INTEGRATED BEHAVIOR THERAPY FOR EXCLUSIVELY-ANXIOUS SELECTIVE MUTISM: A NONCONCURRENT MULTIPLE BASELINE DESIGN ACROSS FIVE PARTICIPANTS

By

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ABSTRACT

INTEGRATED BEHAVIOR THERAPY FOR EXCLUSIVELY-ANXIOUS SELECTIVE MUTISM: A NONCONCURRENT MULTIPLE BASELINE DESIGN ACROSS FIVE PARTICIPANTS

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Selective mutism (SM) is a rare childhood anxiety disorder, which may be markedly detrimental to a child's academic performance and social functioning if left untreated (American Psychiatric Association [APA], 2013). *Integrated Behavior Therapy for Selective Mutism* (IBTSM; Bergman, 2013) is the only manualized treatment approach developed specifically for children with SM. Previous investigations provide evidence for its efficacy in a clinical research setting (Bergman, Gonzalez, Piacentini, & Keller, 2013), and examined its effectiveness using 8-session (Cotton-Thomas, 2015), 12-session (Siroky, Carlson, & Kotrba, 2017), and 35-session (Khan & Renk, 2018) versions of IBTSM when implemented in real-world conditions (e.g., school setting, community-based clinics). The present study used a nonconcurrent multiple baseline single-case design to examine the adherence, effectiveness, and acceptability of a condensed version of IBTSM (i.e., 16 sessions in a community-based clinic) in decreasing social anxiety levels and speech avoidance for five children (ages four to eight years) diagnosed with SM, exclusively-anxious subtype. Novice clinicians were able to implement the condensed (16-session) version of IBTSM with excellent adherence (i.e., average = 97%) for all five participants over an average of 19 weeks (*Range* = 16-22 weeks). Visual analyses did not demonstrate a replicated intervention effect across all five children. However, Tau-U effect size indices and Reliable Change Index (RCI) calculations demonstrated significant individual improvements in social anxiety levels and speaking behaviors over the course of treatment.

Notably, three of five children (60%) no longer met diagnostic criteria for SM at the end of treatment. Surprisingly, two children who presented with comorbid anxiety disorders (generalized anxiety, separation anxiety) saw a removal of these diagnoses by the end of treatment as well. All five caregivers rated the condensed version of IBTSM as an acceptable treatment approach overall for their child's mental health challenges, with specific endorsements of acceptability in the areas of time required and treatment quality. A majority of caregivers also rated IBTSM as effective in addressing their child's needs. Future research directions and study implications are provided.

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CHAPTER 1

INTRODUCTION

Selective Mutism (SM) is a rare behavioral disorder in which children persistently fail to speak in certain settings, though they exhibit typical speech in others. Children with SM may speak effortlessly in comfortable contexts (e.g., at home, with caregivers, close friends), but later withhold speech when prompted to respond verbally in other situations (e.g., at school, in public). This lack of speech must be evident for at least one month and cannot be better explained by developmental delays, speech and language impairments, or poor knowledge of the spoken language within these social settings (American Psychiatric Association [APA], 2013). Despite its low prevalence rate, with less than two percent of children receiving a diagnosis of SM (Bergman, Piacentini, & McCracken, 2002; Elizur & Perednik, 2003), the potentially debilitating effects of SM on later development demand a need for increased awareness to promote the early identification of SM and to disseminate potential evidence-based treatment (EBT) options for children with this disorder.

Scholars agree that SM is likely caused by excessive anxiety when faced with an expectation to speak in certain situations (Cohan, Chavira, & Stein, 2006; Muris, Hendriks, & Bot, 2016). These conclusions are due to consistent findings that SM and social anxiety are closely linked with regard to both etiological theories and approaches for intervention. Two recent comprehensive reviews draw from the SM literature to support a developmental psychopathology model, which integrates biological, psychological, and social factors that may explain why children with SM experience feelings of physiological and/or psychological discomfort in some social situations (Muris & Ollendick, 2015; Viana, Beidel, & Rabian, 2009). Theories discussing potential temperamental, genetic, and environmental causes also reiterate

common characteristics between SM and social anxiety within these domains. High rates of comorbid social anxiety in samples of youth with SM add to this theory suggesting that SM may be a more severe form of social anxiety (Ford, Sladeczek, Carlson, & Kratochwill, 1998; Gensthaler et al., 2016; Viana et al., 2009). The most telling result of these aggregated findings can be seen in the recent classification of SM as an anxiety disorder in the fifth edition of the Diagnostic and Statistical Manual for Mental Disorders (DSM-5; APA, 2013).

Although not all cases with SM present with comorbid social anxiety, the association between SM and social anxiety is often used to help conceptualize why SM symptoms (i.e., speech avoidance) persist, later informing the course of treatment for children with SM (Muris & Ollendick, 2015). While a variety of factors may initially lead to heightened anxiety in situations where speaking is expected, the continual reinforcement of withheld speech may be best explained via a behavioral conceptualization. Persistent avoidance of social interactions is a key characteristic of social anxiety and is viewed as an attempt to reduce anxious feelings and thoughts (APA, 2013). Similarly, speech avoidance during anxiety-provoking situations naturally reinforces the likelihood that a child with SM will avoid speech again in similar situations. Kotrba (2015) and Bergman (2013) both discuss a "cycle of avoidance" where every instance of avoidance removes aversive sensations associated with speaking, and the removal of the expectation leads to relief. This maladaptive pattern becomes negatively reinforced with each paired association of avoidance, removal of speaking expectations, and resulting feelings of relief. This conceptualization of SM would also view an effective treatment for SM as one that disrupts the cycle of avoidance through gradual exposure to feared stimuli, or situations with opportunities to practice, rather than avoid, speech in a supportive environment.

Psychosocial treatment approaches (e.g., cognitive-behavioral therapy, behavioral therapy) are currently supported as the most effective option for both social anxiety and SM (Cohan et al., 2006; Viana et al., 2009), though there is a clear lack of options for evidence-based treatments meant specifically for children with SM (Chorpita et al., 2011). Given the young age of onset, as well as the behavioral conceptualization of speech avoidance, behavioral therapy may be most appropriate for this young population. Behavioral therapy for SM typically involves common behavioral techniques such as graduated exposure and a contingency management plan to continually reward the successful completion of exposure tasks (Bergman, Gonzalez, Piacentini, & Keller, 2013; Kotrba, 2015; Muris & Ollendick, 2015). Other strategies include the use of shaping to increase expectations for different types of speech, fading new people into comfortable contexts (i.e., stimulus fading), and generalization across settings and people (Zakszeski & DuPaul, 2016). Unfortunately, the rarity of SM makes it unlikely that clinicians will encounter cases of SM and, in turn, they may not know how to effectively diagnose or treat it when cases do arise. For these reasons, there is a need for a standardized approach to behavioral therapy for SM that can be easily disseminated into community-based clinical settings to treat children with SM.

Bergman (2013) sought to address this noticeable gap in the current state of evidence-based practice for SM by developing *Integrated Behavior Therapy for Selective Mutism* (IBTSM). IBTSM utilizes behavioral techniques in a standardized approach via a manualized treatment program for children ages four to eight years diagnosed with SM. One benefit of a manualized approach is the potential ease of implementation in new contexts (Kendall, Gosch, Furr, & Sood, 2008). IBTSM may be particularly useful for clinicians who serve children with SM since behavioral strategies (e.g., fear hierarchy, graduated exposure paired with positive

reinforcement, contingency management) are typically effective for encouraging speech and are appropriate for the developmental level of this young population. Manuals may also help to ensure treatments are carried out as intended, which, in turn, may promote greater treatment adherence and acceptability (Perepletchikova & Kazdin, 2005). Additional considerations for the use of a manualized approach to behavioral therapy include the need for standardized treatments to inform evidence-based practice (American Psychological Association [APA], 2006), and the potential for manuals to contain resources to facilitate generalization of gains across contexts (Addis, Wade, & Hatgis, 1999).

Preliminary evidence suggests IBTSM is a potentially efficacious treatment for children with SM (Bergman et al., 2013). Bergman and colleagues (2013) used a randomized controlled trial (RCT) to demonstrate the efficacy of IBTSM for 21 children with SM. By the end of treatment (20 sessions across 24 weeks), the IBTSM group made significant improvements in speech and social anxiety, with some children demonstrating significant growth after only 12 weeks (i.e., at the midpoint assessment). Specifically, 25% (n = 3) of children who received IBTSM no longer met criteria for SM at the midpoint assessment and two-thirds (67%, n = 8) saw a removal of diagnosis by the end of 20 sessions and 24 weeks of treatment. While these results support the possible efficacy of IBTSM, Bergman and colleagues (2013) recommended future studies examine the effectiveness of IBSTM in community-based settings, with consideration to whether a modified treatment length could produce comparable gains.

Three studies, including one unpublished manuscript, have attempted to explore the effectiveness of IBTSM with modifications to treatment length and/or the setting in which IBTSM is implemented. The first study condensed IBTSM to 12 sessions over 18 weeks, though all treatment components were kept the same (Siroky et al., 2017). In this single-case

investigation involving a replicated AB design, an independent clinician implemented IBTSM with high fidelity (96.7%). Two four-year-old boys participated, each diagnosed with SM, though they presented with very different characteristics and associated symptoms. For instance, one child had comorbid social anxiety disorder and withheld speech primarily in the school setting. The second child, however, showed oppositional behaviors (e.g., ignoring parent commands) in addition to his lack of speech at home, in school, and in public. Despite these varying presentations, both participants showed increases in speaking behaviors and significant reductions in caregiver-reported social anxiety levels by the end of treatment. The child with the primarily anxious presentation at baseline no longer met criteria for SM by the three-month follow-up. Additionally, caregivers reported high levels of overall satisfaction with the condensed version of IBTSM (i.e., 12 sessions over 18 weeks) when implemented in a community-based clinical setting.

An unpublished investigation by Cotton-Thomas (2015) implemented a brief version of IBTSM (i.e., 8 sessions across 8 weeks) in a school setting. Effect sizes for direct observations of speech suggested a meaningful increase in non-vocal speech in the school setting after 8 weeks of treatment, though minimal gains were seen in vocal speech. All three children saw a significant improvement in caregiver-reported functional communication at the end of treatment as well, though teacher ratings did not result in a replicated effect. Caregivers and school professionals both viewed the 8-session, 8-week IBTSM as an acceptable treatment option for students with SM. The author of this study did not, however, assess or report changes in clinical diagnoses for the three participants between baseline and the end-of-treatment time points, making it difficult to determine whether treatment outcomes were clinically meaningful.

Finally, Khan and Renk (2018) used a 35-session version of IBTSM to treat a five-year-old boy with SM and comorbid speech/language deficits. He also presented with clinically elevated symptoms of separation anxiety. Given this child's age, symptoms, and impairment at baseline, IBTSM was selected as the most appropriate treatment option. He received 35 total sessions of IBTSM, with five follow-up sessions. Treatment was extended due to a need to move rather slowly through the child's fear hierarchy and to allow for continued services while the child underwent a speech/language evaluation. However, as the child became more verbal with his therapist, his speech deficit served as a noticeable barrier for progress and his need for more targeted speech therapy became more evident. Thus, treatment transitioned from IBTSM to speech therapy. Although this child continued to demonstrate limited verbal communication even with familiar adults, parent ratings by the end of treatment indicated that his symptoms of anxiety, depression, and withdrawal all fell within normal limits for same-age peers. Clinical diagnoses were not reassessed at the end of treatment.

These early investigations provide some indication of the potential transportability and effectiveness of IBTSM in applied settings, though there is clear evidence to suggest that 8- and 12-session versions of IBTSM, across 8 to 18 weeks, are insufficient to yield the type of significant, replicable improvements in SM symptomology expected of an EBT. The mixed results from previous research on IBTSM suggest a need to better understand for whom and under which circumstances this treatment approach is consistently effective (i.e., external validity). Reviews of the SM treatment literature agree that behavioral therapy is generally effective for samples of children with SM (Muris & Ollendick, 2015; Pionek Stone, Kratochwill, Sladeczek, & Serlin, 2002), but Cohan and colleagues (2008) argue that differing presentations of SM may benefit from more targeted treatment approaches. Scholars of SM are continuing to

explore potential subtyping classification systems for SM, given that many children with SM may also present with mild oppositional tendencies or have a comorbid speech/language disorder (APA, 2013; Muris & Ollendick, 2015). Four studies have recently attempted to provide evidence for SM subtypes, though each resulted in a different set of potential classifications (Cohan et al., 2008; Darr et al., 2016; Diliberto & Kearney, 2016; Mulligan et al., 2015). Cohan and colleagues (2008), as well as the unpublished manuscript by Darr et al. (2016) were consistent with their findings for at least three subtypes of SM, distinguishing between those who are exclusively-anxious, those with mild oppositional behaviors and anxiety, and those with a speech/language disorder or communication delay and anxiety as well.

This confirmation of potential SM subtypes leads to further questions on whether distinct treatment approaches are needed to best address the needs of children with SM with varied clinical presentations. Cohan and colleagues (2008) argue for such a treatment planning process. They suggest behavioral therapy alone may be ideal for children who present with the proposed subtype of exclusively-anxious SM, while children with mild-oppositional/anxious tendencies might benefit from an approach that includes a more structured contingency management plan to reinforce compliance. Children with a comorbid communication-delay would likely need supplemental speech therapy along with behavioral therapy. This call for subtype-informed treatment aligns with recent trends in clinical practice that seek to distill specific components of EBTs and analyze their effectiveness to match client characteristics (Chorpita, Daleiden, & Weisz, 2005). However, the absence of any EBT designed specifically for children with SM (Chorpita et al., 2011) makes it difficult for clinicians who are less familiar with this disorder to use such a distillation and matching process to address individual needs. In turn, Comer and

Barlow (2014) note that manualized interventions, rather than modular approaches with less structure, may be particularly useful for low-incidence disorders like SM.

With a clear lack of research on SM interventions, scholars must first determine which interventions and/or strategies work for which specific groups of individuals with SM (i.e., age ranges, subtypes of SM) and the conditions necessary to promote change (e.g., clinic or school context, length and dosage of treatment) before a complex model can inform decisions about a modular approach to treatment. A manualized treatment such as IBTSM may be ideal for this purpose as it will be important to identify a structured, evidence-based approach that can be replicated with close adherence to the manual when implemented in new contexts among a range of clinicians and with clearly-specified populations (i.e., subtypes) of youth with SM.

In sum, initial support for IBTSM as a potential EBT for children with SM is encouraging, yet incomplete. Currently, one randomized controlled efficacy trial, two single-case designs, and one clinical case study have been conducted to specifically explore the utility of IBTSM. Taken together, this work points to the potential efficacy of IBTSM and serves as informative evidence of which contextual manipulations (e.g., length of treatment, community-based clinic vs. school setting) may produce replicable treatment effects. Early attempts to modify treatment length echo the call from treatment developers (Bergman et al., 2013) who suggest a shorter approach may still yield meaningful improvements in SM symptomology, though studies exploring a 12-session, 18-week clinic-based version of IBTSM (Siroky et al., 2017) and an 8-session, 8-week school-based version IBTSM (Cotton-Thomas, 2015), were limited by insufficient baseline data points, making it difficult draw conclusions about casual effects following the introduction of IBTSTM as an intervention. Inconclusive results regarding IBTSM's effectiveness may also reflect varied effects for children with distinct clinical

presentations of SM, as was suggested by Siroky and colleagues (2017), though these hypotheses have yet to be empirically tested. Thus, in order to expand upon these initial findings, future studies should carefully examine the effectiveness of IBTSM using a feasible and sufficient treatment dosage (e.g., 16-session version) for children with the proposed exclusively-anxious SM subtype in a community-based clinical setting.

Answering such questions will require close adherence to research design standards, ensuring that scholars can draw valid conclusions about treatment effects. While RCTs are typically the gold standard for validating potential EBTs (APA, 2006), exploring IBTSM's effectiveness using single-case experimental designs may be ideal given the anticipated small population of youth with the hypothesized subtype of exclusively-anxious SM. Single-case research may address practical limitations of RCTs as well, particularly when studying low-prevalence disorders. Simply recruiting a large enough sample to draw valid conclusions may require a significant amount of time, funding, and resources (Kratochwill et al., 2012).

The APA Presidential Task Force on Evidence-based Practice (2006) notes that single-case methodologies are especially valuable in examining functional relationships between an intervention and the observed treatment outcomes within the context of individual clients. Recent guidance about single-case research design standards (e.g., call for interobserver agreement for dependent variables, minimum number of attempts needed to demonstrate a replicated effect, minimum number of data points per phase) has also helped to ensure greater confidence with which scholars can make causal inferences about intervention effectiveness (Kratochwill et al., 2012). Investigations following these standards allow scholars to examine replicated treatment effects for participants who serve as their own control (Christ, 2007; Kratochwill & Levin, 2010). Finally, sound single-case investigations of manualized treatments

such as IBTSM can be used within future meta-analyses to inform evidence-based practice when treatment effect sizes for individual subjects and/or for a group of subjects are presented (Horner, Carr, Halle, McGee, Odom, & Wolery, 2005; Maggin & Chafouleas, 2012).

Current Study

Considering the need for standardized SM treatment options to inform evidence-based practice with this population, and the limitations of the current SM treatment literature, the present study critically examined the treatment adherence, effectiveness, and acceptability of a condensed version of IBTSM (i.e., 16 sessions over an average of 19 weeks) administered in a community-based clinical setting to five children ages four to eight with exclusively-anxious SM. A 16-session version was selected due to the treatment developers' note that significant effects may be seen in a shorter amount of time (i.e., less than 20 sessions over 24 weeks), though the lack of replicated effects in studies examining 8-session and 12-session versions of IBTSM indicate a potential need for increased treatment length. Additionally, 16 sessions is a common length for other manualized psychosocial treatment approaches for children with anxiety (e.g., *Coping Cat*). The session structure and key components of IBTSM (e.g., in- and out-of-clinic exposures guided by a fear hierarchy, contingency management), as outlined in the treatment manual (Bergman, 2013), remained the same. This investigation serves as an essential step in the process of verifying IBTSM as a potential EBT meant explicitly for children with exclusively-anxious SM.

In order to draw such conclusions about the overall utility of IBTSM, the present study measured treatment adherence, effectiveness, and caregiver-rated acceptability across all five children with an exclusively-anxious presentation of SM. Clinician self-report ratings and direct observations of treatment sessions were used to assess overall treatment adherence according to

pre-developed session checklists derived from the IBTSM manual (Bergman, 2013). Visual analyses and effect size calculations were used to test hypotheses related to the effectiveness of IBTSM in producing replicated treatment effects for these five children with exclusively-anxious SM. Finally, acceptability was measured by caregiver ratings at the end of treatment.

CHAPTER 2

LITERATURE REVIEW

A careful review of the literature on SM and its treatment informed four research questions, hypotheses, and the overall design of this study. The following topics are covered in greater detail in the section below: (a) diagnostic criteria, associated features, prognosis, and potential subtypes of SM, (b) etiological theories and specific conceptualizations of SM as it relates to social anxiety, (c) the current evidence on effective treatment options for SM, and (d) methodological approaches for examining effective interventions for low-prevalence disorders, specifically the use of single-case research.

Selective Mutism

Diagnostic criteria. Selective mutism (SM) is a rare childhood behavioral disorder, resulting from excessive anxiety in speaking situations (APA, 2013). The primary symptom of SM is described as a persistent lack of speech in social situations where speaking is expected, though typical and spontaneous speech may be observed in other settings. Mutism (i.e., speech avoidance) is "selective" due to the inconsistency of speech across multiple contexts (e.g., at school, in public, with unfamiliar people). For example, a child may communicate without trouble while at home with family or in comfortable situations but may fail to speak when prompted in public or at school. SM symptoms must substantially interfere with the individual's social, academic, or occupational functioning. The fifth edition of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5; APA, 2013) notes a number of exclusionary criteria to consider before a diagnosing SM (see Table 1 on next page). For instance, it is common for children to be exceptionally shy or inhibited at the beginning of a major transition (e.g., attending a new school, family moving to a new community). As such, the child's lack of speech must

occur for at least one month, which may not be limited to the first month of school. The child's lack of speech cannot be better attributed to other impairments such as a hearing impairment, or developmental, learning, or psychotic disorders. Finally, SM is not typically diagnosed in children whose families have recently immigrated to a new country or who speak English as a second language. A clinical diagnosis should be informed by an extensive review of the child's familial, developmental, and social-emotional history to rule out other contributing factors that may better explain the child's persistent pattern of withheld speech.

Table 1.

Diagnostic Criteria for Selective Mutism in the DSM-5

A.	Consistent failure to speak in specific social situations in which there is an expectation for speaking (e.g., at school) despite speaking in other situations.
B.	The disturbance interferes with educational or occupational achievement or with social communication.
C.	The duration of the disturbance is at least 1 month (not limited to the first month of school).
D.	The failure to speak is not attributable to a lack of knowledge of, or comfort with, the spoken language required in the social situation.
E.	The disturbance is not better explained by a communication disorder (e.g., childhood-onset fluency disorder) and does not occur exclusively during the course of autism spectrum disorder, schizophrenia, or another psychotic disorder.

(Adapted from APA, 2013; p. 127)

Associated characteristics. Children with SM may also present with a wide variety of associated behaviors in addition to key diagnostic markers. Many children with SM tend to feel heightened fear in social settings and increased social withdrawal (APA, 2013; Bergman, Piacentini, & McCracken, 2002; Steinhausen & Juzi, 1996). Carbone and colleagues (2010) also report diminished social skills in a sample of young children with SM. In their study comparing child-, parent-, and teacher-reported social skills across groups of children with SM, children

with other anxiety disorders, and a control group, teachers rated children with SM as demonstrating significantly lower levels of social assertion, self-control, and overall social skills. Marked impairment in social, academic, or occupational performance is a main criterion for SM diagnosis, but the repeated avoidance of speech in common settings (e.g., at school, with peers) may cause untreated symptoms to become increasingly worse with time (Ford et al., 1998).

Children with SM have been shown to underperform academically when compared to same-age peers without SM. The accuracy of certain academic performance tasks is important to consider, however, as certain assessments may be invalid for these children if speaking is required (Steinhausen & Juzi, 1996). Other evidence shows children with SM as having average intelligence when verbal demands are removed (Remschmidt et al., 2001). In addition to potential social and academic performance deficits, studies report anywhere from 4%-42% of children diagnosed with SM also have comorbid speech and language impairments (Manassis et al., 2007; Mulligan et al., 2015; Steinhausen, Wachter, Laimböck, & Winkler Metzke, 2006). These findings together suggest that children with SM may have adequate academic and cognitive abilities, but the verbal demands of certain academic assessments make it difficult to properly evaluate students' skills (Bergman et al., 2002).

Another common feature of SM includes mild oppositional or seemingly defiant behaviors (APA, 2013). Caregivers of children with SM have reported more frequent temper tantrums, noncompliance, or argumentativeness, in conjunction with the defining symptom of a persistent lack of speech (Ford et al., 1998). These behavioral presentations often lead to a misconception about SM and the child's intent to withhold speech as an act of defiance. However, scholars argue that a lack of speech may actually reflect a child's unique reaction to intense anxiety caused by the expectation to speak, rather than willful oppositionality (Dummit et

al., 1997; Ford et al., 1998; Keeton, 2013; Kristensen & Torgersen, 2002). Results from a study by Cunningham, McHolm, and Boyle (2006) also refute this claim. The authors analyzed a variety of parent- and teacher-reported behaviors thought to be associated with SM (e.g., anxiety at school, separation anxiety, depressive symptoms, oppositional behavior, verbal deficits) and found that children with SM were not significantly more likely to display oppositional behaviors compared to a community control group. As a whole, caregivers more frequently report inhibited temperament instead.

Proposed clinical profiles of SM. Recent investigations have attempted to explore and describe the diversity of these associated characteristics, with some suggesting potential subtypes of SM. Subtyping for SM may be useful for practice and research for a number of reasons. First, subtypes allow treating clinicians to better understand and observe child behaviors, as well as caregiver perceptions of these symptoms. Subtypes may also help to select and implement the most appropriate treatment approach, given the potential for a variety of contributing factors to SM symptoms. Cohan and colleagues (2008) were the first to examine potential subtypes in 130 children between 5 and 12 years of age with a primary diagnosis of SM. As a whole, children in this sample reported clinical levels of social anxiety and syntax problems with regard to spoken communication. Using parent ratings of social anxiety, internalizing behaviors, externalizing behaviors, aspects of language and communication, and overall psychosocial impairment, a three-factor model demonstrated the best fit. This model resulted in the following potential subtypes of SM: exclusively-anxious, anxious-communication delayed, and anxious-mildly oppositional. More details about these subtypes, as well as others described in additional studies exploring SM subtype classifications, can be found in Table 2 (page 16).

Table 2.

Summary of Studies Exploring SM Subtyping Systems

Study	*Measures*	*Analyses Used*	*Suggested SM Subtypes*	*Significant Comorbid Symptoms by Subtype*
Cohan, Chavira, Shipon-Blum, Hitchcock, Roesch, & Stein (2008)	ADIS-P, SMQ, SASC-R, ECBI, CCC-2, CAIS-P, CBCL, VABS-II	Latent profile analysis, ANOVA	(1) Anxious-mildly oppositional (2) Anxious-communication delayed (3) Exclusively anxious	(1) Social anxiety, borderline significant oppositional behaviors and syntax deficits (2) Social anxiety, clinically significant syntax deficits, borderline significant speech deficits (3) Social anxiety only
Mulligan, Hale, & Shipon-Blum (2015)	SM-CDQ	Qualitative analysis and coding, cluster analysis, ANOVA	(1) Global (2) Anxiety/Language (3) Low Functioning Mutism (4) Sensory/Pathology Mutism (5) Emotional/Behavioral	(1) Higher overall academic, social-emotional, and behavioral functioning, no speech deficits (2) Comorbid anxiety disorders, comorbid speech/language impairments (3) Academic impairment, sensory issues, poor executive functioning, emotional lability (4) Severe sensory issues, delayed motor skills, separation anxiety symptoms (5) Oppositional behaviors, emotional lability, executive functioning concerns
Diliberto & Kearney (2016)	ADIS-P, CBCL	Descriptive analyses, exploratory factor analysis, confirmatory factor analysis	(1) Anxious Behavior Profile (2) Oppositional Behavior Profile	(1) Social problems and social anxiety symptoms, aggressive behaviors (2) Aggressive behaviors, oppositional/defiant behaviors, social problems
Darr, Kotrba, & Carlson (2016)	Diagnostic intake interview and developmental history, SMQ, BCI, BASC-2	Cluster analysis, ANOVA	(1) Exclusively-anxious (2) Communication delayed/bilingual-anxious (3) Mildly oppositional/ sensitive-anxious	(1) Anxious symptoms, minimal defiance (2) Speech delays or language disorders, spoke more than one language, comorbid anxiety (3) Parent-reported oppositional and defiant behaviors, comorbid anxiety

ADIS-P = Anxiety Disorders Interview Schedule for DSM-IV-Parent Version, SMQ = Selective Mutism Questionnaire, SASC-R = Social Anxiety Scale for Children-Revised, ECBI = Eyberg Child Behavior Inventory, CCC-2 = Children's Communication Checklist, CAIS = Child Anxiety Impact Scale–Parent Version, CBCL = Child Behavior Checklist, VABS-II = Vineland-II Adaptive Behavior Scales– Parent/Caregiver Rating Form, SM-CDQ = Selective Mutism Comprehensive Diagnostic Questionnaire, BCI = Behavioral Concerns Inventory, BASC-2 = Behavior Assessment System for Children, Second Edition

Similarly, Mulligan and colleagues (2015) explored different subtypes of SM, noting the need for treatment options that fit with each child's presentation of SM diagnosis and comorbid symptomology. This study used the Selective Mutism Comprehensive Diagnostic Questionnaire (SM-CDQ; Shipon-Blum, 2003) to assess 186 children with a primary SM diagnosis. The SM-CDQ asks about the child's developmental and medical history, potential speech/language impairments, and their behaviors at home, school, and other social settings. The SM-CDQ also includes a brief rating scale at the end of the form, known as the Mutism Behavior Rating Scale (MBRS), which consists of eight subscales used to categorize the level of impairment within the categories of: Academic, Esteem, Flexibility, Sensory, Anxiety, Executive, Oppositional, and Labile. Parent-reported scores on the MBRS subscales were analyzed via cluster analysis, yielding a five-factor model as the best fit for this sample. Five subtypes were derived from this model: Global Mutism, Emotional/Behavioral Mutism, Anxiety/Language Mutism, Low Functioning Mutism, and Sensory Pathology Mutism (Mulligan et al., 2015).

In another recent study, Diliberto and Kearney (2016) further explored different profiles of children with SM. Their analysis included parent reports of Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2001) social problems and aggressive behaviors for a group of 57 children with SM. The confirmatory factor analysis yielded a significant two-factor model, which differentiated between two groups: one presenting with primarily anxious behaviors and the other with primarily oppositional or defiant behaviors. These factors were generally negatively associated, with profiles of anxious behaviors inversely related to aggressive behaviors. In addition, children with an exclusively-anxious profile showed increased aggressive, but not oppositional or defiant, behaviors. It should be noted, however, that these

authors did not attempt to examine a subtype of SM representing children with co-occurring communication delays, as others have done (Cohan et al., 2008; Mulligan et al., 2015).

To address previous gaps in this particular line of research, Darr and colleagues (2016) continued with this exploration of potential subtypes through an analysis of 120 pre-existing clinical records of SM cases from a community-based clinic that specializes in the treatment of SM. Data in this study consisted of parent reports of developmental history, medical history, parent psychopathology, as well as a Behavior Concerns Checklist (BCI). A portion of the sample had available data using the Selective Mutism Questionnaire (SMQ; Bergman, Keller, Piacentini, & Bergman, 2008) and the Behavior Assessment System for Children, Second Edition (BASC-2; Reynolds & Kamphaus, 2004). A cluster analysis based on these assessments demonstrated a three-factor model to include an exclusively-anxious SM group, a communication-delayed/bilingual-anxious SM subgroup, and a mildly-oppositional/sensitive-anxious SM subgroup. While standardized assessment tools (e.g., SMQ, BASC-2) were predictive of some variables includes in the cluster analysis, parent reports via diagnostic intake interviews and the BCI tended to have higher predictor importance. Of the 120 youth in this sample, 47% reported having some comorbid anxiety disorder at the intake assessment. This rate of prevalence, along with the lack of any significant difference in anxiety symptoms between groups, these investigators included the "anxious" label within each of the three classifications to denote the persistent presence of anxious symptomology among all of the SM cases, regardless of varied additional characteristics (e.g., oppositional behaviors, bilingualism, speech delays).

Each of these four studies highlights distinct behavioral patterns of youth with SM in an attempt to verify subtype classification systems. These early findings are relevant for informing future research on SM with the end goal of improving the effectiveness of SM treatment

approaches. Results from these subtyping analyses yielded anywhere from two to four different subgroups within samples of youth with a primary diagnosis of SM, though assessment procedures varied across all studies. The current state of the literature on SM subtypes indicates a need for greater clarification and support for these tentative classifications as well as some standardized procedure to determine subtype in samples of diverse youth with SM. With such a system in place for SM, clinicians may be able to more accurately target problematic behavior patterns and match each client with an appropriate treatment plan to promote optimal effectiveness and acceptability.

Prevalence and prognosis. Despite these varied presentations and associated features, SM is a relatively rare disorder among school-age children. The most recent prevalence studies suggest less than two percent of children in receive a diagnosis of SM (Bergman et al., 2002; Elizur & Perednik, 2003). This rate is quite low when compared to other childhood anxiety disorders such as separation anxiety disorder (4%) and social anxiety disorder (5%) but is comparable to well-studied disorders such as Autism Spectrum Disorder (ASD) which reports a prevalence rate of just under 2% (APA, 2013). Diagnosis of SM is slightly more common in females than males, with an average female to male ratio of about 2:1 (Viana et al., 2009).

SM is typically considered a childhood disorder. The average age of onset for SM is before age five, though adolescent-onset SM has been reported as well (Ford et al., 1998; Sutton, 2013). Ford and colleagues (1998) report a delay of 7.5 months to 29.7 months, on average, between initial symptoms and referral for treatment. The delay in identification and proper diagnosis may be more pronounced for adolescents with SM, attesting to the need for early identification efforts to ensure children with SM receive timely and effective treatment. Children with SM are often first identified after the transition into regular schooling (Viana et al., 2009).

Descriptive studies report school as the setting where speech avoidance occurs most frequently and the context in which anxiety is most severe (Black & Uhde, 1995; Dummit et al., 1997). As described above, the debilitating effect of anxiety and avoidant behaviors (i.e., failure to speak) in the school setting can serve as a significant hindrance to a child's academic learning and overall social-emotional development.

Within the last decade, scholars of SM have begun to explore how symptoms during childhood can affect well-being later in life. Adults who report pervasive childhood SM symptoms (i.e., occurring across multiple settings) were more likely to report later phobic disorders, and were less likely to show improvement even years after treatment (Steinhausen et al., 2006; Sutton, 2013). These outcomes were significantly more pronounced when compared to other adults who reported less pervasive SM symptoms during childhood. Walker and Tobbell (2015) recently conducted a study involving four adults with SM to elicit their subjective definitions and interpretations of the disorder. All participants were diagnosed with SM during their childhood or early teenage years. Overall, qualitative interviews revealed that, "adults with SM may be characterized by a profound sense of loss, of one's identity, of one's past and future, and of one's social interactions" (p. 468). Although scarcely presented in the literature, cases with enduring SM show greater symptom severity with age and greater interference with social or occupational functioning due to the salience of repeated avoidance patterns over time (Pionek Stone et al., 2002; Sutton, 2013). With age of onset occurring at a particularly crucial time in development, careful evaluation, including differential diagnosis and assessment for comorbid disorders, will be essential for children referred for SM treatment as these unique characteristics may inform the course of treatment.

SM and social anxiety. The DSM-5 (APA, 2013) reclassified SM, moving it out of the "disorders experienced in childhood" to its current listing under anxiety disorders. This revision was likely due to the growing evidence suggesting children with SM fail to speak due to overwhelming anxiety associated with speech or verbal social interactions. For example, Ford and colleagues (1998) extensively reviewed phenomenological characteristics of individuals with SM. Their results highlighted behavioral similarities between SM and social anxiety including more frequent internalizing behaviors, as compared to externalizing behaviors, and observable variation in the length, volume, and spontaneity of speech in social settings. Due to these close similarities, children with SM are commonly diagnosed with comorbid social anxiety (Black & Uhde, 1995). Viana and colleagues (2009) outlined a number of published studies reporting comorbidity rates of 60% or greater in samples of children with SM (Kristensen, 2000; Manassis et al., 2007). Further, Yeganeh and colleagues (2003) explored clinical distinctions between SM and social anxiety. Though all children in the SM group (n = 23) also met criteria for comorbid social phobia, clinician ratings of social anxiety levels were higher for children with SM, as compared to children with social phobia alone. Interestingly, these distinctions were not significant when looking at child self-reports of social anxiety. Similar findings were seen in a second study by the same researchers (Yeganeh, Beidel, & Turner, 2006).

Etiological link between SM and social anxiety. Considering the heterogeneity of SM and its associated characteristics**,** there is limited evidence pointing to a single etiological theory for this disorder. Historically, SM was viewed as a response to childhood trauma or other adverse life events, though years of aggregated research concluded that trauma is not significantly associated with the onset of SM symptoms (Ford et al., 1998; Kotrba, 2015). More recent literature suggests SM is more closely associated with social anxiety, which results from a

combination of genetic, temperamental, environmental, and neurodevelopmental factors (Muris & Ollendick, 2015).

Past research has demonstrated high heritability of anxiety disorders, including SM. Kristensen and Torgersen (2002) found that mothers of children with SM were more likely to report being shy or having social anxiety (38.9%) compared to control families (i.e., with a child with SM; 3.7%). This trend was seen in fathers of the sample as well (31.5% vs. 0.9% in control families). Chavira, Shipon-Blum, Hitchcock, Cohan, and Stein (2007) also support previous literature on some familial association of anxiety and tendencies to withhold speech. In a sample of 70 children with SM, 37% of caregivers reported social phobia and 18% reported avoidant personality disorder. These percentages were significantly higher than control families for both disorders (14% and 5%, respectively). Clearer genetic links are beginning to emerge in the SM literature. For instance, Stein and colleagues (2011) studied one particular variation of the CNTNAP2 gene in 99 families who reported having at least one child with SM. This variation was associated with language impairments or socially anxious traits, including diagnoses of social anxiety. In this same paper, the authors assessed a larger sample (N = 1028) of young adults and found similar, significant results. Based on these findings together, there is growing evidence from a genetic perspective that SM is closely associated with some form of underlying general or social anxiety.

A child's temperament may serve as another contributing factor in the manifestation of SM symptoms. Drawing from the larger research on childhood anxiety disorders, behavioral inhibition has been shown to significantly predict anxiety disorders. Individuals with an inhibited temperament tend to fear new situations or stimuli and have difficulty with major transitions like switching schools or moving to a different neighborhood (Ford et al., 1998). Few

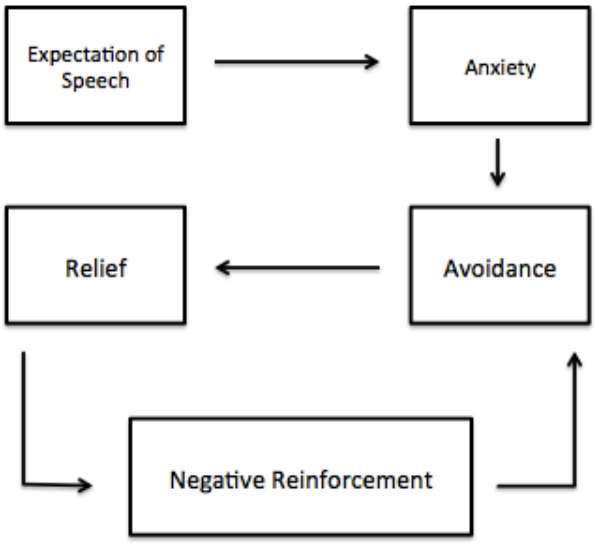
studies explicitly examine the association between behavioral inhibition and SM, though a recent study by Muris, Hendriks, and Bot (2016) may be the first in a much-needed line of research on the behavioral basis of SM. Their results showed significant positive correlations between behavioral inhibition and SM symptom severity, as well as social anxiety in general. Children with SM and increased levels of behavioral inhibition tended to speak fewer words during speech tasks. However, regression analyses indicate that the relationship between behavioral inhibition and parent-reported SM symptoms may not be direct, with social anxiety as a potential mediator. Thus, behavioral temperament may be related to anxiety disorders in a more general sense, rather than a specific disorder like SM.

A common misconception regarding SM is the notion that symptoms are directly caused by a major traumatic event. While some children suffering from post-traumatic stress disorder (PTSD) may withhold speech for a significant period of time following a traumatic experience, there is little evidence to suggest that initial symptoms of SM consistently co-occur with a traumatic event (Black & Uhde, 1995; Gensthaler et al., 2016; Muris & Ollendick, 2015). With that being said, certain environmental factors may contribute to the onset or increased severity of SM. Early research suggests that SM may be more common in children with divorced parents (Hayden, 1980) or parents experiencing marital conflict (Elizur & Perednik, 2003). Parental social isolation or general social deficits may also provide youth with inadequate modeling for socialization (Viana et al., 2009). Further research is needed to clarify the role of parental and familial factors, as a recent study suggests that parenting strategies, behaviors, and attitudes are reportedly similar for children with SM and those without (Alyanak et al., 2013).

To capture the heterogeneity of SM, Muris and Ollendick (2015) propose a *developmental psychopathology model* incorporating biological, psychosocial, and genetic

factors to explain the complexities related to how and why SM symptoms initially occur. Muris and Ollendick (2015) argue, "SM does not develop as the result of one deterministic variable, but rather is due to complex interactions among various vulnerability factors, which heighten the probability (risk) that this psychiatric condition will occur" (p. 161). This model reflects the growing understanding that psychopathology may be best explained by a biopsychosocial framework, which in turn informs integrative or multimodal forms of treatment for SM (Bergman et al., 2013; Oerbeck, Johansen, Lundahl, & Kristensen, 2012); Oerbeck et al., 2014). Such a statement, along with the growing evidence of high comorbidity with social anxiety, suggests that SM should be conceptualized as a complex and multifaceted disorder, with similar underlying mechanisms as other anxiety disorders.

Behavioral conceptualization of SM. A developmental psychopathology model may be helpful to assess an individual's level of risk for SM diagnosis, but a behavioral conceptualization may help to highlight the mechanisms for why SM symptoms persist, and even



Adapted from *Treatment for Children with Selective Mutism: An Integrative Behavioral Approach* by R. L. Bergman, 2013, p. 9. Copyright 2013 by Oxford University Press.

Figure 1. Behavioral conceptualization of selective mutism.

worsen, over time (APA, 2013; Muris & Ollendick, 2015; Viana et al., 2009). Bergman (2013) and Kotrba (2015), explicitly describe a behavioral conceptualization of SM to explain why children persistently avoid speech (see Figure 1).

When faced with the expectation to speak in certain settings, children with SM feel physiological symptoms of anxiety (e.g., increased heart rate, sweating, shortness of breath, shaking hands). These sensations cause significant discomfort and may lead to negative thoughts or strong emotions as well. In an attempt to reduce their anxious feelings, the child may choose to avoid speech if given the opportunity. The removal of the expectation to speak is effective, but the immediate relief negatively and naturally reinforces their avoidant behavior. An association is made that avoidance leads to relief, and the child is more likely to avoid speech the next time they feel anxious when prompted to speak (Bergman, 2013; Kotrba, 2015). Treatment for SM often assumes this behavioral conceptualization to disrupt the "cycle of avoidance," while also keeping in mind the many individual, environmental, and contextual factors that may explain SM onset (Cohan et al., 2006).

Treatment for SM

Psychosocial treatment. Due to the low prevalence of SM and related lack of research, intervention for SM is often informed by evidence-based treatment options for social anxiety (Sharp, Sherman, & Gross, 2007). Psychosocial treatment (e.g., behavioral therapy, cognitive-behavioral therapy, parent-child interaction therapy) is a popular approach to treating social anxiety disorder. These forms of treatment have also been effective in remediating SM symptoms, particularly with children (Muris & Ollendick, 2015; Pionek Stone et al., 2002). Extensive reviews of the treatment literature list behavioral and cognitive-behavioral therapies as

the most common and generally most effective form of treatment for SM (Cohan et al., 2006; Muris & Ollendick, 2015; Viana et al., 2009).

Behavioral therapy. Behavioral therapy may be the ideal form of treatment for children with SM due to the young age of onset and the widely understood conceptualization of SM as an anxiety disorder (Pionek Stone et al., 2002). Treatment for other anxiety disorders (e.g., separation anxiety, specific phobia, social anxiety) typically includes behavioral strategies to encourage individuals to learn about anxious thoughts and feelings (i.e., psychoeducation), with an emphasis on gradual exposure to the feared situation paired with opportunities to practice coping strategies (Vecchio & Kearney, 2009; Zakszeski & DuPaul, 2016). In treating SM, these same behavioral tools may be used to interrupt the cycle of avoidance. Gradual exposure, often paired with rewards for speech, helps to ensure that the child's silence is no longer reinforced by the removal of the expectation to speak (Kotrba, 2015). Behavioral treatment approaches for SM and other anxiety disorders often use a hierarchy of feared situations (i.e., "fear hierarchy" or "fear ladder") to inform symptom severity and to guide gradual exposure exercises.

A final important component of behavioral therapy for SM is generalization from one context to another. Since many children with SM tend to fear very specific situations or settings (e.g., speaking to same-age peers in school, responding to adults when others are watching), stimulus fading may be used to transfer speech from one comfortable person or setting to another less comfortable situation (Bergman, 2013). Severe SM cases may require the use of shaping, which keeps environmental variables constant while slowly increasing expectations for speech (Kotrba, 2015). For example, a child with SM who only speaks to the therapist in a whisper may receive positive reinforcement (e.g., stickers, points for speech) for answering questions from the therapist in a loud, audible voice but later may only receive stickers for longer responses even if

they maintain adequate volume. Positive reinforcement is often powerful in supporting these behavioral strategies, particularly for younger children with SM (Ford et al., 1998). A combination of some or all of these behavioral techniques may help to maximize the effectiveness of behavioral therapy for children with SM.

Cognitive behavioral therapy. Cognitive behavioral therapy (CBT) has recently received more attention in the SM literature. Cognitive components of treatment for SM tend to apply similar behavioral strategies with an additional focus on meta-cognitive techniques to identify anxious feelings, become aware of potential triggers or antecedents, restructuring of distorted expectations associated with speech, and employing relaxation strategies (Fung, Manassis, Kenny, & Fiksenbaum, 2002). One limitation in using this treatment for cases of SM, though, is the notion that CBT may be less effective for younger children. Certain techniques such as identification of one's thoughts and the ability to challenge them through cognitive restructuring are central to CBT and may be above the developmental capabilities of young children who are often diagnosed with SM (Bergman et al., 2013).

Differential treatment for clinical profiles of SM. Given preliminary evidence suggesting various subtypes of SM exist (e.g., exclusively-anxious, mild-oppositional/anxious, communication-delayed/bilingual anxious), it follows that certain treatment approaches may be more or less effective for these proposed subgroups. For instance, Diliberto and Kearney (2016) note that a child presenting with an exclusively-anxious profile may benefit from typical psychosocial treatment approaches (e.g., behavioral therapy, cognitive behavioral therapy) within the school setting or other feared situations, whereas a child whose symptoms are consistent with a communication-delayed/bilingual anxious subtype may require specific therapy targeting both anxious feelings and expressive language difficulties. A purely behavioral

treatment will likely prove to be ineffective. On the other hand, children with mild oppositional tendencies may respond best to a combination of behavioral techniques across settings and parent-driven contingency management.

A recent case study by Skedgell, Fornander, and Kearney (2017) highlights the utility of such a process that involves assessment of comorbid behavioral concerns, clear case conceptualization, and personalized treatment planning. The authors present the case of a six-year-old boy whose symptoms were present for two full years prior to referral to the clinic. During the intake assessment, this participant met criteria for SM and oppositional defiant disorder (ODD). Using standardized measures of speaking behaviors at home and at school, as well as descriptive and observational functional assessments, the clinicians conceptualized his SM as both anxiety- and oppositional-based, with specific issues related to school refusal. These data informed a personalized treatment plan, which consisted of 10 individual sessions emphasizing parent-based contingency management to encourage compliant behavior, followed by 12 sessions of group behavioral therapy with two other children with SM. The group-based therapy plan included psychoeducation, somatic anxiety management techniques, self-modeling, exposure activities, and the use of positive reinforcement throughout. By the end of treatment (i.e., 22 sessions), the participant saw a removal of both SM and ODD diagnoses. His mother also reported successful generalization to the school setting where her son was able to go to school and enter other classrooms to speak to other teachers without difficulty. Improvements were maintained at the six-month follow-up.

Caregiver involvement in treatment. Regardless of subtype, treatment for children with SM typically involves parents or caregivers throughout. The young age of onset is one practical reason for a strong parent component, but the pervasiveness of SM symptoms across settings

also requires a change in caregivers' day-to-day responses (Kotrba, 2015). Caregivers will often need explicit education about the causes, conceptualization, and contributing factors associated with SM. Additionally, caregivers will need to learn how their own behaviors affect their child's SM symptoms, particularly when they attempt to "rescue" their children when they are prompted to speak but fail to do so. For instance, caregivers are quick to recognize their child's feelings of anxiety or fear and, with only good intentions, may try to speak for their child in order to relieve their discomfort. Modeling appropriate responses and behavioral strategies for caregivers is a critical component during behavioral therapy for SM as well (Vecchio & Kearney, 2009). Caregiver involvement may also affect treatment outcomes for youth given the high heritability of anxiety disorders, including SM (Stein et al., 2011), but the level of involvement varies by treatment and its impact is still poorly understood.

School involvement in treatment. School involvement is also highly recommended during treatment of SM given the tendency for SM symptoms to be more severe in this particular setting (Ford et al., 1998; Kotrba, 2015). Currently, few school-based behavioral interventions for SM have been developed and empirically studied (Beare, Torgerson, & Creviston, 2008; Kern, Starosta, Cook, Bambara, & Gresham, 2007; Mitchell & Kratochwill, 2013). These studies typically apply behavioral strategies (e.g., gradual exposure, contingency management) within a school or classroom environment. One notable school-based intervention study by Oerbeck and colleagues (2012) included a sample of seven preschool children (ages 3-5). Investigators described it as a multimodal treatment approach, though behavioral techniques were used a means to elicit speech in children. By the end of treatment, children showed significant gains in caregiver-reported speaking behaviors. Teacher-reported speech also increased significantly, with maintenance one year after treatment completion.

A second study went on to investigate this same intervention with both preschool and school-age children (ages 3-9) via RCT (Oerbeck et al., 2014). Notably, their results showed younger children (ages 3-5) making significantly greater improvement on teacher-reported speech in school compared to older children in the treatment group. These differential treatment gains were seen at a one-year follow-up where 78% of younger children, compared to the 33% of older children, no longer met criteria for SM (Oerbeck, Stein, Pripp, & Kristensen, 2015). Notably, these studies did not attempt to capture changes in social anxiety levels. Although these findings certainly highlight the significant impact of early intervention efforts for children with SM, there is still a need for careful investigations of behavioral interventions implemented across contexts to determine their effect on both speech avoidance and anxiety levels for children with SM.

Manualized treatment approaches. Despite some preliminary progress toward identifying effective strategies for treating SM, as well as contexts in which treatment can and should be carried out, evidence has yet to support one standardized treatment specifically targeting SM. Manualized treatment approaches offer a variety of benefits to clinicians and clients. First, manuals for psychosocial treatment approaches have been found to enhance overall treatment integrity, while also reducing potential variability across cases (Perepletchikova & Kazdin, 2005). For this reason, the presence of a manual is often one criterion used to determine whether a treatment program is evidence-based (Kazdin & Weisz, 2010). The additional benefit of widespread dissemination using treatment protocols helps to ensure clinics have access to reputable resources, strategies, and guidance for evidence-based approaches in treating various disorders, especially those with low incidence rates like SM (Comer & Barlow, 2014).

A number of published manuals for treating anxiety disorders in children and adolescents demonstrate well-established efficacy and effectiveness. For example, *Coping Cat* (Kendall & Hedtke, 2006) is a popular manualized EBT for children with anxiety. *Coping Cat* uses a cognitive-behavioral approach to teach children how to identify their anxious feelings and thought patterns and use adaptive coping strategies. The first half of treatment focuses primarily on learning and conceptualizing these cognitive strategies, while the second half of treatment provides opportunities for youth to practice these skills through gradual exposures. This manualized approach to treating anxiety is widely supported in the literature. Results from both single-case investigations (Kane & Kendall, 1989) and randomized-controlled trials (RCTs; Kendall, 1994; Kendall et al., 1997) demonstrate the consistent effectiveness of *Coping Cat* in successfully relieving anxious symptoms after 16 sessions across 16 weeks of treatment.

The *Coping Cat* manual is meant to be fairly adaptable, allowing practitioners to focus on individual clients' fears or specific symptomology and may be used with children with SM. Husdon, Krain, and Kendall (2001) specifically outline potential adaptations for *Coping Cat* to meet the needs of youth with SM (e.g., emphasizing the importance of rewarding the child's speech). However, there is little research to prove the efficacy of manualized treatments for anxiety, like *Coping Cat*, when used to specifically treat children with SM. Thus, it is unclear whether popular EBTs for broader anxiety disorders are equally effective for youth with SM. Although SM is a relatively rare disorder, clinicians in community-based settings should be prepared to provide adequate care for all potential client needs, including SM. Bergman (2013) developed *Integrated Behavior Therapy for Selective Mutism* to specifically address this SM treatment gap in the anxiety disorder treatment literature.

Integrated Behavior Therapy for Selective Mutism. Integrated Behavior Therapy for Selective Mutism (IBTSM; Bergman, 2013) is the first and only manualized program designed to treat children with SM. IBTSM consists of 20 in-clinic sessions over the course of 24 weeks, with one caregiver-only session at the beginning of the program. IBTSM is intended to treat children with SM, specifically children ages 4-8, given the young age of onset for SM (i.e., before age 5). IBTSM is primarily behavioral in orientation since the developmental capabilities of younger children with SM may limit their ability to effectively understand and apply cognitive strategies. The manual guides clinicians in employing common behavioral strategies (e.g., systematic desensitization, fear "ladder" or hierarchy, contingency management) to increase speech. Together with the caregiver and the child, the clinician uses a Situation Rating Form (Appendix A) to list a number of "easy," "medium," and "hard" situations relative to the child's current level of SM severity. These situations then inform the child's individualized fear ladder (Appendix B), which is used to guide the remainder of treatment. It should be noted, however, that IBTSM does not specifically guide therapists in shaping speech for children with severe forms of SM. Bergman (2013) does provide some suggestions on how clinicians can elicit communication with children who have yet to develop rapport however, there is no explicit direction provided for cases where the child is completely non-communicative (e.g., uses neither verbal nor nonverbal communication).

Bergman (2013) included a caregiver-only pre-treatment session where the clinician and caregivers meet to review the purpose and general progression of IBTSM. Caregivers provide detailed information about their child's speech in and across various contexts to help guide the remainder of treatment. This session also serves as an opportunity to discuss the phenomenology, associated symptoms, and common misconceptions about SM. Caregivers

receive orientation to IBTSM and its central features (e.g., contingency management, graded exposures, transfer of control). After the pre-treatment session, caregivers are encouraged to contact their child's teacher and/or school principal to notify them of his or her involvement in treatment. Bergman (2013) elaborates on IBTSM by stating, "the integrated nature of the therapy refers to the goal of integrating input from the clinician with that from the parents and teacher… together these individuals form a treatment team that constructs and implements exposure assignments" (p. 11). There is some guidance for communicating with caregivers, teachers, and other school personnel as a part of the supplemental components of the treatment manual, though it is minimal as caregivers are expected to serve as the primary means of communication between the therapist and the teacher. Clinicians wishing to engage in direct communication with the child's school teacher may need to establish a plan early on to supplement those suggestions and activities outlined throughout IBTSM.

Compared to previous manualized treatments for anxiety in general (e.g., *Coping Cat*; see Table 3), IBTSM focuses on behavioral strategies to encourage speech with less emphasis on the child's cognitive appraisal of anxiety symptoms and problem-solving. More time is given to building rapport and introducing behavioral practices (e.g., feelings chart, fear hierarchy, rewards system) to support the remainder of treatment. Exposure-based sessions begin much earlier in IBTSM (i.e., Session 3) than *Coping Cat*, which recommends introducing imaginal or *in vivo* practice during Session 10. All subsequent sessions involve both the child and at least one caregiver and focus on in-session exposures. Early on, the treating psychologist facilitates exposure activities and provides direct instruction to caregivers for out-of-session exposures. Bergman (2013) incorporates a "transfer of control" process over the course of treatment in

Table 3.

Comparison of Two Manualized Anxiety Interventions: IBTSM and Coping Cat

IBTSM (20 treatment sessions, 24 weeks)		**Coping Cat (16 treatment sessions, 16 weeks)**	
Pretreatment (Parent Only)	Assessment and Psychoeducation	*Session 1*	Building Rapport and Treatment Orientation
Session 1	Introduction, Rapport Building	*Session 2*	Identifying Anxious Feelings
Session 2	Rapport Building, Reward System, Feelings Chart	*Session 3*	Identifying Somatic Responses to Anxiety
Session 3	Classroom Chart, Fear Ladder, Exposure Practice	*Session 4*	First Meeting with Parents (Parent-only)
Sessions 4–9	Initial Exposure Sessions	*Session 5*	Relaxation Training
Session 10	Midpoint Session	*Session 6*	Identifying Anxious Self-Talk, Learning to Challenge Thoughts
Sessions 11-14	Intermediate Exposure Sessions	*Session 7*	Reviewing Anxious & Coping Self-Talk, Developing Problem Solving Skills
Session 15	Exposure, Introduction to Transfer of Control	*Session 8*	Introducing Self-Evaluation and Self-Reward and Review
Sessions 16-17	Exposure, Additional Transfer of Control	*Session 9*	Second Meeting with Parents (Parent-only)
Sessions 18-19	Exposure, Transfer of Control, Progress Review	*Sessions 10-11*	Practice Low Anxiety-Provoking Situations Using Exposure Tasks
Session 20	Relapse Prevention and Graduation	*Sessions 12-13*	Practice Moderately Anxiety-Provoking Situations Using Exposure
		Sessions 14-15	Practice High Anxiety-Provoking Situations Using Exposure
		Session 16	Practice High Anxiety-Provoking Situations, Commercial, & Terminating Treatment

which caregivers slowly take on greater responsibility in selecting and implementation strategies outside the clinic. Each IBTSM session concludes with an additional behavioral "homework assignment" for the child to complete at home, school, or in the community (e.g., at a restaurant, during a playdate with peers). These exposures become progressively challenging as children work through their fear hierarchy over time. The manual also includes time for the clinician and the caregiver to review events from the previous week, and opportunities to work through common barriers to treatment implementation.

In a clinical pilot study, IBTSM was found to be moderately efficacious (Bergman et al., 2013). Participants (N = 21) were randomly assigned to either the 20-session, 24-week IBTSM program or a 12-week wait list. Caregivers reported on their child's speaking behaviors and social anxiety levels at pre-treatment, midpoint (12 weeks), post-treatment, and a follow-up at 36 weeks. Teacher ratings were also included to assess whether generalization across settings (i.e., from the clinical setting to the school setting) occurred as a result of treatment. By the end of treatment, 67% (n = 8) of all children receiving IBTSM (n = 12) demonstrated clinically significant improvements as indicated by a removal of SM diagnosis based on the end-of-treatment diagnostic interview. Caregiver ratings showed statistically significant improvements in both speech and social anxiety by the end-of-treatment assessment, whereas teacher ratings indicated that only speech, and not social anxiety, in the school setting improved significantly. Additionally, 25% (n = 4) of the active treatment group no longer met criteria for SM by the midpoint (i.e., after 12 weeks of IBTSM). The results from this preliminary study demonstrate IBTSM's potential efficacy for treating children with SM using a standardized treatment manual. However, research is needed to verify the effectiveness of this approach in a community-based clinic through an independent investigation (i.e., without involvement from the developer).

Current state of evidence-based treatment for SM. Techniques and approaches for SM treatment have been described in the literature, though few have garnered enough empirical support to be considered evidence-based. The field of psychology has seen substantial growth within recent decades as it seeks to define and disseminate guidelines for evidence-based psychological practice (EBPP). Initial statements about "empirically-supported therapies" (ESTs), which are now more commonly referred to as evidence-based treatments (EBTs), listed a number of criteria based on the best available research for a given disorder or problem behavior

and labeled treatments on the basis of their efficacy (Chambless & Hollon, 1998). The APA (2006) Presidential Task Force on Evidence-Based Practice distinguished EBPP and ESTs by stating, "ESTs are specific psychological treatments that have been shown to be efficacious in controlled clinical trials, whereas EBPP encompasses a broader range of clinical activities (e.g., psychological assessment, case formulation, therapy relationships)" (p. 6).

While these earlier reports were useful in identifying efficacious treatment options, Kazdin and Weisz (2010) deemphasized the need for a categorical system by level of evidence and, rather, summarized the field's general understanding of evidence-based psychotherapy. They outlined six common criteria, which need to be observed in at least two investigations of a given treatment: (1) clear specification of the patient population, (2) random assignment to treatment conditions, (3) use of a manual to note intervention procedures, (4) use of multiple measures, including those assessing the target behavior, (5) evidence of statistically significant differences between treatment and control conditions at the end of treatment, and (6) evidence of a replication of treatment effect, ideally conducted by independent researchers.

The field is currently making strides to also ensure that efficacious EBTs demonstrate adequate external validity, showing their effectiveness in real-world contexts under less controlled circumstances and with diverse populations. Chorpita and colleagues (2011) addressed these recent efforts by reviewing the literature on EBTs for various psychiatric disorders. Table 4 represents the two highest levels of five proposed in this study, which were derived from the APA Division 12's Task Force on Promotion and Dissemination of Psychological Procedures (1995). The general purpose of this movement was, and continues to be, to identify and disseminate standardized treatments successfully demonstrating efficacy for

Table 4.

Strength of Evidence for Treatments for Children and Adolescents

Level 1: Best Support
i. At least two randomized trials demonstrating efficacy in one or more of the following ways: a. Treatment was superior to placebo (pill or psychological) or another active treatment b. Treatment demonstrated equivalent effects compared to other groups representing one or more Level 1 or Level 2 treatments in a study using sufficient statistical power AND treatment showed significant pre-post changes in the target group as well as groups compared to which the target group is tied. Treatments who have previously qualified using ties are ineligible for the present comparison. ii. Experiments conducted using a treatment manual
Level 2: Good Support
i. At least two studies using experimental design have shown statistically significant effects compared to waitlist or no-treatment control groups. Although recommend, manuals, independent investigations, and/or specification of the sample is not required for this level. OR ii. One experiment comparing two or more groups with a manual to guide treatment, clear specification of these groups, and evidence of efficacy via: a. Effects indicating superiority compared to placebo (pill or psychological) or other treatment approach. b. Effects equivalent to a previously-established treatment (using qualifying definition in 1ib.

Adapted from Chorpita et al. (2011)

the target behavior or disorder and to guide practitioners in selecting and implementing "best practices" to ensure clients receive high-quality and appropriate treatment. It should be noted that even within Chorpita and colleagues' (2011) extensive review of efficacious and potentially-effective treatments for disorders in childhood and adolescence, no treatment was recommended for children with SM. As the first manualized treatment for children with SM, IBTSM serves as an excellent option for a potential EBT for SM.

As the field develops innovative approaches to inform treatment selection and implementation, the literature on SM continues to lack support for one treatment approach for a specific population of children with SM. Guidance on evidence-based approaches to psychological treatment may transition away from standardized treatment protocols and instead work to distill specific components to match with individuals' presenting concerns (Chorpita et al., 2005). However, this model may be the most appropriate approach with common disorders

or problem behaviors, particularly when a large body of literature exists to support treatment options for these target populations. Comer and Barlow (2014) argue that such a transition may be difficult and unreasonable when used with low-incidence disorders like SM, stating specifically, "low base rate disorders that are best treated with specialized, uncommon methods are not addressed within treatment algorithms designed to address the broad majority of individuals seeking care" (p. 8). There is a profound need to bolster the literature on SM treatment approaches, with the goal of finding and disseminating EBTs for children with SM.

Adherence and acceptability in treatment research. While models for identifying and developing EBTs often emphasize the treatment effects (i.e., changes in dependent variable associated with the introduction of the intervention), two additional components are critical in the process considering whether an intervention should be widely-disseminated. Treatment adherence is an essential aspect of intervention research and is defined as "the extent that the therapists implement the therapy as intended" (Chambless & Hollon, 2012; p. 540). Perepletchikova and Kazdin (2005) point to the many ways treatment adherence, as one component of overall treatment integrity, informs scholars' evaluation of treatment effectiveness. First, the degree to which a clinician adheres to the treatment protocol as intended may affect the experimental validity of an intervention study. If the goal is to make inferences about a change in the dependent variable(s) after the introduction or manipulation of an independent variable (i.e., the intervention), the independent variable must be employed as it was designed. Finally, adherence data are essential in drawing inferences about the external validity of the intervention (Perepletchikova, Treat, & Kazdin, 2007). Attempts to produce replicable effects across investigations assume precise adherence to treatment procedures (i.e., high levels of adherence). Aside from its significance to intervention effectiveness research, Perepletchikova and Kazdin

(2005) also argue that adequate treatment integrity – including adherence, clinician competence, and treatment differentiation – may boost the overall success of the intervention. Researchers evaluating the effectiveness of a treatment, specifically those seeking to provide support for a standardized approach as a possible EBT, would be remiss to neglect the role of treatment adherence in supporting their observed results.

Even if a treatment is implemented as intended and results in the anticipated outcomes, perceptions of treatment acceptability may impede later dissemination and implementation. Treatment acceptability is another critical component of evaluating effectiveness, especially when scholars seek to translate a treatment program from research (i.e., efficacy trial) to practice (i.e., effectiveness research in applied settings). Acceptability can be defined as, "a judgment of whether treatment procedures are effective, reasonable, fair, or appropriate for a given problem or client" (Perepletchikova & Kazdin, 2005; p. 369). Kazdin (1980) initially highlighted the significance of treatment acceptability by noting that multiple interventions may be equally effective but vastly different with regard to their perceived acceptability. In this same paper, Kazdin (1980) explored variables affecting treatment acceptability and found that knowledge of the treatment's effectiveness had no significant influence on its overall acceptability. The structure and perceived aversive side effects may be more predictive of acceptability ratings than an understanding of how it has worked in the past. Clients' perceptions of treatment acceptability may influence treatment adherence as well (Perepletchikova & Kazdin, 2005). Client and stakeholder (e.g., caregivers, teachers) buy-in is important for ensuring full involvement and active participation throughout. A clinician's best efforts to implement a treatment as designed may still yield poor results if the client or his/her caregivers find the treatment as a whole to be unreasonable or ineffective.

Future steps toward an EBT for SM. With this general understanding of the EBT movement and the paucity of intervention research for SM, it is clear that a lack of any transportable and acceptable EBT targeting SM may serve as a major barrier to many children with SM receiving timely treatment. As the only widely-available manualized treatment for SM, IBTSM shows promise as a potential EBT but has yet to be validated to the extent necessary. The preliminary RCT by Bergman and colleagues (2013) serves as a vital first step as it meets all six of the Kazdin and Weisz (2010) criteria listed above, though future support for its effectiveness in new contexts and with new, diverse samples will be essential. Sheridan (2014) proposes one helpful conceptualization of intervention research as a linear process toward large-scale effectiveness research (Table 5).

Table 5.

Ten Steps Along the Intervention Research Trajectory

Step 1: Identify an issue or problem
Step 2: Create strategies
Step 3: Pilot/assess feasibility
Step 4: Evaluate with intensity/precision; small sample
Step 5: Replicate and extend with new sample, problem, context
Step 6: Develop theory
Step 7: Test on larger scale
Step 8: Assess mechanisms of change (theory)
Step 9: Investigate influential contextual/situational variables
Step 10: Test effectiveness on large scale

(Sheridan, 2014; p. 300)

Using this model, recent studies specifically examining IBTSM have already helped to fill in some steps along this path. The developers of IBTSM clearly identified the issue of the debilitating nature of SM and the need for early intervention (Step 1). They have compiled strategies from previous research on behavioral therapy into a single manual for dissemination (Step 2). They have compiled strategies from previous research on behavioral therapy into a single manual for dissemination (Step 2). Additionally, this manual has been piloted and found

efficacious with one sample of children with SM (Step 3; Bergman et al., 2013). Two single-case investigations have examined the intensity of IBTSM within small samples and the context in which it is effective (Steps 4-5; Cotton-Thomas, 2015; Siroky et al., 2017).

Siroky, Carlson, and Kotrba (2017) carried out IBTSM in a community-based clinical setting with two young boys with SM. Treatment length was shortened from 20 sessions over 24 weeks to 12 sessions over 18 weeks, though none of the key components of IBTSM were removed. The participating psychologist implemented this condensed IBTSM with excellent fidelity (M = 96.7%). After 12 sessions, caregivers of both children reported some increase in speaking behaviors as well as decreases in social anxiety levels. Effect sizes indicate a meaningful reduction in social anxiety in both cases when comparing means from the baseline to the midpoint (d = 1.28) and to the end-of-treatment time points (d = 2.69). Three months following the end-of-treatment, reductions in social anxiety reached clinical significance for both participants. One child no longer met criteria for SM at this follow-up assessment. An important consideration for this study was the differing presentations of the two cases. The child who saw a removal of SM diagnosis began treatment with what may be considered an "exclusively-anxious presentation" of SM and a comorbid diagnosis of social anxiety. The second child showed more frequent oppositional behaviors, corroborating Diliberto and Kearney's (2016) argument that behavioral treatment alone may be differentially effective for proposed subtypes or presentations of SM. With only a replicated treatment effect pertaining to social anxiety symptoms, this early effectiveness study points to a number of new questions to be answered through ongoing research.

Similarly, Cotton-Thomas (2015) used a multiple baseline single-case design to examine an eight-session version of IBTSM in a school setting for three children (ages 6, 8, and 10) with

SM. Effect sizes using the *no assumptions* approach were reported on both different types of speech assessed through systematic direct observations. Mean effects for non-vocal speech were large (d = 0.84) though vocal speech yielded no meaningful effect (d = -0.08). Additionally, caregiver ratings of participants' functional communication reached clinical significance, though these effects did not generalize to the school setting as teacher-reported changes in functional communication were not significant. Conclusions are also limited by the lack of data on treatment adherence for this eight-session school-based version of IBTSM. While this study provides some evidence that a brief version of IBTSM may encourage more frequent non-verbal communication in the school setting, it highlights some potential reasons as to what may not be effective for children with SM. The brevity of treatment may be one potential reason for the minimal gains seen in these children. Additionally, Bergman (2013) recommends IBTSM for children between the ages of four and eight and one child in this study was above this age range (i.e., age 10).

Results from these studies highlight some control future directions for research related to effective EBTs for children with SM. First, they show some evidence that even condensed version of IBTSM may produce significant gains in one or more symptoms of SM (e.g., social anxiety, functional communication) in children. In addition, both studies demonstrated adequate overall acceptability of condensed versions of IBTSM when implemented in community-based clinic and in a school setting. Thus, even with minimal improvements in participants' verbal communication, caregivers and school professionals viewed IBTSM as an acceptable treatment approach. However, both studies are limited by methodological concerns (e.g., insufficient baseline data to meet single-case design standards, a lack of description about treatment modifications and/or implementation), making it difficult to accurately draw conclusions about a

functional relationship between outcomes and the treatment itself. Mixed results from these investigations of brief versions of IBTSM also suggest a longer treatment approach may be necessary for meaningful changes in primary (i.e., verbal communication) and secondary symptoms (i.e., social anxiety, non-vocal speech) of SM over time.

Considering both the strengths and limitations of this preliminary research on the effectiveness of IBTSM, it will be critical for researchers to further explore those missing steps in the trajectory toward EBTs (Sheridan, 2014) in order to move IBTSM research from efficacy to effectiveness for samples of children with SM. The replicated single-case design by Siroky and colleagues (2017) also points to the importance of assessing for proposed SM clinical profiles to ensure children's presentations are closely aligned with the proper treatment approach (e.g., behavioral therapy for exclusively-anxious SM). Single-case research may be an ideal approach to conduct more precise evaluations of IBTSM in a smaller sample (Step 4) and to test the effectiveness of IBTSM through replications in community-based settings (Step 5). These investigations should also seek to carefully examine treatment adherence as clinicians implement IBTSM in new contexts, as well as acceptability ratings from relevant stakeholders.

Single-Case Research

Single-case research for intervention effectiveness. Traditionally, investigations of intervention effectiveness require rigorous research methodology to demonstrate a causal relationship between an intervention and the expected outcomes. In broader scientific research, RCTs are generally preferred as they allow investigators to exert greater control over potential confounding variables. This experimental control helps to increase the accuracy with which causal inferences are made. However, scholars in behavioral, educational, and pharmacological research have begun to demonstrate the rigor of single-case research designs for drawing valid

conclusions about intervention effectiveness for individual cases or experimental units (e.g., classrooms, schools; Kratochwill et al., 2012; Shadish & Sullivan, 2011). In addition, certain characteristics of single-case research may be particularly suitable when studying low-incidence behaviors or behavioral disorders like SM (Kratochwill et al., 2012). For instance, single-case investigations elude the practical issue of attempting to recruit a large sample. Given the rarity of SM, a well-developed RCT for this disorder would require significant time, resources, and funding. In single-case experimental studies, each participant serves as his or her own control, rather than an additional group of independent participants (Kratochwill & Levin, 2010).

Sound single-case research involves frequent and repeated measurement of dependent variables (Horner et al., 2005). This consistent assessment allows investigators to visually plot change over time, which aids in the interpretation of a treatment effect. If a noticeable change in the dependent variable occurs relatively soon after the independent variable is introduced, especially when an effect is replicated within or across subjects, investigators can be fairly confident of a functional relationship (Horner et al., 2005). Using SM as an example, a hierarchy of changes can be predicted across multiple dependent variables given the behavioral conceptualization of SM where youth first experience anxiety in certain situations where speech is expected and then withhold speech (i.e., speech avoidance) until the expectation is removed and they experience relief from physiological and/or psychological discomfort.

When examining treatment effects, reductions in anxiety would be noticed first. Reduced levels of anxiety should be followed by improvements in nonverbal communication, then increases in responsive speech (i.e., verbal communication after being prompted to do so), and ultimately in spontaneous speech (i.e., verbally communicating without being prompted to do so). These changes to might follow an individual's hierarchy of feared stimuli such as

improvements in home, then certain community settings, and finally in school (Kotrba, 2015; Skedgell et al., 2017). Replication of the effect across dependent variables and across subjects is particularly crucial to maintaining experimental control, given the inherent difficulties with generalizing results across multiple diverse participants. Finally, investigators using single-case experimental designs can employ randomization techniques to protect the internal validity of the study, and to more clearly demonstrate discriminant validity (Kratochwill & Levin, 2010).

A number of designs can be used within single-case research to critically examine intervention effectiveness. In the field of behavioral and educational research, single-case investigations seek to demonstrate a replicated treatment effect through repeated changes between a baseline (A) phase and a treatment (B) phase. These repeated transitions from A to B (e.g., A-B-A-B), when paired with continuous data collection, allow investigators to observe behavioral changes associated with the presence or absence of an independent variable (Horner et al., 2005). In behavioral and education science, an intervention is typically implemented with the goal of yielding meaningful and lasting improvements in problematic behaviors. Thus, the withdrawal of the intervention may be ineffective in reducing dependent variables back to baseline or may be unethical when the pattern of behavior significantly impairs the child's functioning (Christ, 2007). In some instances, a return to baseline may diminish improvements in symptoms (e.g., speaking behaviors, anxiety levels), which would be considered counterproductive to the goal of the treatment (Kratochwill et al., 2012). Single-case research using a multiple baseline design, instead of reversal or withdrawal designs, may be ideal when attempting to verify the effectiveness of an intervention for small samples of children diagnosed with a chronic or debilitating disorder like SM.

Multiple baseline design. Multiple baseline designs are similar to typical AB designs (i.e., one baseline phase followed by one intervention phase), but include an attempt to replicate the effect across three or more participants, behaviors, or settings (Kratochwill et al., 2010). The AB schedule is staggered, wherein the length of the baseline (A) phase varies across all units. For example, one unit (e.g., participant, classroom, behavior of interest) may be randomly assigned to a five-week baseline with five weeks of intervention, while a second unit is assigned three weeks of baseline with seven weeks of intervention. Typically, three attempts to demonstrate replication are necessary to draw valid conclusions about the overall effect of the intervention using a multiple baseline design (Horner et al., 2005).

This replication, either within a single case or across multiple cases, helps to draw valid conclusions about the treatment effect because it points to whether there is noticeable and consistent co-variation between the observed changes in dependent variables and the introduction or re-introduction of the independent variable (Horner et al., 2005). As long as the independent variable involves an active manipulation, three replications of such an effect across participants confirm this functional relationship and help to eliminate questions of potential threats to experimental control.

Studies involving multiple baseline designs also avoid ethical and practical concerns with the withdrawal of a necessary and potentially beneficial treatment (Watson & Workman, 1981). By repeating a simple AB design at staggered and potentially random time points, observable changes in the target behavior(s) can be seen across multiple subjects and the treatment need not be withdrawn to determine a treatment effect. The use of randomization within a multiple baseline design may result in greater confidence of causal inferences; however, given the practical limitations when attempting to randomly-assign predetermined, staggered baseline

phase lengths to participants, randomization is not necessary to meet single-case research design standards (Kratochwill & Levin, 2010; Kratochwill et al., 2012). Multiple baseline designs may be able to help clinicians understand the time required to see significant changes in client outcomes since the treatment phase is uninterrupted. Finally, if a number of diverse individuals show the anticipated response to an intervention, one can assume with greater confidence that others will show similar gains. While the notion of wide-spread generalizability is limited in single-case research, between-subjects multiple baseline designs may be used to test the external validity of an intervention (Horner et al., 2005). If an intervention, implemented with high overall adherence, produces replicable gains across multiple participants, it is more likely to be effective in other, diverse samples as well.

Concurrent and nonconcurrent multiple baseline designs. Multiple baseline designs offer investigators some flexibility with regard to treatment start points and the schedule of implementation. It is important to distinguish between concurrent and nonconcurrent multiple baseline designs, particularly with a design carried out across subjects. Concurrent designs involve a simultaneous initiation of data collection, beginning with the baseline phase, for every case. Baseline data collection continues until one subject establishes a stable baseline, after which the independent variable is introduced. This process continues successively across all subjects once they each reach baseline stability. Since treatment implementation is contingent upon baseline stability, concurrent designs have the benefit of avoiding potential threats to internal validity such as history or participant maturation (Christ, 2007). A nonconcurrent design, on the other hand, makes it possible for data collection to take place at dispersed time points. Watson and Workman (1981) note that nonconcurrent designs may be ideal for clinicians or investigators working in applied contexts. While concurrent designs may help to maintain

greater experimental control by avoiding potential participant mortality (i.e., exclusion of subjects who fail to demonstrate a stable or extreme enough baseline), nonconcurrent designs may be more feasible when conducting community-based investigations since the clinician need not see all cases on the same schedule.

Some scholars of single-case research criticize nonconcurrent multiple baseline designs (e.g., Harris & Jenson, 1985), arguing the inability of this design to adequately address other threats to internal validity. However, Christ (2007) contends that while the short duration and the *a priori* designation of baseline phases may make mortality of participants more common, other threats to internal validity are weakened due to the qualities of multiple baseline designs. These characteristics include: (a) well-defined experimental manipulations of the independent variable, (b) hypotheses proposed a priori, (c) frequent formative assessments of the dependent variables, (d) visual depictions of whether changes in the dependent variables coincide with manipulation of the independent variable, and (e) opportunities to demonstrate replicated effects across multiple data series or subjects. Additionally, when using multiple baseline single-case designs with individuals with a long history of symptoms, one can infer that baseline data are representative of more pervasive behavior patterns such as the ongoing and pervasive avoidance of speech in children with SM. Thus, with the contingency on baseline stability, participants have likely experienced symptom stability for a significant period of time prior to baseline data collection. Christ (2007) also notes that while history may threaten validity in nonconcurrent designs, such an event is very unlikely to coincide with the phase change from baseline (A) to intervention (B) across all subjects and is much more probable *within* a single phase. Since these above characteristics are essential for a multiple baseline experiment, whether concurrent or

nonconcurrent in nature, Christ (2007) concludes that both variations "are sufficiently robust to contribute meaningfully to the scientific literature" (p. 457).

Analysis of single-case research.

Visual analyses. Visual analyses are commonly employed first to examine intervention effectiveness within a single-case research design (Harrington & Velicer, 2015; Kratochwill et al., 2012). The use of visual analysis relies on a number of assumptions. First, data must suggest a clear and consistent baseline trend for each participant or unit. Variation in observed behaviors or performance disrupts the internal validity of the study, making it difficult to conclude whether a change in the dependent variable(s) is the direct result of the implementation of the independent variable. Second, a sufficient amount of data is needed to document permanent change or relatively durable improvements after the start of the intervention. Typically, a minimum of five data points per phase are recommended with multiple baseline designs to meet design standards for single-case research (Kratochwill et al., 2012). Once these assumptions are met, investigators can begin to visually compare data between phases and across participants to determine if a noticeable treatment effect occurred.

Kratochwill and colleagues (2012) list six characteristics to use in critically evaluating changes within phases (i.e., examining data from the treatment phase only) or between phases (i.e., comparing baseline and treatment phases). These include: (1) level, (2) trend, (3) variability, (4) immediacy of effect, (5) overlap, and (6) consistency of patterns within similar phases across multiple participants. These six features require careful interpretation of plotted data points. Level is the overall mean score within each phase. Visibly, this may look like the best-fitting horizontal line that captures a single average score for a given phase. Trend, on the other hand, reflects the slope of a line that best fits the change in data over the course of the

treatment phase. Variability can be conceptualized as the visible range, variance, standard deviation, or general scatter from the line that best fits the plotted data. If variability is small, data would be closely, if not perfectly, situated along the best-fitting line; however, widespread variability affects the internal validity of the treatment since an inconsistent pattern questions the extent to which changes in the dependent variable are a direct result of the treatment itself.

The immediacy of effect is the amount of time between the implementation of the independent variable and the first sign of a visible change in the dependent variable. The *What Works Clearinghouse* Single-case Design Technical Documentation Manual (Kratochwill et al., 2010) recommends comparing the level of the final three baseline data points to the first three data points in the treatment phase. Observable change within this window provides a stronger argument for a true treatment effect, unless a delayed or prolonged effect is anticipated. During the visual analysis, investigators should also consider what percentage of data from different phases overlap in level or trend. The final characteristic, consistency of data across similar phases, is particularly important for multiple baseline designs. For multiple baseline investigations, this may be conceptualized as a visual indication of a replicated effect. When looking at similar phases across units (e.g., comparing data from the treatment phase of each participant), a consistent trend in data demonstrates greater confidence with which one can assume the independent variable is functionally related to the observed change in the dependent variables, even with staggered start points and across unique individuals,

Quantitative analyses for single-case research. While visual analyses are crucial to, and sufficient for, the evaluation of intervention effectiveness (Kratochwill et al., 2012), investigators may wish to supplement visual indicators of a treatment effect with more quantitative analyses. Scholars in single-case research have employed quantitative and statistical approaches to verify a

functional relationship between changes in the outcome variables (i.e., dependent variables) and the intervention (i.e., independent variable). Visual analyses may demonstrate a repeated functional relationship between the independent variables and dependent variables across subjects; however, the size or magnitude of this relationship (i.e., treatment effect) cannot be gleaned from visual analyses alone.

Effect sizes. Effect sizes help to quantify the magnitude of treatment effects to support the overall social validity of the treatment (Horner et al., 2005). In addition, effect sizes are necessary for aggregating results from multiple single-case investigations on a given intervention or treatment approach. Researchers can assess the combined effects of the treatment to inform evidence-based practice via meta-analyses of single-case investigations (Horner, Swaminathan, Sugai, & Smolkowski, 2012). A number of effect sizes have been created specifically for single-case research designs. For example, Parker, Vannest, and Davis (2011) reviewed nine potential effect size estimates using nonoverlapping data. Non-overlapping data are those data points on a plotted graph that do not exceed the extreme (i.e., minimum or maximum) point identified in the other phase. Non-overlap methods for calculating effect sizes are more robust than standard mean, median, or mode calculations, but are relatively simple to derive from all available visual points on a graph (Parker, Vannest, & Davis, 2014). Examples of non-overlap methods include: Percent of data Exceeding the Median (PEM), Extended Celeration Line (ECL), Percent of Non-Overlapping Data (PND), Percent of All Non-Overlapping Data (PAND), Non-Overlap of All Pairs (NAP), and Tau-U indices.

One particularly useful effect size is known as the Tau-U index. The Tau-U index combines the use of all non-overlapping data with consideration of potential trend in the treatment phase to determine the magnitude of change over time (Parker, Vannest, Davis, &

Sauber, 2011). An advantage of the Tau-U index is that it allows one to identify, and correct for, significant baseline trends prior to determining the statistical significance of changes in dependent variables over time. According to Parker and colleagues (2011b), the first step in calculating the Tau-U effect size is to identify the number of pairwise comparisons for all data points in both the baseline and treatment phases by multiplying the number of Phase A data points by the number of Phase B data points. Then, specific comparisons are made by pairing up each Phase A data point with each Phase B data point (i.e., all possible pairs). Next, codes are given to each pairwise comparison using one of three possible codes: (a) improvement over time (+), (b) reduction or decrease over time (-), or (c) tie (T). The difference between the number of positive codes and the number of negative codes yields $S_{\text{novlap.}}$ Finally, the Tau-U index is calculated by dividing S_{novlap} by the total number of pairs, identified in the first step. Tau-U is presented as a percentage, representing "the percent of non-overlapping data minus the percent of overlapping data" (p. 2, Parker et al., 2011b).

Single-case investigations of SM treatment. The rarity of SM in clinical populations has historically precluded scholars' ability to carry out large-scale studies to examine the appropriate treatments for SM. While there are a small number of descriptive studies with larger samples of children and adolescents with SM (see Ford et al., 1998), many experimental investigations of SM treatment involve a small number of participants. In a recent review of SM treatment research, Zakszeski and DuPaul (2016) point out both the paucity of research on SM treatment in general, as well as the lack of attention to research design standards to support causal inferences. Across all 21 studies included in their review, a total of 129 children with SM were represented in the treatment literature specifically targeting SM symptoms. The largest sample size reported in any one study consisted of 24 children with SM (Lang et al., 2016). This

likely reflects anticipated barriers to recruiting a sufficient number of children with SM. As a result, researchers often use single-case methodologies to explore intervention effectiveness in smaller samples. Out of the 21 studies reviewed, the most common methodology was single-case experimental design (n = 9, 43%). The authors note, however, that only two of these employed a design that allowed for valid conclusions about a functional treatment effect (Beare et al., 2008; Vecchio & Kearney, 2009). Based on this review, it is imperative that SM scholars pay careful attention to single-case research design standards if they hope to build the literature for later meta-analyses necessary for informing evidence-based practice.

Research Questions and Hypotheses

As an important step in verifying IBTSM as a potential EBT for children with SM, the current study examined the adherence, effectiveness, and acceptability of the manualized IBTSM treatment program (Bergman, 2013) in alleviating SM symptoms in five children referred to a community-based clinic using a nonconcurrent multiple baseline design. With limited results on the effectiveness of 8-session and 12-session versions of IBTSM, a 16-session version of IBTSM may be sufficient to produce replicable improvements across these five cases since this represents the mean length between the original version of IBTSM (i.e., 20 sessions over 24 weeks) and the most recent effectiveness trial which yielded some significant effects on children's social anxiety after receiving 12 sessions of treatment over 18 weeks (Siroky et al., 2017). Additionally, due to recent studies suggesting children with distinct presentation of SM may respond differently to certain treatment approaches, a focus on children with the proposed subtype of exclusively-anxious SM was important to determine whether IBTSM may be most beneficial for children with this presentation of SM. The current study examined four research questions and hypotheses (see Table 6 on p. 55).

Question 1. Can a condensed (i.e., 16-session) version of IBTSM be carried out as intended by novice clinicians in a community-based clinical setting across all five cases?

One of the benefits of manualized treatments is the increased likelihood that treatments will be carried out as intended (Perepletchikova & Kazdin, 2005). Five children with SM received 16 sessions of IBTSM from clinicians in a community-based clinical setting, with treatment lasting 19 weeks, on average (*Range* = 16-22 weeks). The use of a therapist binder, derived from the IBTSM treatment manual (Bergman, 2013), along with individual session checklists was used to support clinicians in the implementation of IBTSM for these five cases. Additionally, study clinicians received training from a clinical child psychologist who specializes in treating children with SM. It was hypothesized that study clinicians would implement this condensed version of IBTSM with high treatment adherence. More specifically, an overall adherence percentage of 80% or higher was expected, which is considered the minimum rating required for adequate treatment adherence (Perepletchikova & Kazdin, 2005). This hypothesis is in line with the excellent treatment adherence for IBTSM reported by Bergman and colleagues (2013; *M* = 99.3%) and Siroky et al. (2017; *M* = 96.7%).

Question 2: Will a condensed version of IBTSM lead to a decrease in caregiver-rated social anxiety levels across the baseline phases and treatment phases for five children with exclusively-anxious SM?

Based on findings from the original IBTSM pilot study, a noticeable reduction in social anxiety symptoms across these five cases by the end of treatment was expected. Bergman and colleagues (2013) found significant decreases in caregiver-reported social anxiety for participants in the treatment group when compared to the waitlist group. These improvements were seen at as early as the midpoint assessment (week 12) and were maintained through end-of-

treatment and the 36-week follow-up. Similarly, a visible decrease in the overall level of social anxiety ratings, as compared to each child's baseline scores, and a steady downward trend relatively soon after the first treatment session were anticipated. Moderate to large effect sizes for caregiver-reported social anxiety were expected.

Question 3: Will a condensed version of IBTSM lead to improvements in observed speaking behaviors across settings between the baseline phases and treatment phases for five children with exclusively-anxious SM?

The overall goal for the current study was to provide evidence for the potential effectiveness of a condensed IBTSM in reducing speech avoidance for this sample of five children with exclusively-anxious SM. After 16 sessions of IBTSM, visible improvements in the overall level and slope of caregiver-reported speaking behaviors across contexts, compared to each child's baseline data, were anticipated. Caregiver ratings of observed speech were used to capture how far each child progresses along a common hierarchy of speaking behaviors (i.e., from non-communicative to responsive speech to spontaneous speech) at home, in school, or in other public situations. An increase in responsive words spoken during weekly analog behavioral observations shortly after treatment begins was also expected. Using a two-part analysis, it was also hypothesized that changes would be clinically meaningful. This hypothesis is consistent with previous results from Bergman and colleagues' (2013) original pilot study where children receiving IBTSM made significant improvements in caregiver-reported speech and words spoken to teachers by the end of treatment. Additionally, 67% (n = 8) of in the treatment group longer met criteria for SM after IBTSM, suggesting these changes were clinically significant for a majority of the sample.

Question 4: Will caregivers rate the condensed (i.e., 16-session) version of IBTSM as an acceptable treatment approach for children with exclusively-anxious SM, specifically in the areas of treatment quality, effectiveness, and time required?

High overall treatment acceptability across all five participants was anticipated. This prediction was based on results from Bergman and colleagues (2013), who reported high caregiver satisfaction (M = 3.79/4.00, SD = 0.29) on the Client Satisfaction Questionnaire (CSQ; Hargreaves & Attkisson, 1978) after children received 20 sessions of IBSTM over 24 weeks. In a preliminary community-based study, caregivers of two children rated a condensed version (i.e., 12 sessions over 18 weeks) of IBTSM as highly acceptable across all three of these domains (Siroky et al., 2017). High ratings related to time required were expected across all five cases as well, given the present study's attempt to reduce the length of treatment to 16 sessions while retaining all key components of IBTSM. This structured approach to SM treatment was also expected to receive high ratings of treatment quality given its early focus on orienting caregivers and children to the goals of treatment and the use of the "transfer of control" process. Perepletchikova and Kazdin (2005) note that stakeholders' knowledge about the treatment mechanism(s) and treatment goals may boost treatment acceptability. Finally, given anticipated effectiveness across all five children, it was hypothesized that caregivers would view the condensed version of IBTSM as effective in reducing SM symptoms for their child.

Table 6

Research Questions with Corresponding Hypotheses and Measures

Research Question	**Hypothesis**	**Measures**
Q1: Can a condensed (i.e., 16 sessions) version of IBTSM be carried out as intended by novice clinicians in a community-based clinical setting across all five cases?	Study clinicians will carry out condensed IBTSM (i.e., 16 sessions) as intended for all five cases, as indicated by an aggregated adherence score of 80% or higher.	*Entire treatment phase*: Adherence checklists derived from treatment manual (Bergman, 2013) *Four (25% per case) sessions:* Direct observations of in-clinic sessions
Q2: Will a condensed version of IBTSM lead to a decrease in caregiver-rated social anxiety levels across the baseline phases and treatment phases for five children with exclusively-anxious SM?	Five cases will demonstrate significant decreases in caregiver-rated social anxiety levels, as compared to baseline, after receiving a condensed version of IBTSM.	*Pre/Post:* Social Anxiety Scale for Children – Parent Form, Total Score *Daily during baseline,* *Weekly during treatment*: Social Anxiety Scale for Children – Parent Form, FNE subscale
Q3: Will a condensed version of IBTSM lead to improvements in caregiver-reported speaking behaviors across settings between the baseline phases and treatment phases for five children with exclusively-anxious SM?	Five cases will demonstrate significant improvements in caregiver-reported speaking behavior across contexts, when compared to baseline ratings, after receiving a condensed version of IBTSM.	*Pre/Post:* Selective Mutism Questionnaire – Total Score Anxiety Disorders Interview Schedule for Children – Parent Interview (ADIS-P) *Daily during baseline,* *Weekly during treatment:* Brief Rating of Observed Speaking Behaviors *Weekly:* Frequency count of words spoken during analog observations
Q4: Will caregivers rate the condensed (i.e., 16-session) version of IBTSM as an acceptable treatment approach for children with exclusively-anxious SM, specifically in the areas of treatment quality, effectiveness, and time required?	Caregivers of all five participants will rate condensed IBTSM as an acceptable treatment for SM with regard to quality, effectiveness, and time required.	*End-of-treatment:* Treatment Evaluation Questionnaire – Parent Form

CHAPTER 3

METHODS

Participants

Five children (ages 4-8 years) were enrolled as participants in the present study. After receiving approval from the Michigan State University – Biomedical Institutional Review Board (MSU-BIRB), recruitment followed the steps described in the *Procedures* section (page 69). Paper fliers (Appendix C) outlining the goal, length, cost, and potential risks/benefits of the study were distributed at both clinic locations and shared on the clinic's website for others to access. A total of 13 families contacted the study coordinator to express interest. Of these, seven families (53.8%) moved on to the screening process to determine eligibility, while the others (n = 6; 46.2%) were not eligible due to distance (i.e., lived out of state, lack of availability in schedule to travel to clinic) or initial exclusion criteria (e.g., recently enrolled in intensive-dose behavioral therapy for SM). Following screening with the remaining seven families, the first five children who met eligibility criteria were enrolled. One child was determined ineligible based on exclusionary criterion for SM symptom severity (i.e., symptoms too severe based on SMQ total score at baseline) and one child was eligible but returned information shortly after the fifth family was enrolled. Table 7 provides baseline demographic information about

Table 7

Baseline Demographics by Participant

	Age	**Sex**	**Race**	**ADIS-P, Clinical Severity Rating (CSR)**				**SMQ**
				Selective Mutism	*Social Phobia*	*GAD*	*Separation Anxiety*	*Total Score at Baseline*
Child 1	8	M	Biracial	8	8	5	5	16
Child 2	7	M	Caucasian	8	5	n/a	n/a	13
Child 3	6	F	Caucasian	4	n/a	n/a	n/a	17
Child 4	6	M	Caucasian	6	6	7	n/a	16
Child 5	4	F	Caucasian	7	n/a	n/a	n/a	15

Note: A CSR of 4 or higher indicates severity meeting clinical significance.

each of the five children enrolled in the present study. Three of five participants (60%) were male, and four of five participants (80%) were White/Caucasian. Ages of participating children ranged from four to eight (M = 6.2 years) at baseline. All families received one pre-treatment intake session and 16 active treatment sessions at no cost. Families also received $200 of compensation ($50 at Session 1, $100 at Session 8, $50 at Session 16) for travel to/from the clinic, rewards/tokens to support the contingency management component of treatment, and for their active participation in the study.

Inclusion and exclusion criteria. In order to be eligible to participate, children had to have been between the ages of four and eight at the time of recruitment. Bergman (2013) specifies this age range for IBTSM given the emphasis of behavioral, rather than cognitive, approaches and the typical age of onset for SM. All five children presented with symptoms consistent with a diagnosis of SM, as confirmed by a semi-structured caregiver intake interview used for all cases receiving treatment from the participating clinic and a supplemental comprehensive diagnostic interview (i.e., Anxiety Disorders Interview Schedule for Children – Parent Interview [ADIS-P]).

Due to early research suggesting that behavioral therapy may be most appropriate for children with the proposed subtype of exclusively-anxious SM, children eligible for this study also needed to demonstrate symptoms consistent with an exclusively-anxious presentation of SM, rather than mildly-oppositional/sensitive anxious SM or bilingual/ communication-delayed anxious SM (see Darr et al., 2016; Cohan et al., 2008). All five participants demonstrated stable symptoms (e.g., persistently low frequency of speaking behaviors across settings) during the baseline phase. Finally, because IBTSM does not incorporate strategies for eliciting speech in very severe SM cases (e.g., shaping; see Kotrba, 2015), participants were screened for moderate

SM severity, as indicated by a total score on the Selective Mutism Questionnaire (Bergman et al., 2008) between 13 and 27 (i.e., mean for children with SM plus two standard deviations above the mean). All children presented with elevated symptoms of social anxiety at baseline, as rated by caregivers using the Social Anxiety Scale for Children-Revised, Parent Form (SASC-R, Parent Version; La Greca & Stone, 1993). However, following enrollment in treatment, a calculation error was discovered in Child 4's SASC-R total score at baseline. Although this child's baseline score of 46 on the SASC-R at baseline fell below the identified cut-off for eligibility based on the clinical cut-off for boys (*Total* = 50; La Greca, 1999; La Greca & Stone, 1993), supplemental data gathered through the diagnostic interview with caregivers also demonstrated clinically elevated symptoms of social anxiety consistent with a clinical diagnosis.

The integrative nature of IBTSM relies on the consistent involvement from caregivers and their willingness to communicate with school personnel. To ensure caregivers' understanding of this aspect of IBTSM, caregivers were asked to confirm their commitment to engaging in and facilitating exposure activities prior to proceeding with the screening procedures, and again during the caregiver consent process at the first appointment. To avoid potential conflicting or invalid results for this treatment effectiveness study, participants must not have been receiving any other form of treatment for SM, including other psychosocial treatment approaches or psychopharmacological treatment. Although not a formal inclusion criterion, it was recommended that participants be enrolled in a full-time school program (e.g., preschool, early childhood center, elementary school) for the entirety of the study (Bergman, 2013). Four of five participants were enrolled in full-time school for a majority of the time they were in treatment, and the fifth participant (Child 5) was enrolled in part-time preschool for three afternoons per week.

Measures

Treatment adherence. In order to assess overall treatment adherence, the first author developed checklists (Appendix D) for each session based on session descriptions/goals listed in the IBTSM manual (Bergman, 2013). Immediately following each session, study clinicians indicated the extent to which they implemented each session component using a four-point scale, with options ranging from 0 to 3. Specific item-level responses included: "No attempt was made", "Attempted but not successful", "Attempted and partially successful" and "Successful." Percentages of adherence were derived from item-level responses after coding them into dichotomous No/Yes categories. A score of 0 or 1 ("no attempt" or "unsuccessful attempt") was coded as "No" while a score of 2 or 3 ("partially successful attempt" or "successful attempt") was coded as "Yes." The number of components coded as "Yes" was divided by the total number of required components to yield an adherence percentage. Although not a primary focus of this present study, strength of adherence can be assessed using this four-point scale, with higher scores indicating stronger adherence to the required IBTSM components.

At the end of treatment, total adherence percentages were calculated for each participant to determine overall treatment adherence for this study. Four (i.e., just over 20% of 16 treatment sessions) in-clinic sessions for each participant were randomly selected for observation (live or via videotape) to assess inter-observer agreement for overall adherence. The primary investigator viewed videotapes of the randomly-selected sessions and completed a separate session checklist for the observed session (i.e., Session 5 checklist if observing the child's fifth treatment session) to note the number of items the observed clinician completed. Inter-observer agreement, calculated using the percentage of agreement between clinicians and the first author, was 94.2%. Kratochwill et al. (2010) note that inter-observer agreement of 80% or higher is adequate.

Table 8.

Overview of Assessment Plan by Phase

Study Phase	Assessment Plan	Variable
Intake	Behavioral Concerns Inventory*	*Clinical profile of SM
	Clinical Intake Interview with Developmental and Medical History*	
	Social Anxiety Scale for Children – Parent Version (Full scale)*	Social anxiety severity
	Selective Mutism Questionnaire (Full scale)*	SM symptom severity
	Anxiety Disorders Interview Schedule for Children – Parent Interview	Clinical diagnoses
	Screen for Child Anxiety Related Disorders – Parent Version	
Baseline/Treatment	*Once weekly:* Analog observations	Words spoken
	Once weekly: Session checklists	Treatment adherence
	Daily during baseline, *Weekly during treatment:* Social Anxiety Scale for Children – Parent Form (FNE subscale)	Caregiver-reported social anxiety levels
	Daily during baseline, *Weekly during treatment*: Brief Ratings of Observed Speaking Behaviors	Caregiver-reported speaking behaviors across contexts
	Four times per case: Direct observation of treating clinicians	Treatment adherence (inter-observer agreement)
End-of-treatment	Treatment Evaluation Questionnaire	Treatment acceptability
	Social Anxiety Scale for Children – Parent Version (Full scale)	Social anxiety severity
	Selective Mutism Questionnaire (Full scale)	SM symptom severity
	Anxiety Disorders Interview Schedule for Children – Parent Interview	Clinical diagnoses
	Screen for Child Anxiety Related Disorders – Parent Version	

Multiple baseline measures. The use of multiple, valid measures in single-case research is key to ensure greater confidence with which accurate conclusions can be drawn about each of the four research questions. Multiple measures were used to assess the following dependent

variables: treatment adherence, clinical profile of SM, caregiver-reported social anxiety levels, caregiver-reported speaking behaviors, observed words spoken, and caregiver satisfaction with treatment. Table 8 outlines the assessment plan across three phases – intake, baseline/treatment, and end-of-treatment – related to treatment adherence, effectiveness, and acceptability.

Social anxiety. Caregivers completed the eight-item *Fear of Negative Evaluation (FNE) subscale* from the SASC-R (Appendix E) to capture participants' social anxiety symptoms related to perceptions of peer evaluation. Caregivers completed this brief rating every day during the baseline phase, then weekly during the treatment phase. The SASC-R contains three subscales, which categorize items into the areas of (1) fear of negative evaluation (FNE; 8 items), (2) social avoidance and distress in new situations (SAD-New; 6 items), or (3) general social avoidance and distress (SAD-G; 4 items). Caregivers completed the full SASC-R scale at baseline via the screening packet, and again at the end-of-treatment (after Session 16). Subscale items a five-point scale (1 = Not at all, 3 = Sometimes, 5 = All the time). As with the full scale, there are no clinical cut-offs for subscales on the SASC-R but Ginsburg, La Greca, and Stone (1998) report a mean of 23.30 (*SD* = 9.15) on the FNE subscale for socially anxious school-age children. The FNE subscale was selected because it demonstrates high reliability ($\alpha = .86$; La Greca & Stone, 1993) and represented moderate severity (i.e., neither highest nor lowest mean scores) in a previous study of IBTSM for children with SM in a community-based setting suggesting adequate sensitivity to potential treatment effects over time (Siroky et al., 2017).

Observed speaking behaviors. As a part of this single-case experimental design, frequent assessment of the primary symptom of SM (i.e. speech avoidance) was essential for determining the presence of a treatment effect. Two measures of observed speaking behaviors were used to track incremental changes across baseline and treatment phases for all five cases of children with

exclusively-anxious SM. These include: (1) caregiver ratings of speaking behaviors across settings and (2) number of words spoken in weekly analog behavioral observations.

Brief ratings of observed speaking behaviors. Along with the SASC-FNE subscale, caregivers completed Brief Ratings of Observed Speaking Behaviors (BROSB; Appendix F) every day during baseline and once per week during treatment to monitor incremental changes in speaking behaviors. The BROSB is a Direct Behavior Rating (DBR; Chafouleas, Riley-Tillman, & Christ, 2009) created specifically for the present study. DBRs are typically brief, repeatable assessments through which an observer (e.g., caregiver) provides a rating using operationally-defined target behaviors to monitor progress over time (Christ, Riley-Tillman, & Chafouleas, 2009). DBRs with multiple items per scale (DBR-MIS) are shown to be sensitive to changes in behavior over time, and they require fewer observations to reach adequate dependability when compared to single-item DBRs (Volpe & Briesch, 2012).

The BROSB is a DBR-MIS consisting of three items, derived from the three subscales of the Selective Mutism Questionnaire (Bergman et al., 2008): home, school, and other social situations. Each item asks caregivers to rate the frequency and type of their child's speaking behaviors for a given day/week using a seven-point Likert scale. These responses, ranging from 0 to 7, include: "Does not communicate (neither verbal/non-verbal)," "Infrequently communicates nonverbally," "Frequently communicates nonverbally (but no speech)," "Responds verbally, but does so infrequently," "Frequently responds verbally," "Consistently responds verbally but does not spontaneously initiate speech," "Spontaneously initiates verbal speech but does so infrequently," or "Frequently and spontaneously initiates verbal speech." Total scores range from 0 to 21, with higher scores on the BROSB indicating increased frequency and complexity of speech across settings.

Analog behavioral observations. Analog behavioral observations (ABO) were used to supplement caregiver ratings of speaking behaviors and to obtain quantitative data on speech frequency. ABOs are used to create an environment in which the target behavior can be observed naturally, while also exerting some control over the observation context to prevent potential extraneous variability (Heyman & Slep, 2004). As a part of each treatment session, the child, his or her caregiver, and the treating clinician engaged in an analog activity that encouraged speech. The goal of this observation was to assess the number of words spoken in a controlled situation with consistent expectations for speech. Caregivers were asked to follow a pre-developed protocol (Appendix G), in which they asked their child to look at a picture book and describe the image on the page. Clinicians used a basic fading in technique, where they gradually increased their attention to the child's speech. The extent to which clinicians were present and attentive to a child's speech was determined by each child's baseline fear hierarchy, which was established during the first intake session and varied across all five cases. Bergman's (2013) Situation Rating Form (Appendix A) and Fear Ladder (Appendix B) were used to identify parameters for this analog observation of medium difficulty for each child.

Clinicians were asked to note the number of words spoken during each ABO; however, project assistants reviewed four randomly-chosen ABO sessions (i.e., 25% of a maximum of 16 total observations) to determine inter-observer agreement using percentage of agreement between the clinician and the designated reviewer. Supplemental data were gathered regarding the child's spontaneous speech (i.e., instances of speech in the absence of a prompt), though this data did not serve as a main dependent variable given anticipated infrequency of spontaneous speech in clinical samples of children with SM. For every word spoken, the child received one sticker or token, which they used to redeem a prize at the end of the treatment session. Each

child received a sticker or token for any word spoken and the child's description did not need to be accurate. This task was selected so that speech and subsequent positive reinforcement was not contingent upon a child's academic, cognitive, or expressive language abilities. Each ABO lasted five minutes and took place before each of the 16 treatment sessions. The final ABO for session 16 took place during the final five minutes of the last treatment session.

Baseline to end-of-treatment measures. Due to the behavioral conceptualization upon which IBTSM was developed, it is likely to be most effective for children with the "exclusively-anxious" subtype of SM as described by Cohan and colleagues (2008) and supported in recent work by Darr et al. (2016). The focus on this exclusively-anxious SM presentation was derived from the differential effectiveness for IBTSM implemented with two different cases of children with SM (Siroky, Carlson, & Kotrba, 2017) Specifically, the child who presented with primarily anxious symptoms and no notable comorbid behavioral concerns experienced significant improvements in SM symptoms over the course of treatment, and no longer met criteria at the three-month follow-up. In contrast, the child who presented with more oppositional behaviors, as reported by the treating clinicians, did not see this magnitude of improvement by the end-of-treatment or the follow-up time points. This effort to align children's clinical profile or subtypes of SM with treatment has recently been presented by Skedgell, Fornander, and Kearney (2017). Participants needed to first meet criteria for the exclusively-anxious SM clinical profile through confirmation of an absence of oppositional-anxious or anxious-language impaired subtypes, as these behavior patterns and/or comorbid impairments likely require additional strategies (e.g., focus on positive reinforcement for compliance, speech therapy addressing articulation or expressive language deficits) beyond the scope of IBTSM. Unfortunately, there is no consistent, validated approach to subtype determination described in the SM literature. For the purposes of

this study, subtype determination used multiple assessments of speaking behaviors, anxiety symptoms, aggressive behaviors, and history of speech delay.

Rule-out assessment. Following Darr and colleagues' (2016) analysis of caregiver rating scales and reports of developmental history to inform SM subtype, the *Behavioral Concerns Inventory* and a comprehensive intake interview was used to rule out the presence of characteristics associated with other potential subtypes of SM (e.g., mildly-oppositional/sensitive anxious SM, bilingual/communication-delayed anxious SM). According to Darr et al. (2016), the BCI is a 28-item checklist used to help caregivers indicate specific concerns they have about their child's behavior. This list contains items describing various internalizing (e.g., "Seems sad," "worries a lot") and externalizing (e.g., "Disobeys parents," "Gets angry easily") behaviors. Specific items on the BCI were found to yield high variable predictor importance in highlighting children with SM who also displayed oppositional or aggressive behavior. These included: "Argues a lot," "disobeys parents," "Fights with other students," and "Takes things that don't belong to him/her" (Darr et al., 2016). If a caregiver reported two or more of these behaviors on the BCI, the child's presenting symptoms would not be considered exclusively-anxious SM and this family was not asked to participate in the study.

Additionally, a *comprehensive clinical intake interview* was administered during the pre-treatment intake session to gather information about the child's developmental history, medical history, and current diagnoses. Participating clinicians completed the intake interview using the clinic's own interview form, which includes an opportunity for caregivers to report the child's developmental milestones, such as toileting, motor skills, and language development. Caregivers were also asked to report family history of anxiety, including SM, or other psychiatric disorders. Since Darr and colleagues (2016) found that caregiver report of speech delay yielded high

predictor variable importance (1.00), caregivers must confirm the absence of a speech delay during the developmental milestones section of the clinic intake interview as a part of the present clinical profile assessment. None of the caregivers interested in participating in this study endorsed a history of speech/language delays.

Social Anxiety Scale for Children-Revised (SASC-R). Given the clear link between SM and social anxiety, caregivers completed the *Social Anxiety Scale for Children-Revised, Parent Form* (SASC-R, Parent Version; La Greca & Stone, 1993; Appendix H) to assess whether each child presented with elevated levels of social anxiety reflective of an exclusively-anxious SM clinical profile. The SASC-R full scale was administered at pre-treatment and post-treatment to compare baseline and end-of-treatment scores for clinical significance. The SASC-R consists of 18 items asking caregivers to rate the severity of their child's social anxiety symptoms. Caregivers use a five-point rating scale (1 = Not at all, 3 = Sometimes, 5 = All the time) to indicate the extent to which each of the 18 statements is true for their child. Total scores on the SASC-R range from 18-90, with higher scores reflecting more severe social anxiety symptoms. Currently, the SASC-R, Parent Version has no cut-off score to denote clinical significance, though the accompanying manual states that school-age boys receiving a score at above 50 and school-age girls with a score at or above 54 indicate high levels of social anxiety (La Greca, 1999; La Greca & Stone, 1993). The full caregiver form demonstrated adequate reliability in the IBTSM pilot study ($\alpha = .87$; Bergman et al., 2013) and in a school-based sample of children ($\alpha = .91$; Bergman et al., 2002). As a part of the clinical profile assessment, children eligible for treatment were required to demonstrate a total SASC-R score at or above 50 (boys) or 54 (girls).

Selective Mutism Questionnaire (SMQ). Caregivers completed the *Selective Mutism Questionnaire* (SMQ; Bergman et al., 2008; Appendix I) at baseline, with the screening packet,

and again at the end of treatment. The baseline administration was used to determine the child's SM clinical profile, while a comparison between baseline and post-treatment scores was used to examine changes in SM severity over time. The SMQ contains 17 items, which allow caregivers to assess the frequency of their child's speech across three contexts: Home, School, and Other/Public. Possible total scores range from 0-51, with low scores indicating less frequent speech. Although there is no cut-off score for the SMQ indicating clinical severity, Bergman and colleagues (2008) report a mean total score of 12.99 (*SD* = 7.23) for children with a primary diagnosis of SM. This investigation also demonstrated the accuracy of the SMQ in assessing baseline SM symptoms, as well as its sensitivity to behavioral change in response to treatment. The full SMQ scale has adequate discriminant and convergent validity (Bergman et al., 2008), and the most recent investigation of IBTSM revealed a Cronbach's alpha of $\alpha = .78$ (Bergman et al., 2013). Since IBTSM does not include shaping procedures for children with more severe presentations of SM, children with moderate SM symptoms only were asked to participate. Moderate SM severity was indicated by a range of scores, derived from the population mean and two standard deviations above this mean (i.e., range of 13-27).

Anxiety Disorders Interview Schedule (ADIS-P). The *Anxiety Disorders Interview Schedule for Children, Parent Version* (ADIS-P; Silverman & Albano 1996) was used to assess diagnostic status for all five cases and to corroborate the rule-out assessment, ensuring participants did not present with other common childhood disorders for which other treatment would be indicated. Caregivers completed the ADIS-P at two points: prior to the first active treatment session and after the final active treatment session (i.e., Session 16). The ADIS-P is a comprehensive structured interview schedule used to measure a child's symptoms of anxiety and the extent to which these anxious symptoms interfere with functioning in school, at home, or in

social situations. Caregiver responses yield a clinical severity rating (CSR) for each anxiety disorder, which can range from zero to eight. A CSR at or above four reflects anxious behaviors within the clinical range and serves as an indicator that these symptoms meet diagnostic criteria (Silverman & Albano, 1996). According to Silverman, Saavedra, and Pina (2001), the ADIS-P demonstrates good to excellent reliability ($\kappa = 0.65\text{-}0.88$) for diagnosing anxiety disorders listed in the DSM-IV. Other studies also support the ADIS-P as a valid measure for identifying childhood anxiety disorders (Wood, Piacentini, Bergman, McCracken, & Barrios, 2002).

Screen for Child Anxiety Related Disorders (SCARED). Caregivers also completed the *Screen for Child Anxiety Related Disorders – Parent about Child Version* (SCARED, Birmaher et al., 1999; Appendix J) at baseline and at the final treatment session. The SCARED is a brief broadband measure used to assess the frequency and severity of various symptoms in children. The SCARED consists of 41 items describing anxious thoughts, feelings, or behavioral/physiological symptoms. Parents use a three-point scale, ranging from 0 ("Not true or hardly ever true") to 3 ("Very true or often true") to rate how true each statement is for their child. Scores on the SCARED are categorizes into five subscales: Panic Disorder/ Somatic Symptoms, Generalized Anxiety Disorder, Separation Anxiety, Social Anxiety Disorder, and Significant School Avoidance. Total scores range from 0 to 123, with higher scores indicating more frequent anxiety symptoms. A total score at or above 25 warrants clinical attention as the presence of an anxiety disorder is likely. Birmaher and colleagues (1999) report high internal consistency for the 41-item measure, reporting an approximate reliability of $\alpha = .90$ for both the Child and Parent versions of this form.

Treatment acceptability. Since treatment acceptability may affect overall success and integrity of the treatment (Perepletchikova & Kazdin, 2005), the present study assessed caregiver

acceptability ratings of the condensed IBTSM. Caregivers completed the *Treatment Evaluation Questionnaire* (TEQ; Kelley, Heffer, Gresham, & Elliott, 1989; Appendix K) at the end of treatment to rate their overall satisfaction with IBTSM. The TEQ contains 21 items on which respondents rate the general quality and acceptability of the treatment approach using a 6-point Likert scale (1 = strongly disagree, 6 = strongly agree). The TEQ also contains three subscales assessing Acceptability (11 items), Effectiveness (8 items), and Time Required (2 items). The overall score is the sum of all item responses and ranges from 21 to 126, with higher total scores indicating greater satisfaction. When interpreting the Acceptability, Effectiveness, and Time subscales, scores at or above 55, 36, and 9, respectively, typically indicate high satisfaction (Kratochwill, Elliott, Loitz, Sladeczek, & Carlson, 2003). Thus, a total score of rating of 110 or above indicates high overall acceptability. Overall treatment acceptability was calculated by aggregating ratings across all five cases and comparing this score to the cut-offs listed above. The TEQ was adapted into a questionnaire format from the Treatment Evaluation Inventory (Kazdin, 1980), which has high internal consistency $\alpha = .97$.

Procedures

The purpose of the current study was to examine the adherence, effectiveness, and acceptability of a condensed, 16-session version of IBTSM for five children with exclusively-anxious SM carried out in a community-based clinical setting. Upon receiving approval from the MSU-BIRB, five children were recruited from a community-based psychology clinic in mid-Michigan, which specializes in childhood anxiety disorders including SM. Caregivers were oriented to the study and treatment, with a careful review of the following information: (1) the importance and extent of their involvement in IBTSM; (2) the purpose of the study, potential benefits and risks, and anticipated costs for their participation; and (3) the process and frequency

of data collection throughout the baseline and treatment phases. Caregivers also received a supplemental binder with information about each of the 16 IBTSM sessions, including session goals and homework assignments, with an additional section delineating the roles of all participating stakeholders. Parental consent was obtained at each caregiver-only treatment session. Additionally, the primary investigator was available throughout treatment to provide additional support. Study clinicians, but not caregivers, sought guidance from the primary investigator throughout the duration of the study.

Condensed IBTSM implementation. Study clinicians implemented a 16-session, condensed version of IBTSM, which took an average of 19 weeks (*Range* = 16-22 weeks) to complete from start to finish. Only one participant (Child 3) completed all 16 sessions in 16 consecutive weeks. All others took longer due to weather conditions, illnesses, winter breaks, or planned vacations. Regardless of the length of treatment, each family still received 16 full sessions of IBTSM, which maintained the same structure and key components of IBTSM in its original 20-session, 24-week format (Table 9). The 16-session format was selected due to the mixed, but promising, findings from the 12-session, 18-week version of IBTSM examined by Siroky and colleagues (2017). Sixteen weeks is a common length of psychosocial treatment for children (e.g., *Coping Cat;* Kendall & Hedtke, 2006) and represents the mean between 12 sessions (Siroky et al., 2017) and 20 sessions (Bergman et al., 2013). Sessions took place in the same community-based psychology clinic every week, lasting approximately 50 minutes each, with analog behavioral observations taking place for five minutes prior to the start of the session.

The first two sessions of IBTSM focused on building rapport between the child and the clinician, particularly in cases where the child struggles to speak with the clinician. The goal of

Table 9

Integrated Behavior Therapy for Selective Mutism Sessions, Original and Condensed Versions

IBTSM Sessions (24 weeks, 20 treatment sessions total)		**Condensed IBTSM Sessions (*M* = 19 weeks, 16 treatment sessions)**	
Pretreatment (Parent Only)	Assessment and Psychoeducation	*Pretreatment Intake Session*	Assessment and Psychoeducation
Session 1	Introduction, Rapport Building	*Session 1*	Introduction, Rapport Building
Session 2	Rapport Building, Reward System, Feelings Chart	*Session 2*	Rapport Building, Reward System, Feelings Chart
Session 3	Classroom Chart, Fear Ladder, Exposure Practice	*Session 3*	Classroom Chart, Fear Ladder, Exposure Practice
Sessions 4–9	Initial Exposure Sessions	*Sessions 4–7*	Initial Exposure Sessions
Session 10	Midpoint Session	*Session 8*	Midpoint Session
Sessions 11-14	Intermediate Exposure Sessions	*Sessions 9-10*	Intermediate Exposure Sessions
Session 15	Exposure, Introduction to Transfer of Control	*Session 11*	Exposure, Introduction to Transfer of Control
Sessions 16-17	Exposure, Additional Transfer of Control	*Sessions 12-13*	Exposure, Additional Transfer of Control
Sessions 18-19	Exposure, Transfer of Control, Progress Review	*Sessions 14-15*	Exposure, Transfer of Control, Progress Review
Session 20	Relapse Prevention and Graduation	*Session 16*	Relapse Prevention and Graduation

Session 1 was to help the child and his or her caregiver to become better oriented with IBTSM and the types of activities they would complete throughout. For example, the clinicians explained the reward system used during treatment and assigned the first homework assignment to be completed at home before Session 2. During Sessions 1 and 2, clinicians were prompted to attempt to elicit speech from each child as well. Session 3 was similar in that the primary goal was to increase the child's comfort in the clinic and to encourage more frequent speech with the clinician. This session also included a more detailed discussion about the fundamental behavioral tools used in IBTSM such as the feelings "thermometer," the fear hierarchy or "ladder" on which the child and caregiver ranked speaking situations from least to most fearful, and a classroom list to identify peers who may be helpful or familiar for future practices with communicating verbally in social situations.

Once both the child and his or her caregiver were sufficiently comfortable with these behavioral techniques, the primary focus of treatment shifted to in-session and out-of-session exposure assignments (Sessions 4-7). Together with the study clinician, the caregiver and child outlined specific settings, people, or situations that challenged the child sufficiently without causing excessive distress. Caregivers and their children worked together to select a reward or prize the child could earn upon successfully completing each assignment. Caregivers were encouraged to reward the child immediately after reaching their goal. For example, Child 5 tended to be more comfortable speaking to adults but continued to consistently avoid speech with classmates at school. To work toward this goal, Child 5's clinician and their caregivers planned out-of-session exposures involving planned communication with a familiar classmate in a comfortable setting like the child's home. Future steps involved speaking to this same classmate in a less formal setting at school and then eventually, practicing speaking to one or more classmates in their classroom. Specific details for each exposure assignment varied by participant, as these practices were developed based on each child's specific "fear ladder" and caregivers' ability to facilitate activities outside of the clinic.

The midpoint session (Session 8) served as an opportunity to check-in with caregivers and participants. Caregivers were given the chance to discuss any major obstacles encountered thus far in treatment. More specifically, study clinicians provided caregivers with guidance on how to continue to facilitate a relationship with their child's teacher. Caregivers were asked to request teacher feedback on any noticeable changes in the child's speech or levels of social anxiety at school. Clinicians were able to share and discuss data from the first half of treatment to celebrate any improvements seen in the dependent variables (e.g., caregiver-reported speaking behaviors, words spoken during observations, caregiver-reported social anxiety).

During the second half of treatment, caregivers began to take on greater responsibility in facilitating exposure tasks and selecting appropriate rewards for their child's positive behaviors. One of the key components of IBTSM is this "transfer of control" process, where adults start to mediate exposure assignments independently, beginning with Session 11. Bergman (2013) notes that, "Instead of the therapist leading the process, parents, with supervision from the therapist, will take the lead in the child's treatment. They will take the primary responsibility for devising and assigning speaking tasks, revising tasks (if necessary), setting goals, managing behavioral reward system, and the like" (p. 15). Using this process, caregivers were expected to be well-prepared to guide their child through additional exposure tasks if SM symptoms return after treatment ends. Each week, caregivers completed homework assignments pertaining to their child's "fear ladder." The use of the "fear leader" ensured that exposures during the second half of treatment were becoming progressively more difficult for the child and focused on helping them to generalize progress to the classroom, social situations with familiar peers, or spontaneous conversations with unfamiliar people in public. With greater control over treatment, caregivers were expected to devise appropriate assignments during therapy sessions with feedback from their child's clinician, as needed.

The final session (Session 16) focused primarily on follow-up discussions, celebrations, and relapse prevention. Study clinicians reviewed progress made by the child over the course of treatment, and children received a certificate of completion. As with the midpoint session (Session 8), any and all improvements since the start of treatment were celebrated. Clinicians allowed caregivers opportunities to ask questions or voice concerns about treatment ending. Finally, caregivers received additional education about how to continue facilitating exposures in public and at school. Each family was also offered the opportunity to continue working with

their child's clinician, although additional treatment sessions would require payment based on the clinic's typical session costs since funding for treatment sessions as a part of the present study covered one caregiver-only session and 16 follow-up treatment sessions.

Project personnel.

Study clinicians. Three doctoral-level students in school psychology, who were working as psychology interns at the participating clinic, served as clinicians in the present study to implement condensed IBTSM (i.e., 16 sessions) for five participants with exclusively-anxious SM. All clinicians received training on common behavioral techniques (e.g., shaping, fading, grade exposure, contingency management) typically used to treat children with SM from their direct clinical supervisor. This supervising psychologist has a PhD in clinical psychology and is an internationally-renowned expert on SM. An additional training session was held by the primary investigator/study coordinator to review IBTSM and its structure, to explain their roles and responsibilities with the project, and to model the ABO procedure. Clinicians received copies of a supplemental manual (separate copies for each participant), with additional guidance for how to carry out a condensed, 16-sesison IBTSM. This 16-session IBTSM manual was derived from the full IBTSM manual created by Bergman (2013). Clinician manuals included detailed explanations of each treatment session with example scripts from the original manual, as well as strategies for eliciting speech, templates for letters to teachers and school staff, and handouts to help guide treatment activities/assignments.

Project assistants. Two graduate-level students in school psychology were hired to assist with data collection and analysis for this study. The primary investigator met with both project assistants together at the beginning of data collection (i.e., prior to first baseline assessment) to provide direct instruction for observing and coding ABOs for words spoken, observing and

coding IBTSM sessions for adherence, and conducting visual analyses for multiple baseline designs. The primary investigator observed four, randomly-selected ABOs and IBTSM sessions via videotape to assess for inter-observer agreement. For the ABO tasks, clinicians and the second observer counted the number of words each child used to describe the picture. For treatment sessions, the primary investigator observed four randomly-selected sessions for each case (i.e., 25% of the total) to assess adherence. ABOs and IBTSM sessions were randomly selected before data collection began.

One of these two project assistants supported the data analysis portion of the study. To help orient this project assistant to single-case design and the steps for visual analysis, they were asked to carefully review the *What Works Clearinghouse* Single-case Research Technical Documentation Manual (Kratochwill et al., 2010) and to practice on graph templates prior to final data analysis. Following this practice, the project assistant received de-identified graphs with the study results and supplemental Visual Analysis Guides to conduct visual analysis once more. Inter-rater reliability was perfect between the primary investigator and project assistant ($\kappa = 1.00$). Project assistants received $10/hour of work they contributed to the project as a whole. Inter-rater reliability was adequate when analyzing final, de-identified graphs ($\kappa = 0.60$; 80% agreement), though both reviewers agreed perfectly about conclusions related to replicated effects across all five cases.

Study Phases

Study overview. At the pre-treatment intake session, caregivers received clear guidance on the goals and procedures of the present study. Caregiver responsibilities for IBTSM were fourfold: (1) attend and participate as needed in weekly sessions, (2) complete out-of-session homework assignments each week, (3) facilitate communication with their child's teacher, and

(4) complete questionnaires regularly regarding their child's speech and social anxiety levels. Participating families were also oriented to the general purpose of the study and any anticipated risks or benefits to their involvement. Baseline data collection began on the same say of the pre-treatment session and ended on the day of the child's first treatment session.

Recruitment and consent/assent procedures. Participants for the present study were recruited primarily through the participating community-based psychology clinic in mid-Michigan. The primary investigator used the Exclusion and Inclusion Clinician Sheet (Appendix L) to guide the recruitment process. Fliers approved by the MSU B-IRB were distributed throughout both clinic locations in mid-Michigan, as well as on the clinic's website and social media pages (Appendix C). Families who expressed interest in the present study received more information about the expectations for participation, including where weekly sessions would occur. Those who continued to be interested received an initial screening packet with the clinic's Behavioral Concerns Inventory, the SMQ, the SASC, and the SCARED to help the study investigator determine eligibility. Thirteen families in total expressed interest in participating in the study, with seven moving on to the screening process to determine eligibility. The six families who did not move forward with screening were not eligible due to distance (i.e., lived out-of-state or too far way to travel to the clinic each week) or initial exclusion criteria due to recent involvement in a week-long, intensive-dose treatment program for SM. Of the seven families who underwent the screening process, the first five eligible children were enrolled. One child was determined ineligible due to elevated SM severity, and one child met eligibility criteria but returned completed screening forms shortly after the fifth family had been enrolled.

Once each of the five children were enrolled in the study, caregivers were asked to schedule the first two sessions – the parent-only pre-treatment appointment and the first active

treatment session with their child – to ensure baseline length lasted no more than nine days. In addition to the pre-treatment session components as listed in the IBTSM manual, clinicians administered the comprehensive diagnostic interview to confirm SM diagnosis at this appointment. Caregivers received a baseline packet containing multiple copies of the BROSB and SASC-FNE at this parent-only session, which they completed daily until the first treatment session (Session 1). Parental consent (Appendix M) was obtained at the pre-treatment intake session as well. Prior to asking for consent, clinicians reviewed the consent form, which discussed the overall purpose of the present study, an overview of IBTSM, potential risks and benefits of involvement, as well as any anticipated costs to the family for participation. Caregivers were also made aware of their rights as participating families at this time. An additional child assent form (Appendix N) was administered to the child at the beginning of the first session, as the child was not present at the first parent-only pre-treatment session. Clinicians obtained verbal assent from the child but, given expected difficulties with eliciting speech at baseline, options for nonverbal assent (e.g., pointing, nodding) were provided as well.

Baseline phase. After the initial screening procedures and the first pre-treatment session, clinicians administered baseline assessment packets to participating caregivers. Given the practical limitations of enrollment through a community-based clinic, randomization to a baseline schedule was not used in this study. Instead, the first treatment session for IBTSM (i.e., Session 1) was scheduled with the family based on their availability and the clinician's availability, with options for the length of time between the pre-treatment session and the first treatment session. Baseline length options were controlled such that the minimum length was five days and the maximum length was nine days. Additionally, no two children were able to have the same baseline length. In turn, families enrolled later had fewer options to choose from.

This minimum of five data points per phase meets experimental single-case research design standards without reservation (Kratochwill et al., 2012). Caregivers completed the SASC-FNE and BROSB ratings daily starting on the day of the pre-treatment session and ending on the day of the first treatment session (i.e., Session 1).

Treatment phase. Following baseline data collection, all participants received 16 sessions of IBTSM, which maintained the same behavioral techniques, activities, goals, and procedures as IBTSM in its original format but was modified only in length (see Table 9 on page 71). Treatment sessions took place weekly in the community-based clinic. Due to unexpected circumstances (e.g., illness, weather condition) treatment length ranged from 16 weeks to 22 weeks (M = 19 weeks). In order to facilitate appropriate in-session exposure practices, two sessions for Child 5 (Sessions 11 and 12) occurred in a naturalistic setting (e.g., at a local restaurant to practice ordering, at a playground to practice interacting with peers). This child's clinician still completed session adherence checklists for those sessions. However, even when session practices occurred in other settings, all analog observations still took place before each treatment session and in the same setting (i.e., in the exact room in the clinic) to ensure environmental control.

Caregivers continued to complete questionnaires assessing their child's social anxiety pertaining to fear of negative evaluation (SASC-FNE) and their child's speech across settings (i.e., BROSB) weekly over the course of treatment. Caregivers completed the full scales for the SMQ, the SASC-R, and the SCARED once more at the end-of-treatment (Session 16) to capture anticipated global improvements as a result of this treatment approach. To further assess clinical significance of treatment gains after receiving IBTSM, the diagnostic interview was administered (i.e., ADIS-P) once more after each child's final treatment session.

Study Design

Through the use of multiple measures of treatment adherence, caregiver-reported social anxiety levels and speaking behaviors, as well as caregiver ratings of treatment acceptability, this study aimed to provide additional support for IBTSM as a suitable treatment for children with an exclusively-anxious presentation of SM, as proposed by early studies on different clinical presentations of SM (Darr et al., 2016; Cohan et al., 2008). Five children with a diagnosis of SM, who exhibited no other oppositional/defiant behaviors or speech delays at baseline, received a 16-session version of IBTSM as outlined by the manual via a nonconcurrent multiple baseline single-case design. The nature of this design reflected the plan to initiate data collection at varying time points since concurrent data collection in a community-based clinic would have been much too difficult to coordinate (Watson & Workman, 1981). No randomization was used in this nonconcurrent multiple baseline design due to expected limitations of randomly assigning baseline lengths to families and clinicians working in a community-based clinic setting. Rather, baseline lengths were determined primarily by the family and the clinician's availability, though the first author assisted in scheduling to ensure baseline phases differed for all cases and ranged from five to nine days in length. Once a baseline length had been selected by one family, this option was not available to future enrollees. In turn, families who were enrolled later had fewer options to choose from and the final family was assigned a specific baseline length after other baseline lengths had been chosen.

Caregivers completed daily questionnaires about their child's fear of negative evaluation (i.e., social anxiety) and observed speaking behaviors across contexts during the baseline phase. During the treatment phase, caregivers were only required to complete these questionnaires once per week. Behavioral observations took place before each treatment session to supplement

caregiver ratings of speaking behaviors. Treatment adherence was assessed using pre-developed checklists, to be completed after each session by study clinicians, and through observations (live or video) for four sessions (i.e., 25% of the total) per participant. At the end of treatment, caregivers rated the acceptability of the 16-session version of IBTSM in the areas of treatment feasibility, effectiveness, and time required.

Data Analysis

Research question one. Treatment adherence was analyzed by computing percentages of session components implemented by the study clinicians throughout IBTSM. A response of 0 (no attempt made) or 1 (unsuccessful attempt) for a given item on session adherence checklists was considered a lack of adherence for that component of IBTSM. A response of 2 (partially successful attempt) or 3 (successful attempt) represented adherence to the treatment task for a given component. At the end of each session, the total number of completed tasks (i.e., the number of items with a score of 2 or 3) was divided by the total number of required tasks and multiplied by 100 to yield a percentage of adherence for each session. At the end of treatment overall, adherence percentages from one pre-treatment session and 16 active treatment sessions were summed and averaged. Individual adherence percentages were generated for each of the five cases, as well as an aggregated average percentage across all participants. Perepletchikova and Kazdin (2005) note treatment implementation that includes 80% or more of the fundamental components is adequate.

Research questions two and three. In order to assess treatment effectiveness, the present study utilized a three-step analysis including (a) visual analyses, (b) effect size calculations, and (c) Reliable Change Index scores.

Visual analysis. First, visual analyses were used to determine the presence of noticeable

changes in each individual child's social anxiety levels and observed speaking behaviors over time. Six characteristics guide the visual analysis of between-phase (comparing baseline to treatment phases for each participant) or within-phase (examining intervention phase data across all cases) changes (Kratochwill et al., 2012). These include level, trend, variability, immediacy of effect, overlap of data between phases, and consistency of patterns within similar phases across multiple cases. With regard to the immediacy of effect, Kratochwill and colleagues (2010) suggest close examination of the last three baseline data points compared to the first three data points in the treatment phase. Given the varied frequency of assessment using the SASC-FNE and the BROSB across phases (i.e., three data baseline points represent three days, three treatment data points represent three weeks), immediacy of effect was evaluated using the first three weeks of treatment, compared to each case's entire baseline phase.

Minimal variability during baseline was expected, though SM symptoms depend highly on the environment and some spread may be seen within single cases. Due to the nature of SM, all five participants were expected to show consistently high social anxiety levels and consistently low ratings of speech across settings during the baseline phase. Although there is limited data on the immediacy of the effect of behavioral therapy for children with SM, one preliminary study (Siroky et al., 2017) found that one child who made significant gains by the end of treatment began to see some improvement within three sessions. Thus, observable treatment effects within the same time span were anticipated. Specifically, a visible decrease and downward trend in SASC-FNE scores was expected, since lower scores on this measure suggest lower levels of social anxiety. The opposite pattern was expected for ratings of participants' observed speaking behaviors such that, shortly after the implementation of IBTSM, an increase in the overall level (i.e., mean) and slope (i.e., trend) was expected. It was also hypothesized that

observable changes in speaking behaviors would be seen within the first three sessions.

Visual Analysis Guides (see Appendix O) were used to review three graphs for each case (i.e., 15 graphs in total) – one representing SASC-FNE scores as rated by caregivers, one depicting BROSB scores, and the third depicting frequency of words spoken during weekly ABOs. The Visual Analysis Guides included prompts to assess the level, trend, variability, and immediacy of effect for each case, as well as the consistency of patterns across all five cases. Due to the proposed use of effect size calculations, which take into consideration the amount of overlapping data between phases, the Visual Analysis Guides did not include a question about overlapping data. Visual comparison of these characteristics across all five cases was used to determine whether a replicated effect occurred. Generally, "an effect is demonstrated if manipulation of the independent variable is associated with predicted change in the pattern of the dependent variable (with temporal proximity between the two taken into account as well)." (Kratochwill et al., 2012; p. 31). Agreement between project assistants was adequate ($\kappa = 0.60$; 80% agreement). Disagreement only occurred when considering the immediacy of effect.

Effect size. The second step of data analysis proceeded to effect size calculation via Tau-U indices to assess the statistical significance of individual changes in outcome variables over time, and to assist in determining whether baseline trends were significant enough to require correction when examining these treatment effects. An online program was used to calculate Tau-U indices using both SASC-FNE and BROSB scores, for each individual child (http://www.singlecaseresearch.org/calculators/tau-u; Vannest, Parker, Gonen, & Adiguzel, 2016). Data from each child's baseline phase and each child's treatment phase were entered into separate columns (Child 1 baseline, Child 1 treatment, Child 2 baseline, Child 2 treatment, etc.). Using the online software, initial contrasts were computed for each child's baseline to their own

baseline to identify whether Phase A trend control was needed when computing contrasts for between each child's baseline data and their data from the treatment phase. Parker and colleagues (2011b) note that baseline trend control should only be used when "Phase A trend is pronounced and statistically significant" (p. 4). If a baseline-to-baseline contrast was considered statistically significant using a more liberal criterion for statistical significance ($p < .15$), as was recommended Bruni and colleagues (2017), contrasts between baseline and treatment phase data controlled for baseline trend.

Reliable change index. Supplemental Reliable Change Index (RCI; Jacobson & Truax, 1991) scores were used to assess clinical significance in observed changes on measures used at pre-treatment and end-of-treatment time points only (i.e., SASC-R, SMQ, and SCARED full scale scores). The RCI has been found to accurately detect clinical significance for a variety of measures (Gresham & Noell, 1993). To calculate the RCI, each case's baseline score was subtracted from their end-of-treatment score. This difference was then divided by the standard error of measurement for a particular measure. The standard error of measurement represents the standard deviation of the measure, multiplied by the square root of one minus the measure's reliability coefficient (α). An RCI of ±1.96 or higher is indicative of clinical significance (Jacobson & Truax, 1991).

$$\text{RCI} = \frac{X_B - X_A}{\text{SEM}} \qquad \text{SEM} = SD_{measure} \times \sqrt{(1 - \alpha_{measure})}$$

Cronbach's alpha reliability coefficients for both the SMQ (α = .76) and the SASC-R (α = .87) were derived from those reported in Bergman and colleagues' (2013) pilot study for IBTSM. Birmaher and colleagues (1991) reported a Cronbach's alpha of .90 for both the Child and Parent versions of the SCARED. Additionally, previous psychometric assessments for the SMQ, SASC-R, and SCARED measures (Behrens, Swetlitz Pine, & Pagliaccio, 2018; Bergman et al., 2008;

Letamendi et al., 2008), which included children with similar clinical presentations, were used to inform standard deviations for standard error of measurement calculations. RCI calculations were expected to indicate clinically significant reductions in social anxiety levels via SASC-R scores, along with clinically significant increases in speaking behaviors across contexts via SMQ total scores, for all five cases of children with SM.

Research question four. Finally, treatment satisfaction was assessed through caregiver ratings on the TEQ-P for each child. Caregivers completed the TEQ-P at the end of treatment (i.e., after Session 16). An overall score of 110 or higher was used as an indicator of adequate treatment acceptability (Kratochwill et al., 2003), as rated by caregivers of each of the five cases. The TEQ-P also contains three subscales related to treatment Effectiveness, Acceptability, and Time Required. Individual caregiver ratings of 55, 36, and 9 or higher were used to indicate adequate satisfaction across the Effectiveness, Acceptability, and Time Required subscales, respectively. Considering the emphasis on frequent communication between clinicians and caregivers, as well as the preliminary evidence to suggest IBTSM as an acceptable treatment (Bergman et al., 2013; Cotton-Thomas, 2015; Siroky et al., 2017) high levels of overall caregiver satisfaction were expected, with consistently high ratings across the three subscales.

CHAPTER 4

RESULTS

Research Question 1

Can a condensed (i.e., 16 sessions) version of IBTSM be carried out as intended by novice clinicians in a community-based clinical setting across all five cases?

Clinicians' self-report ratings of adherence to session components indicated high adherence when a condensed, 16-session version of IBTSM was implemented in a community-based clinical setting, as reflected in adherence ratings of 95% or higher across participants and an average adherence rating of 97% across all five children (Table 10). These ratings are well above 80%, which Perepletchikova and Kazdin (2005) indicate is an acceptable percentage for treatment adherence. When examining inter-observer agreement across all five cases, observer

Table 10

Treatment Adherence for Condensed IBTSM by Child

IBTSM Session	Child 1	Child 2	Child 3	Child 4	Child 5	Average
Pretreatment	86%	86%	100%	86%	100%	91%
1	100%	100%	100%	100%	100%	100%
2	100%	89%	100%	100%	100%	98%
3	81%*	88%	100%	100%	100%	94%
4	100%	100%	100%	100%	100%	100%
5	100%	100%	100%	100%	100%	100%
6	100%	100%	100%	90%*	100%	98%
7	100%	80%	100%	100%	100%	96%
8	100%	100%	100%	100%	100%	100%
9	80%	100%	100%	100%	100%	96%
10	100%	100%	100%	100%	100%	100%
11	100%	100%	100%	100%	100%	100%
12	100%	100%	100%	100%	100%	100%
13	100%	100%	100%	100%	83%	97%
14	100%	86%	100%	100%	100%	94%
15	100%	64%*	100%	100%	100%	93%
16	100%	100%	100%	100%	100%	100%
Average	**96%**	**93%**	**100%**	**99.5%**	**99%**	**97%**

*When inter-observer agreement was not perfect (100% agreement), the average between the two raters was used.

ratings were highly consistent with clinician self-report ratings of adherence to session components (94.2% agreement).

Research Question 2

Will a condensed version of IBTSM lead to a decrease in caregiver-rated social anxiety levels across the baseline phases and treatment phases for five children with exclusively-anxious SM?

Multiple baseline measures. Visual analysis for social anxiety levels, using the fear-of negative evaluation (FNE) subscale from the SASC-R, did not provide evidence of a replicated treatment effect for reduced social anxiety symptoms across all five children, primarily due to a lack of change in level (i.e., mean) and trend (i.e., slope) in the anticipated direction following the introduction of the intervention (Figure 2). Treatment effects were examined for individual participants using Tau-U effect size calculations. Using the cut-off of $p < .15$ as an indicator of statistical significance (Bruni et al., 2017), two children experienced a significant reduction in social anxiety symptoms over time (Child 1: *Tau-U* = -0.638, $p = 0.035$; Child 3: *Tau-U* = -0.875, $p = 0.001$). Caregiver ratings of social anxiety over the course of treatment for the remaining three participants were non-significant (Child 2: *Tau-U* = 0.00, $p = 1.00$; Child 4: *Tau-U* = 0.00, $p = 1.00$; Child 5: *Tau-U* = 0.00, $p = 1.00$), possibly due to the low ratings of social anxiety at baseline.

Baseline to end-of-treatment measures. Table 11 presents scores on measures of anxiety symptoms, including social anxiety (SASC-R) and features of other anxiety disorders (SCARED, from baseline to end-of treatment. Four of five (80%) children saw a significant reduction in social anxiety symptoms over time (Child 1: *RCI* = -7.99; Child 3: *RCI* = -11.82;

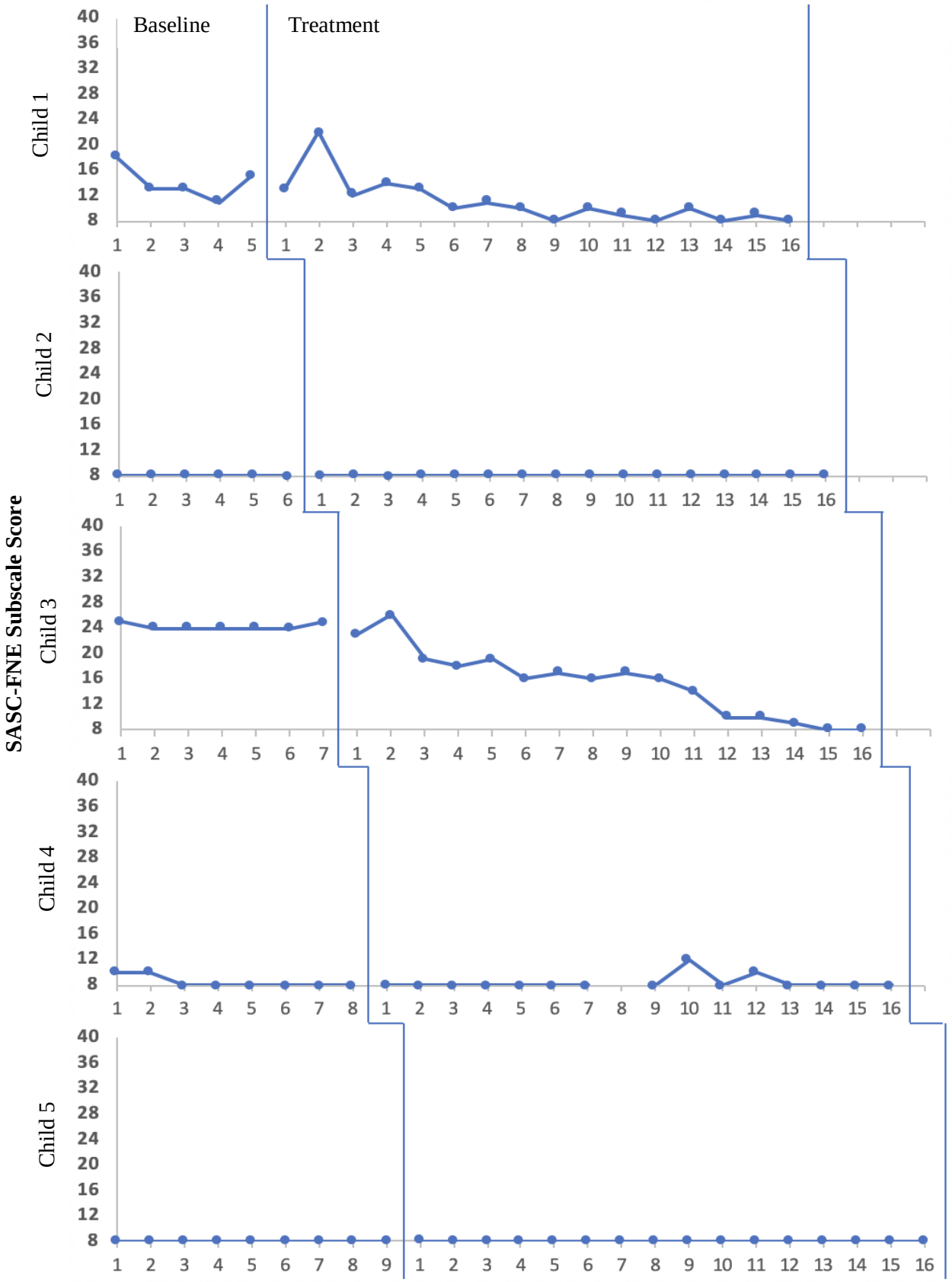


Figure 2. Caregiver-rated social anxiety levels over time

Table 11

Changes in Anxiety Symptoms and Diagnoses from Baseline to End-of-Treatment

Measure/*Child*	Baseline	End-of-Treatment	RCI
SASC-R Total Score			
Child 1	58	33	-7.99*
Child 2	56	51	-1.60
Child 3	64	27	-11.82*
Child 4	46	39	-2.24*
Child 5	67	35	-10.22*
Average (SD)	**58.20 (8.14)**	**37.00 (8.94)**	**-6.71***
SCARED Total Score			
Child 1	35	15	-4.29*
Child 2	27	19	-1.72
Child 3	40	17	-4.93*
Child 4	35	23	-2.57*
Child 5	26	17	-2.57*
Average (SD)	**32.60 (5.94)**	**17.60 (3.58)**	**-3.22***
ADIS-P - Social Phobia CSR			
Child 1	8	*n/a*	--
Child 2	5	6	--
Child 4	6	*n/a*	--
ADIS-P - GAD CSR			
Child 1	5	*n/a*	--
Child 4	7	*n/a*	--
ADIS-P – Separation Anxiety CSR			
Child 1	5	*n/a*	--
Child 4	*n/a*	4	--

*An RCI greater than 1.96 or less than -1.96 indicates a clinically significant change.

Child 4: *RCI* = -2.24; Child 5: *RCI* = -10.22). Supplemental assessments of child anxiety symptoms using the SCARED – Parent Form showed a similar pattern. When examining consistency of change across all five individuals, four children (80%) experienced a clinically significant reduction in overall symptoms of anxiety (Child 1: *RCI* = -4.29; Child 3: *RCI* = -4.93; Child 4: *RCI* = -2.57; Child 5: *RCI* = -2.57). Comparison of baseline to end-of-treatment ADIS-P CSRs indicated meaningful change in comorbid anxiety disorder diagnoses as well, despite having no treatment goals targeting generalized anxiety or separation anxiety symptoms. Of the three children who met criteria for Social Phobia at baseline, one child (Child 4) saw a removal

of diagnosis, according to ADIS-P criteria for clinical diagnosis. Both children who met criteria for Generalized Anxiety Disorder (GAD) at baseline (i.e., Child 1, Child 4), experienced a decrease in GAD symptoms to the point of removal of diagnosis. Similarly, Child 1 met criteria for Separation Anxiety Disorder at baseline, and they no longer presented with clinical levels of symptom severity by the end-of-treatment.

Research Question 3

Will a condensed version of IBTSM lead to improvements in observed speaking behaviors across settings between the baseline phases and treatment phases for five children with exclusively-anxious SM?

Multiple baseline measures. Based on visual analysis of mean item-level scores on the BROSB, no replicated intervention effect was observed in speaking behaviors across all five children, due to a lack of consistent visual changes in the level and slope of speaking behaviors in the hypothesized direction (Figure 3). A noticeable effect was observed for one individual child, Child 5, as indicated by comparison of the mean, slope, variability, and immediacy of change between baseline and treatment phases. Tau-U effect sizes were calculated to examine whether individual children experienced statistically significant improvements in speaking behaviors when comparing data from the treatment phase to their own data from the baseline phase. Only Child 2 and Child 5 experienced statistically significant (i.e., $p < .15$) increases in parent-rated speaking behaviors (Child 2: *Tau-U* = 0.677, $p = 0.017$; Child 5: *Tau-U* = 0.917, $p < .001$). All other Tau-U calculations were non-significant (Child 1: *Tau-U* = -0.050, $p = 0.869$; Child 3: *Tau-U* = -0.071, $p = 0.789$; Child 4 [corrected for baseline trend]: *Tau-U* = 0.172, $p = 0.564$). Similarly, simple visual analysis did not reflect noticeable, consistent improvement of words spoken during ABOs across the five participants (Figure 4). Rather, changes in words

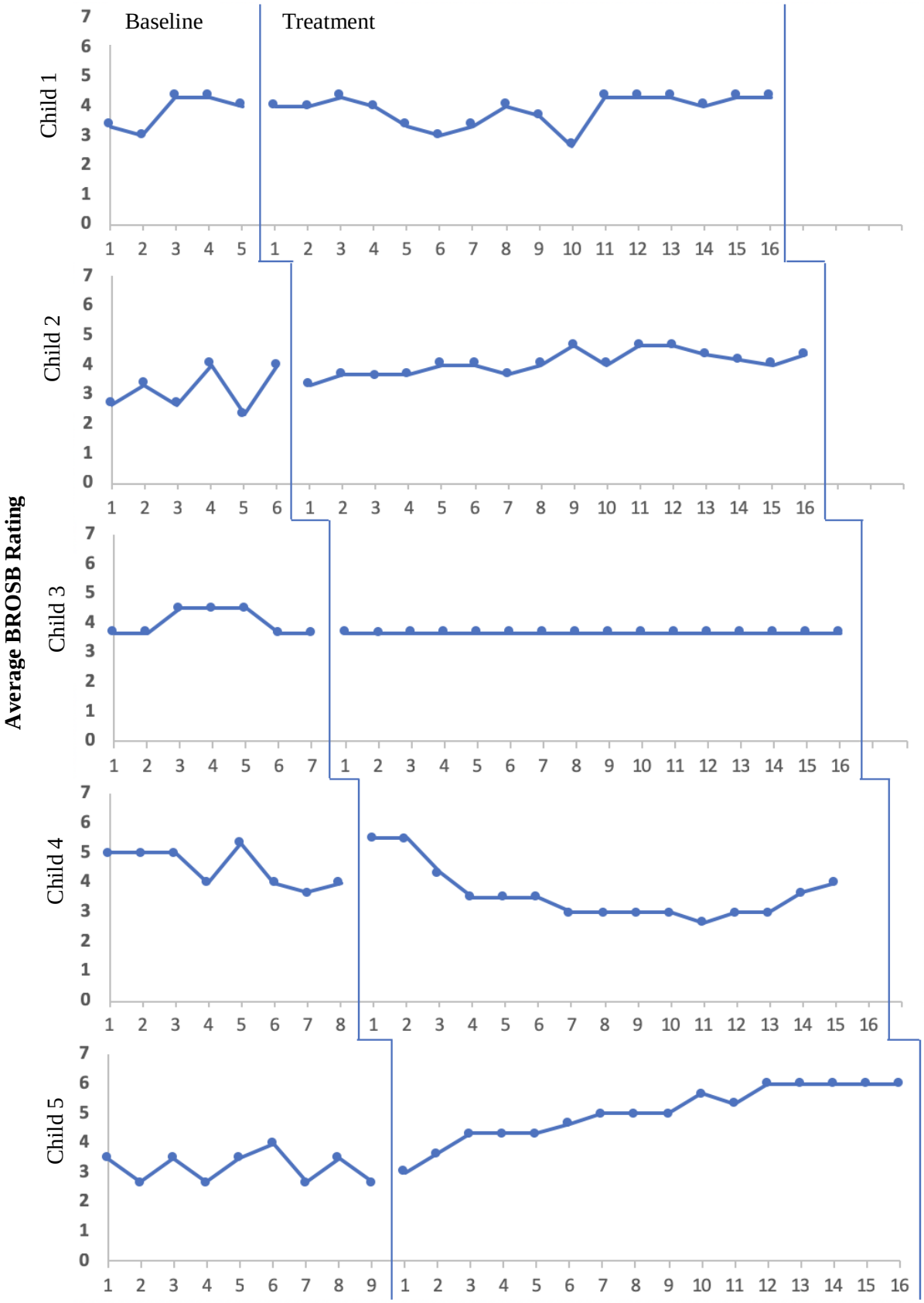


Figure 3. Average scores on Brief Ratings of Observed Speaking Behaviors over time

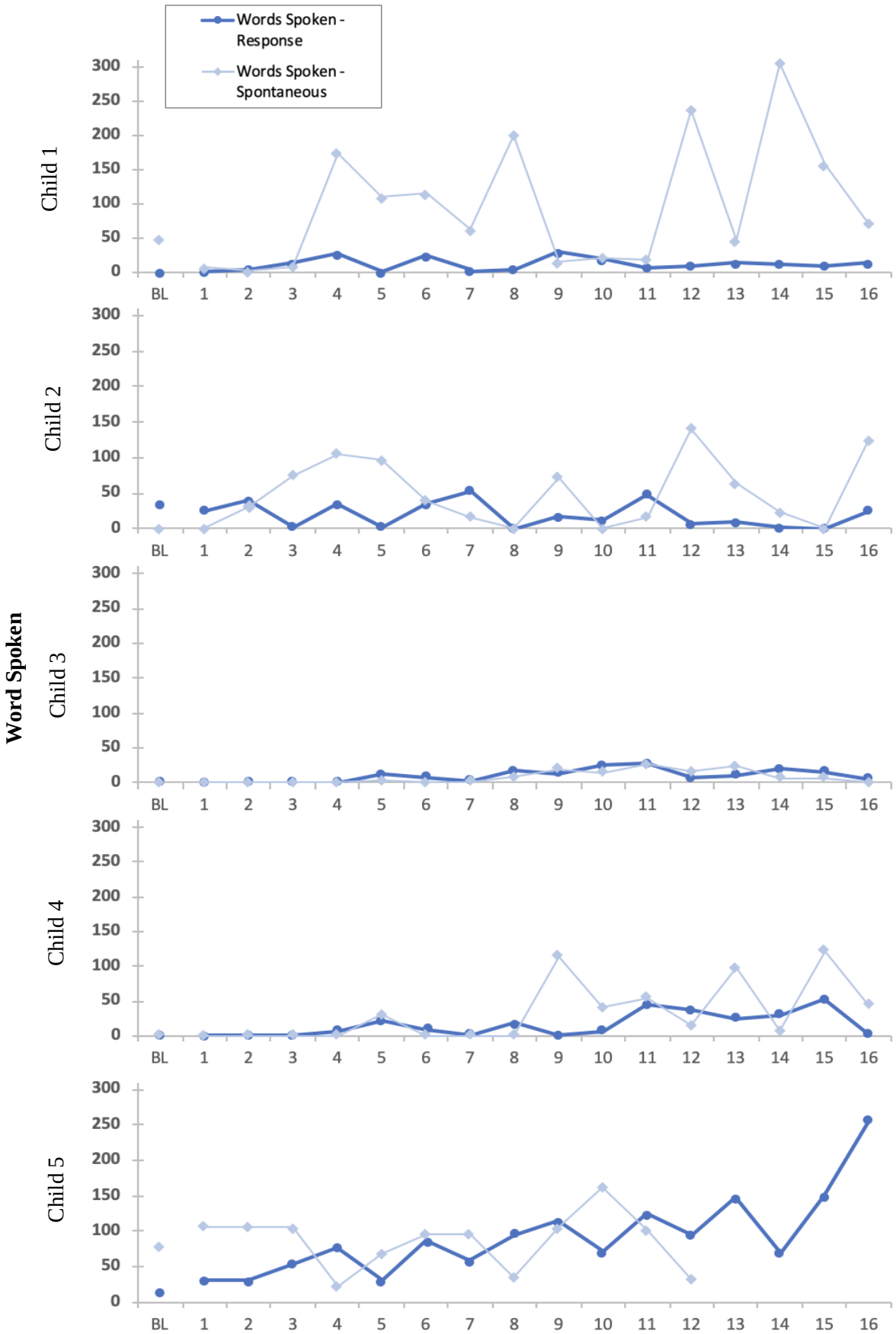


Figure 4. Words spoken over time during analog behavioral observations

spoken as measured by weekly ABOs were fairly sporadic with no clear change once the intervention was introduced.

Baseline to end-of-treatment measures. All five children saw an increase in SMQ scores from baseline to end-of-treatment, indicating an increase in caregiver-rated speaking behaviors over time. Three of five children (60%) were found to experience clinically meaningful increases in speech between baseline and end-of-treatment time points (Child 1: *RCI* = 5.42, Child 4: *RCI* = 4.34; Child 5: *RCI* = 6.23). At baseline, all five children met diagnostic criteria for SM as indicated by ADIS-P CSRs of four or higher (Table 12). By the end of treatment, three children (60%; Child 1, Child 4, Child 5) saw a reduction in the frequency, severity, and impairment associated with SM symptoms, resulting in a removal of diagnosis.

Table 12

Changes in Speaking Behaviors from Baseline to End-of-Treatment

Measure/*Child*	Baseline	End-of-Treatment	RCI
SMQ Total Score			
Child 1	16	36	5.42*
Child 2	13	18	1.36
Child 3	17	24	1.90
Child 4	16	32	4.34*
Child 5	15	38	6.23*
Average (SD)	**15.40 (1.52)**	**29.60 (8.41)**	**3.85***
ADIS-P - Selective Mutism CSR			
Child 1	8	*n/a*	--
Child 2	8	6	--
Child 3	4	4	--
Child 4	6	*n/a*	--
Child 5	7	*n/a*	--

*An RCI greater than 1.96 or less than -1.96 indicates a clinically significant change.

Research Question 4

Will caregivers rate the condensed (i.e., 16-session) version of IBTSM as an acceptable treatment approach for children with exclusively-anxious SM, specifically in the areas of treatment quality, effectiveness, and time required?

A majority of caregivers (*n* = 4; 80%) perceived the condensed version of IBTSM as an acceptable treatment approach overall. All five (100%) caregivers endorsed adequate treatment quality, and all (100%) viewed the condensed version of IBTSM as acceptable with regard to the time required. Caregivers of three participants (60%) perceived treatment to be adequately effective for their child. Scores provided by each caregiver can be found in Table 13.

Table 13

Caregiver-rated Treatment Acceptability for Condensed IBTSM

Participant	Overall Acceptability	Quality	Effectiveness	Time Required
Child 1	114*	66*	32	11*
Child 2	94	56*	25	11*
Child 3	121*	65*	40*	10*
Child 4	118*	61*	40*	11*
Child 5^	118*	63*	37*	12*
Parent 1	119*	63*	38*	12*
Parent 2	117*	63*	36*	12*
Average (*n* = 6)	**114***	**63***	**35***	**11***

*Indicates ratings at or above the cut-off for adequate acceptability.
^Both parents of Child 5 completed separate forms. Scores for Child 5 reflect average total and subscale scores from both parents.

Individual Improvements

Although results did not provide evidence of a replicated intervention effect across all five cases, caregivers perceived the condensed version of IBTSM to be acceptable. This result may be due to the improvements and changes seen by individual children. Child 1, Child 4, and Child 5 saw meaningful improvements in speaking behaviors, as well as significant reductions in social anxiety symptoms, to the extent that none of these children met diagnostic criteria for SM at the end-of-treatment time point. Additionally, Child 1 and Child 5 both experienced a decrease in symptoms of other types of anxiety (e.g., separation anxiety, generalized anxiety) such that they no longer met diagnostic criteria for these comorbid disorders. However, Child 4's

caregiver endorsed clinically elevated symptoms of separation anxiety at the end of the treatment, despite the lack of diagnosis at the baseline timepoint.

Although visual analysis of Child 4's speaking behaviors indicated an overall reduction in the frequency and consistency of their speech across settings, scores from baseline to end-of-treatment indicated significant increases. An important feature of Child 4's clinical presentation was their tendency to use an altered voice, observed to be speech at a much higher pitch than their typical voice, when verbally communicating with unfamiliar peers and adults or when speaking to their caregivers in a novel setting (e.g., the participating clinic). Initial goals for treatment focused on establishing any speech with the clinician in-session; however, after speech had been established and maintained, which occurred around Session 3, individual goals in- and out-of-session focused on differentially reinforcing this child's use of their typical voice rather than their altered voice. This may explain the noticeable decrease in Child 4's mean BROSB ratings around Session 3.

Interestingly, Child 2 and Child 3 were both identified by their distinct clinicians as demonstrating persistent oppositional and defiant tendencies, which were not captured during the intake assessment process. Specifically, both Child 2 and Child 3 were observed to be non-compliant with parent commands, even when commands did not require a verbal response. Child 2 experienced some improvement in SM and anxiety symptoms, though these changes were just below clinical significance. Child 3 did experience a significant reduction in social anxiety levels, though their increased speech across settings fell short of clinical significance as well.

CHAPTER 5

DISCUSSION

Purpose of Study

Selective mutism is a relatively rare anxiety disorder defined by symptoms of persistent speech avoidance in certain social situations, even though fluent and comfortable speech is observed in other settings (APA, 2013). SM typically affects young children, with initial signs first seen before age five (Ford et al., 1998). If not treated effectively, children with persistent SM are more likely to experience significant impairment in their social development, may underperform academically, and may be at-risk for later anxiety or mood disorders (Pionek Stone, 2002; Steinhausen & Juzi, 1996). Thus, early intervention is essential for children with SM to promote their long-term social-emotional, behavioral, and academic well-being.

Since SM and social anxiety are highly comorbid, treatment for SM typically adapts evidence-based approaches developed for youth with social anxiety disorder (Muris & Ollendick, 2015; Pionek Stone et al., 2002; Sharp, Sherman, & Gross, 2007). To date, the most common and effective form of treatment for children with SM is behavioral or cognitive behavioral therapy (Muris & Ollendick, 2015; Viana, Beidel, & Rabian, 2009). Although a number of studies have supported the effectiveness of various adaptations of behavioral or cognitive behavioral therapy for SM (Zakszeski & DuPaul, 2016), no manualized treatment has garnered sufficient research support to be considered an EBT, as defined by Chorpita and colleagues (2011), meant specifically for children with SM. Sheridan (2014) clearly outlines such a progression via a ten-step trajectory for intervention research. This study sought to add to previous literature on IBTSM which has defined a need for SM treatment in children (Step 1), described strategies which can effectively improve functioning (Step 2), and piloted the feasibility and efficacy of

IBTSM via a randomized-controlled study (Step 3; Berman et al., 2013). Steps 4 and 5 of Sheridan's (2014) trajectory address precise investigations of treatment effectiveness when implemented in smaller samples and in various settings. Scholars exploring characteristics of samples of children with SM suggest the presence of at least three subtypes, for which distinct treatment approaches may be particularly beneficial (Cohan et al., 2008; Darr et al., 2016; Diliberto & Kearney, 2016; Mulligan et al., 2015; Skedgell et al., 2017). In an effort to better examine for whom IBTSM might be effective, this study enrolled children with the proposed exclusively-anxious subtype.

The present study sought to contribute to this growing body of research on EBTs for children with SM, using a nonconcurrent multiple baseline single-case design to examine the adherence, effectiveness, and acceptability of the only manualized treatment for children with SM, *Integrated Behavior Therapy for Selective Mutism* (IBTSM; Bergman, 2013). This study builds upon previous investigations of IBTSM, which provide some preliminary evidence for its efficacy (Bergman et al., 2013) and its potential effectiveness in reducing speech avoidance and social anxiety levels in children with SM in real-world contexts using varying treatment lengths (Cotton-Thomas, 2015; Khan & Renk, 2018; Siroky et al., 2017). Given limitations of previous studies (e.g., lack of sufficient baseline data, unclear assessment of intervention adherence, comorbid behavioral or speech/language deficits), a more rigorous single-case investigation was warranted in order to more confidently draw conclusions about treatment effectiveness in specific populations of children with SM (i.e., children with an exclusively-anxious subtype). To address these limitations, the present study was carefully designed to meet standards for single-case research without reservation, as determined by the *What Works Clearinghouse* Single-case Design Technical Documentation Manual (Kratochwill et al., 2012).

Five children between the ages of four and eight with an exclusively-anxious presentation of SM received a 16-session version of IBTSM across an average of 19 weeks (*Range* = 16-22 weeks) in a community-based clinical setting. Novice clinicians implemented this condensed version of IBTSM with excellent adherence (i.e., average = 98%). Incremental assessment of social anxiety levels and speaking behaviors over the course of treatment indicated that each child experienced some individual improvement; however, visual analyses did not result in a replicated treatment effect across all five cases. Despite this lack of replicated effect using visual analyses, a majority of participants experienced statistically significant and clinically reliable reductions in SM symptoms, as demonstrated by Tau-U effect size calculations and RCI scores. More notably, three of five children no longer met diagnostic criteria for SM by the end of treatment and two children (40%) also saw a removal of comorbid anxiety disorder diagnosis by the same time point. Finally, caregivers rated this condensed version of IBTSM as acceptable overall. All five caregivers (100%) perceived IBTSM as acceptable with regard to treatment quality and time required. A detailed discussion of these results within the context of previous research is provided below, followed by implications for future research and practice.

Treatment Adherence

As anticipated, study clinicians implemented the condensed, 16-session version of IBTSM with excellent adherence (ratings of 93% or higher) across all five cases. Only one session (Child 2, Session 15: *M* = 64%) throughout the course of treatment across all five cases fell below what Perepletchikova and Kazdin (2005) note as adequate adherence (i.e., 80%). This adherence rating of 64% was derived from the average between the clinician's self-report rating of session adherence (100%) and the second observer's rating of this following video review (29%). According to the second observer's rating, the clinician only completed two of seven

components listed for Session 15: (1) review general events of the past week and (2) develop, explain, and execute exposures according to individual plan.

Treatment adherence, defined as the extent to which a clinician implements treatment as it is intended (Chambless & Hollon, 2012), is an essential factor in intervention research. Perepletchikova and Kazdin (2005) emphasize the importance of measuring treatment adherence in this line of research, particularly when treatment is replicated, in order to draw valid conclusions about changes in dependent variables following the introduction of the independent variable (i.e., intervention). Given the present study's design to critically examine treatment effectiveness and acceptability across multiple children, adherence was pertinent for answering all research questions. Adherence was measured through clinician self-report ratings using session checklists, supported by high inter-observer agreement (94.2%). This uniformly high treatment adherence seen across all five cases is consistent with previous investigations of IBTSM, which found similarly high ratings of treatment adherence when carried out in a research-based clinical setting (99.3%, Bergman et al., 2013) and a community-based clinical setting (96.7%, Siroky et al., 2017).

The excellent adherence in this study also reflects one of the benefits of manualized treatment approaches given the increased likelihood that clinicians will adhere to treatment components when a manual or guide is available (Addis, Wade, & Hatgis, 1999; Perepletchikova & Kazdin, 2005). An additional factor likely affecting study clinicians' strong adherence to the IBTSM protocol was the weekly supervision each clinician received, which was provided by a licensed psychologist who is a known expert in treatment for children with SM. Consistent supervision and opportunities for discussion about specific treatment elements is a critical element in training programs which adopt and adequately implement EBTs (McHugh & Barlow,

2010). The supervision also provided study clinicians with frequent opportunities to problem-solve potential barriers to treatment effectiveness, supported their use of behavioral strategies used to elicit speech from children, and facilitated the "transfer of control" process to better share skills with caregivers. This process is similar in nature to the supervision offered to clinicians in the randomized-controlled pilot study of IBTSM (Bergman et al., 2013). Bergman and colleagues (2013) explained that clinicians providing treatment to participating children received weekly supervision in a group format (Bergman et al., 2013).

Finally, the structure of IBTSM itself allows for a great deal of flexibility with regard to specific activities to support overall treatment goals. Kendall and colleagues (2008) provide an in-depth discussion about the benefits of maintaining this "flexibility within fidelity" approach when carrying out manualized EBTs, including the increased likelihood that treatment is carried out as intended. Although all IBTSM treatment components remained consistent across all five children, specific activities and in-session exposures varied for each child depending on his or her individual goals. For instance, Child 2 required multiple in-session exposures to simply increase the consistency of their speaking behaviors with his clinician, while Child 5 was able to use in-session exposures to verbally communicate with novel people, in novel settings. Therefore, this flexibility with which clinicians could tailor treatment activities while successfully implementing session components likely contributed to the excellent adherence observed in this study.

Social Anxiety Symptoms

Recent reviews of the SM literature highlight a consistent link between SM and social anxiety, including similar epidemiological characteristics (Ford et al., 1998), high rates of comorbidity (Gensthaler et al., 2016; Kristensen, 2000; Manassis et al., 2007; Yeganeh et al.,

2003; Yeganeh et al., 2006), and similarities in treatment approaches (Sharp, Sherman, & Gross, 2007). Thus, social anxiety symptoms were a primary dependent variable in this study and were closely examined over time using multiple measures. Contrary to the hypothesized outcome, there was no evidence of a replicated treatment effect on social anxiety levels across all five of the participating children in this study following visual analysis. However, all five children experienced reductions in social anxiety symptoms when examining changes in SASC-R full scale scores between baseline and end-of-treatment time points. Four of five (80%) children saw clinically reliable reductions over time as indicated by RCI calculations and two of the three children who met diagnostic criteria for social phobia at baseline saw a removal of diagnosis following 16 sessions of IBTSM over the course of 21 (Child 1) and 19 weeks (Child 4).

These changes in social anxiety symptoms are consistent with previous investigations of IBTSM. In Siroky and colleagues' (2017) replicated AB single-case design examining the effectiveness of a 12-session version of IBTSM in a community-based clinic, both children (n = 2; 100%) experienced significant and reliable reductions in social anxiety symptoms by the three-month follow-up (Child 1: *RCI* = -6.07, Child 2: *RCI* = -3.20). Additionally, Bergman and colleagues (2013) reported comparable results in the RCT piloting IBTSM in a clinical research setting. After receiving 20 sessions of IBTSM over the course of 24 weeks, group-level means on the SASC-R indicated that children in the active treatment group (n = 12) experienced a significant reduction in social anxiety symptoms by the end of treatment. Although the average number of comorbid diagnoses in the active treatment group decreased from baseline (M = 2.38) to end-of-treatment (M = 2.25), this change was not significant.

The present study's incremental assessment of both social anxiety symptoms and speaking behaviors over the course of treatment also allows some discussion on the theorized

mechanism of action used to describe the onset and maintenance of SM symptoms. Given the proposed link between SM and social anxiety, the behavioral conceptualization of SM proposes that elevated symptoms of anxiety may cause or contribute to a child's speech avoidance (Bergman, 2013; Kotrba, 2015). Following this logic, it would be expected that children in the present study would need to experience a reduction in anxiety symptoms in order to experience a similar reduction in SM symptoms (i.e., speech avoidance). When examining within-subject changes in multiple baseline measures of social anxiety (i.e., SASC-FNE subscale) and speaking behaviors (i.e., BROSB), there was no evidence of a pattern consistent with this proposed mechanism of action (i.e., reductions in social anxiety levels did not consistently precede increased speaking behaviors), even when accounting for an expected lag between reduced social anxiety symptoms and increased speech. However, a review of baseline to end-of-treatment scores for each child may align with this proposed conceptualization. Each of the three children who no longer met criteria for SM by the end of treatment also saw clinically reliable changes in symptoms of social anxiety and other anxiety symptoms (i.e., SCARED total scores). Additionally, Child 3 also experienced clinically meaningful reductions in social anxiety symptoms and other symptoms of anxiety, though their SM symptom improvement was just shy of clinical significance (SMQ: *RCI* = 1.90). Therefore, no child in the present study experienced reductions in speech avoidance without also experiencing significant reductions in social anxiety.

Speaking Behaviors

Since speech avoidance is the most distinctive and impairing symptom of SM in children (APA, 2013), speaking behaviors were another primary dependent variable, as measured by caregiver ratings of speaking behaviors (i.e., BROSB) and words spoken during weekly analog behavioral observations. Visual analyses of these two measures did not support the hypothesis

that all five children would experience a significant reduction in speech avoidance when comparing baseline speaking behaviors to changes in speaking behaviors over the course of condensed (i.e., 16 sessions) IBTSM. When examining changes in SMQ total scores from baseline to end-of-treatment, however, it is clear all five children experienced an increase in speaking behaviors over time. Three children (60%) improved to the point of clinical significance (i.e., RCI scores greater than 1.96), and also no longer met diagnostic criteria for SM by the end of treatment (i.e., after 16 sessions over an average of 19 weeks). This remission rate is comparable to the response Bergman and colleagues (2013) found in the randomized-controlled pilot study of IBTSM where 67% of children in the treatment group saw a removal of diagnosis after 20 sessions of IBTSM over 24 weeks. Previous single-case investigations of an 8-session, school-based version of IBTSM (Cotton-Thomas, 2015) and a 12-session, community-based clinic version of IBTSM (Siroky et al., 2017) did not result in consistent, significant treatment effects based on parent- and teacher-reported speaking behaviors. Taken into context with these previous findings, results from the current study suggest that a 16-session version of IBTSM, when implemented in a community-based clinic over the course of 16-22 weeks (*M* = 19 weeks), may have been a sufficient treatment length to consistently yield meaningful improvements in speaking behaviors over time in this sample of five children with an exclusively-anxious presentation of SM.

An important potential reason for a lack of replicated treatment effects is the possible, unintentional inclusion of children presenting with oppositional behaviors, in addition to their SM diagnoses. The rationale for identifying children of an exclusively-anxious subtype of SM follows from previous work on SM classifications, and calls from SM scholars to better understand whether varying presentations require different treatment approaches (Cohan et al.,

2008; Darr et al., 2016; Diliberto & Kearney, 2016; Mulligan et al., 2015; Skedgell et al., 2017). Although all children in the present study were screened for elevated symptoms of defiance or oppositional behaviors prior to enrollment, two children (Child 2, Child 3) were observed to resist compliance to parent commands throughout in-session and out-of-session exposure practices. Child 2 and Child 3 were the only two participants who did not experience statistically or clinically significant improvements in speaking behaviors over the course of treatment, though Child 3 did see a significant reduction in social anxiety levels by the end of treatment. As was hypothesized in an earlier single-case investigation of a 12-week version of IBTSM (Siroky et al., 2017), it is possible that children with mild oppositional behaviors are less likely to experience clinically significant improvements in speaking behaviors over time in response to IBTSM alone. Children who present with elevated defiant behaviors may require adaptations to typical treatment for SM, such as an initial focus on positive reinforcement for compliance prior to targeted exposures to reinforce speech across settings (see Skedgell et al., 2017).

Treatment Acceptability

Caregivers uniformly perceived the condensed, 16-session version of IBTSM as an acceptable treatment approach. These results are in line with previous investigations of IBTSM when implemented in various settings (e.g., research-based clinic, community-based clinic, school) and with modifications to treatment length. In the pilot study for IBTSM, both parents and teachers involved in the care of participating children endorsed high levels of satisfaction with the treatment approach (Bergman et al., 2013). An earlier single-case investigation with two children also indicated high levels of parent and teacher acceptability across both cases, even though treatment effects were only significant for one child (Siroky et al., 2017). Additionally,

Cotton-Thomas (2015) found that school professionals viewed IBTSM as acceptable and feasible for children with SM when carried out in a school setting.

A number of factors may have influenced caregivers' perceptions of IBTSM as an acceptable treatment approach. First, all five children saw some positive change in one or more outcome variables (i.e., social anxiety, speaking behaviors) over time. When comparing baseline to end-of-treatment scores on the SASC-R, SMQ, and SCARED, all children saw a change in the intended direction (i.e., increase in SMQ scores, decrease in SASC-R and SCARED scores) which were either clinically significant or near clinical significance. Each caregiver denied adverse effects to them or their child due to participating in this study. Thus, the presence of positive outcomes and absence of negative outcomes undoubtedly contributes to caregivers' perception treatment as acceptable.

Next, caregivers were made aware of the overall goals and plans for treatment during the pre-treatment intake session. Caregivers and participants were highly involved in selecting appropriate goals for out-of-session exposure activities and homework assignments, a process referred to as "transfer of control" by Bergman (2013). According to Perepletchikova and Kazdin (2005), when caregivers or other stakeholders are aware of the rationale, mechanisms, and goals for treatment, they are more likely to perceive treatment as more acceptable. Thus, the orientation to the condensed version of IBTSM and involvement in learning strategies clinicians used throughout (e.g., psychoeducation, exposure practices, contingency management) may have influenced caregivers' overall perception of IBTSM.

Finally, all five families (100%) decided to continue receiving some form of treatment (e.g., weekly therapy appointments, monthly therapy appointments, school-based consultation, community-based exposures) through the participating clinic, which may serve as an additional,

qualitative indicator of treatment acceptability. For instance, although Child 2's caregivers provided ratings below the cut-off of adequate treatment acceptability, they elected to continue receiving services with the same clinician with a focus on school-based exposures and consultation as this is the area in which Child 2 demonstrated particular impairment.

Comorbid Anxiety and Behavioral Concerns

An unexpected but critical finding in this study was the presence of relevant comorbid issues (e.g., other anxiety disorders, oppositional behaviors) and heterogeneity amongst the study sample, even after targeted efforts to identify and enroll children with a more homogenous clinical presentation of exclusively-anxious SM. Within this sample of five children, three (60%) met criteria for one or more additional anxiety disorders beyond SM at baseline (Child 1: social phobia, GAD, separation anxiety; Child 2: social phobia; Child 4: social phobia, GAD). Individual changes in total and subscale scores on the SMQ, SASC-R, and SCARED over time can be found in supplemental Tables 14-16 (Appendix P).

Although there is no research to date specifically examining how comorbid anxiety disorders affect treatment outcomes for children with SM receiving IBTSM, studies exploring predictors of treatment outcomes for children receiving CBT for anxiety suggest that child demographic variables at baseline (e.g., increased age, comorbid social anxiety, and greater symptom severity) may be associated with poorer treatment outcomes (Hudson et al., 2015; Oerbeck et al., 2014; Wergeland et al., 2016). Contrary to those previous findings, however, both Child 1 and Child 4 saw a removal of all baseline anxiety disorder diagnoses, including SM, by the end of treatment. Unexpectedly, by the end of treatment, Child 4 met criteria for separation anxiety disorder despite a lack of this diagnosis at baseline. Although specific details related to the onset of this child's separation anxiety symptoms were not assessed, it is possible that this

child presented with mild separation anxiety at baseline but, due to their very limited speech, did not express worry or concern about being away from their caregiver to others when separated.

Additional consideration should be given to the emergence of oppositional behaviors in at least two participants in the present study over the course of treatment. A variety of assessments were used to identify and enroll children with an exclusively-anxious subtype of SM, as outlined by Darr and colleagues (2016), including screening for characteristics indicative of other proposed SM subtypes (i.e., oppositional-anxious, speech/language concerns). Even though all five children met criteria for the exclusively-anxious subtype of SM at baseline, both Child 2 and Child 3 were identified by clinicians as presenting with mildly oppositional behaviors throughout treatment. One possible explanation for this change in clinical presentation, despite a demonstrated lack of oppositional behaviors in these two children at baseline, may reflect the changing patterns of parent responses to their child's avoidance strategies when given the expectation to speak. The presence of oppositional behaviors in children with SM is not uncommon (Ford et al., 2008; Keeton, 2013; Yeganeh et al., 2006), though more recent reviews of the SM literature propose that oppositional behaviors may be a result of the child's feelings of anxiety or distress when expected to speak, rather than primary cause for a child's lack of speech (Muris & Ollendick, 2015). Reference to the behavioral conceptualization of SM may help to explain this phenomenon as well (Bergman, 2013). Given the child's noticeable anxiety when in situations where they are expected to speak, it is common for parents or caregivers to negatively reinforce avoidance through "rescuing" (i.e., speaking on behalf the child) or removing the expectation to speak altogether (Kotrba, 2015). In turn, parents may also develop a pattern of assisting their child in avoiding having to speak. Over the course of IBTSM, these caregiver response patterns are continually corrected by clinicians to further reinforce the child's speech,

rather than speech avoidance. Thus, as parent responses change, some children may be more resistant to comply with expectations which they have previously avoided, appearing as oppositional or defiant behaviors given their desire to avoid speaking (Bubier & Drabick, 2009).

Limitations

The following limitations are important to consider in the context of the overall findings of this study: (a) reliability and validity of weekly measures (i.e., BROSB, FNE); (b) lack of empirically-derived assessment to determine SM subtype; (c) non-diverse sample of children; and (d) inconsistent measurement intervals across baseline and treatment phases.

Reliability and validity of multiple baseline measures. A critical consideration when reflecting on the lack of replicated treatment effects across all five cases is the selection of multiple baseline measures to capture changes in dependent variables over time. Since caregivers were asked to provide ratings of social anxiety levels and speaking behaviors each week, there was a need to identify brief measures which caregivers could feasibly complete without compromising validity and reliability. Unfortunately, the selection and analysis of the brief measures used in the present study may have lacked validity, affecting the ability to accurately capture and analyze changes in dependent variables over time.

The BROSB was a Direct Behavior Rating (DBR) developed specifically for this study, which was derived from the SMQ (Bergman et al., 2008) as a brief way for caregivers to indicate the type of speech their child demonstrates at home, in school, and in other social situations. Although DBRs are supported as useful formative assessment tools sensitive to incremental change over time (Christ, Riley-Tillman, & Chafouleas, 2009; Volpe & Briesch, 2012), there have been no previous studies utilizing DBRs for speaking behaviors in children with SM. Thus, it is possible that the BROSB as a tool was insufficient in capturing changes in individual's

speaking behaviors from week to week. For instance, during summer and winter breaks, caregivers were unable to rate their child's speaking behaviors in the school domain, which resulted in decreased total scores on the BROSB. As a result, average item-level scores were used to better assess children's speech across settings over the course of treatment. The use of means, rather than total scores, yielded a smaller range within which to improvements could be seen (i.e., 1-7 compared to 1-21). Additionally, although the BROSB was developed to capture increases in complexity (length of response, length of spontaneous speech) and frequency of speaking behaviors, this measure did not account for whether children were using altered voices or whispers. In turn, two children who provided a verbal response consistently, but who did not spontaneously initiate speech in school, may both have received a score of 5 on the school item, though one child may have been speaking in a whisper while the other child may have been speaking in their full voice. Such a distinction would represent differing severity levels of SM symptoms, though this difference would not be reflected in an examination of the two children's BROSB scores.

Similarly, a proposed reason for a lack of change on the multiple baseline measure of social anxiety (i.e., SASC-FNE subscale) is the unexpectedly low levels even at baseline, leaving minimal room for improvement over time. Using the screening procedures, all five children presented with clinically elevated social anxiety symptoms at baseline via SASC-R total scores. However, when caregivers provided daily ratings on the FNE subscale during the baseline phase, at least three children (Child 2, Child 4, Child 5) presented with minimal scores, leaving little room for improvement or observed change over the course of treatment. The FNE subscale was selected as the multiple baseline assessment due to its high level of reliability ($\alpha = .86$; La Greca

& Stone, 1993), yet results from this investigation put into question the overall face validity of this subscale for use with children with SM.

SM subtype assessment procedures. Another potential limitation of the present study involves the procedure for identifying clinical subtypes of SM. The choice to screen for children with an exclusively-anxious subtype of SM was based on Siroky and colleagues' (2017) findings, which suggested IBTSM may be specifically beneficial to children with that clinical presentation, rather than children with preexisting externalizing behaviors or speech/language concerns. Although preliminary research suggests the presence of at least three subtypes of SM (Cohan et al., 2008; Darr et al., 2016; Diliberto & Kearney, 2016; Mulligan et al., 2015), there are currently no empirically-based assessments for identifying SM subtypes. The procedure used in the present study followed the work of Darr and colleagues (2016), and all children met the inclusion and exclusion criteria meant to specifically recruit and enroll children with a proposed exclusively-anxious subtype of SM; however, at least two of the five children (40%) were observed to be more oppositional in nature than the others, often engaging in non-compliant behaviors even during clinic sessions. In turn, conclusions about the effectiveness of IBTSM for this specific subtype of children with SM are limited by unexpected changes in clinical presentations over the course of treatment in some, but not all, participants.

Non-diverse study sample. Although this study was not intended to generalize results to a larger population of youth, this study is still quite limited by the homogeneity of the sample. Four of five participants (80%) identified as White/Caucasian, with one child (Child 1) identifying as Mixed/Biracial (20%). Additionally, all families involved in this study had access to the time, transportation, and flexibility in their schedule to be able to attend and engage in 16 sessions of IBTSM, resources which may not be readily available to all families in need of

behavioral therapy services for children with SM. It is unclear whether similar results would be found if implemented in another geographic location or if provided to families with more diverse cultural or socioeconomic backgrounds.

Inconsistent measurement intervals across phases. A final potential limitation of this study is the different intervals between data points for the baseline phase (one day), as compared to the treatment phase (one week). The baseline and treatment phases were designed in this way to reduce the amount of time children had to wait prior to the start of treatment, and to support the feasibility of data collection for participating caregivers. Although such a change in intervals between phases is not explicitly a concern identified by the single-case research design standards (Kratochwill et al., 2012), this inconsistency may threaten the internal validity of the design and should be taken into consideration when interpreting results.

Implications for Research

Given a number of significant findings related to the adherence, effectiveness, and acceptability of a 16-session version of IBTSM when implemented in a community-based clinic, results from this study build upon previous investigations of IBTSM to better determine for whom and under which circumstances this treatment can be effective. The present study sought to address aspects of Steps 4 and 5 in Sheridan's (2014) trajectory for intervention research (2014) by implementing a condensed version (i.e., 16 sessions over 16-22 weeks) of IBTSM with precision in a small sample of children with a specific presentation of SM (i.e., exclusively-anxious subtype). Previous research on IBTSM reflect initial steps along this trajectory, though additional research is needed before IBTSM could be considered an EBT for this population using rigorous criteria (Chorpita et al., 2011). Additional research exploring IBTSM's effectiveness is needed to see if significant treatment effects can be replicated in new settings or

with more diverse samples of children with SM. Considerations for future investigations of IBTSM or other SM treatment approaches are provided below.

First, an important feature of IBTSM is its emphasis on caregiver involvement throughout treatment, with increased responsibility as treatment progresses (i.e., "transfer of control"). It is unclear, however, the extent to which caregiver participation and perceptions contribute to IBTSM's effectiveness. Even with this emphasis on caregiver skill training, the present study did not monitor the extent to which caregivers attempted and/or completed out-of-session exposure practices in other settings beyond the participating clinic. Studies examining the effectiveness of CBT for children with anxiety reveal mixed findings regarding the association between homework compliance and treatment outcomes (Hughes & Kendall, 2007; Mausbach, Moore, Roesch, Cardenas, & Patterson, 2010), yet consistent exposure practice outside of the clinic may be particularly important for children with SM so improvements can be generalized from the clinic setting to other contexts or situations where speaking is expected (e.g., novel settings, novel people, increased speaking demands). Future investigations of IBTSM should consider more closely examining the effect of caregiver involvement on treatment outcomes, including caregivers' understanding of treatment processes/goals, engagement during treatment sessions, and adherence to out-of-session assignments.

Additionally, there is a clear need for the identification and clarification of possible subtypes of SM. Multiple efforts (e.g., developmental history checklist, diagnostic interview with parents) were made during the initial recruitment phase to identify children with an exclusively-anxious subtype of SM, since it was hypothesized that other subtypes may show less improvement through the condensed version of IBTSM. However, based on clinician report and observation, at least two children were identified as having potential oppositional/defiant

tendencies over the course of treatment. In turn, it is unclear which subtype would best capture the clinical presentation of these two children given the emergence of more externalizing behaviors over time. Efforts should be made through future research to develop a reliable and valid assessment for SM subtypes, with implications for treatment modifications to address distinct needs for those children with SM and their families.

Finally, future studies would likely be strengthened by a more detailed examination of speaking behaviors in children with SM, including speech complexity, speech across settings, and possible safety behaviors specific to SM (e.g., speaking in a whisper or altered voice). It is well known that children with SM often experience more severe speech avoidance in the school setting as compared to other settings (Black & Uhde, 1995; Dummit et al., 1997). Previous investigations of IBTSM when implemented in a clinical research (Bergman et al., 2013) and a community-based clinic setting (Siroky et al., 2017) assessed treatment outcomes as rated by both parents and teachers. Unfortunately, it was beyond the scope of the present study to closely examine teacher-rated speaking behaviors. In turn, future investigations studying the effectiveness of a 16-session version of IBTSM should consider supplementing caregiver-rating speaking behaviors with teacher ratings to better assess whether comparable gains would be seen across settings using the same length of treatment. Safety behaviors were informally assessed in this study via additional questions on the SMQ; however, a comparison of ratings between baseline and end-of-treatment time points were not suggestive of a noticeable or meaningful change in safety behaviors across these five cases.

Since the completion of data collection for the present investigation, a few studies have begun to address this need for more detailed assessment of SM symptoms. For instance, Gensthaler and colleagues (2018) recently described a novel parent rating scale created

specifically for children with SM, titled the *Frankfurt Scale of Selective Mutism* (FSSM). The FSSM includes two scales including a diagnostic scale, which is used to assess the presence of SM symptoms, as well as a severity scale to assess individual speaking behaviors, patterns, and the severity of speech avoidance. Xu and colleagues (2018) also recently reviewed two pilot studies using passive audio vocal measurement (AVM) to provide more objective data regarding speaking behaviors over time. The use of AVM was found to be feasible and sensitive in capturing variation in vocalizations, which may help to provide greater detail about individual characteristics of each child's pattern of communication.

Implications for Practice

The results of this study may also be used to inform clinical practice when treating children with SM. One finding from this investigation points to the potential feasibility of maintaining high treatment adherence while carrying out IBTSM in a community-based clinic setting. Three novice clinicians were able implement a condensed IBTSM (i.e., 16 sessions over an average of 19 weeks) with high levels of adherence to the session components as outlined in the manual. This adherence was also maintained across all five cases of children with SM, each presenting with varied goals related to speaking behaviors with peers, at school, or in other social situations in public. In the context of intervention research, such a finding is pertinent in order to more clearly examine the effectiveness of a treatment approach when carried out in novel settings. The strong adherence found in the present study, along with similar findings from previous studies on IBTSM (Cotton-Thomas, 2015; Khan & Renk, 2018; Siroky et al., 2017), suggests that IBTSM may be feasibly implemented by novice clinicians in clinical research, school, and community-based clinic settings, though there is a notable need for additional research with targeted to specifically address the remaining steps along Sheridan's (2014)

intervention research trajectory (i.e., develop a theory, test in larger samples, assess mechanisms of change, explore situational or contextual variables affecting treatment effects, and test effectiveness of the developed treatment on a larger scale).

Finally, results from this study provide some support for the presence of multiple subtypes of SM and the need for different treatment approaches to specific address clinical features of each subtype. All five participants presented with symptoms of the proposed exclusively-anxious SM subtype at the baseline time point; however, two children (Child 2 and Child 3) displayed oppositional/defiant behaviors over the course of the active treatment phase. Although it may be challenging to predict which children will see an onset of oppositional behaviors once treatment begins, clinicians treating children with SM would do well to monitor early signs of defiance or oppositional behaviors. This step may be critical in developing an effective and efficient treatment plan for individual children and to determine whether adaptations to IBTSM may be needed.

APPENDICES

APPENDIX A

Situation Rating Form

from IBTSM manual (Bergman, 2013; p. 121)

Instructions: Describe specific situations and how difficult they are. Some of these situations should NOT include speaking and should be very easy situations so that this task is not overwhelming. Others should be speaking situations.

EASY:

Situation: _____

Situation: _____

Situation: _____

MEDIUM:

Situation: _____

Situation: _____

Situation: _____

HARD:

Situation: _____

Situation: _____

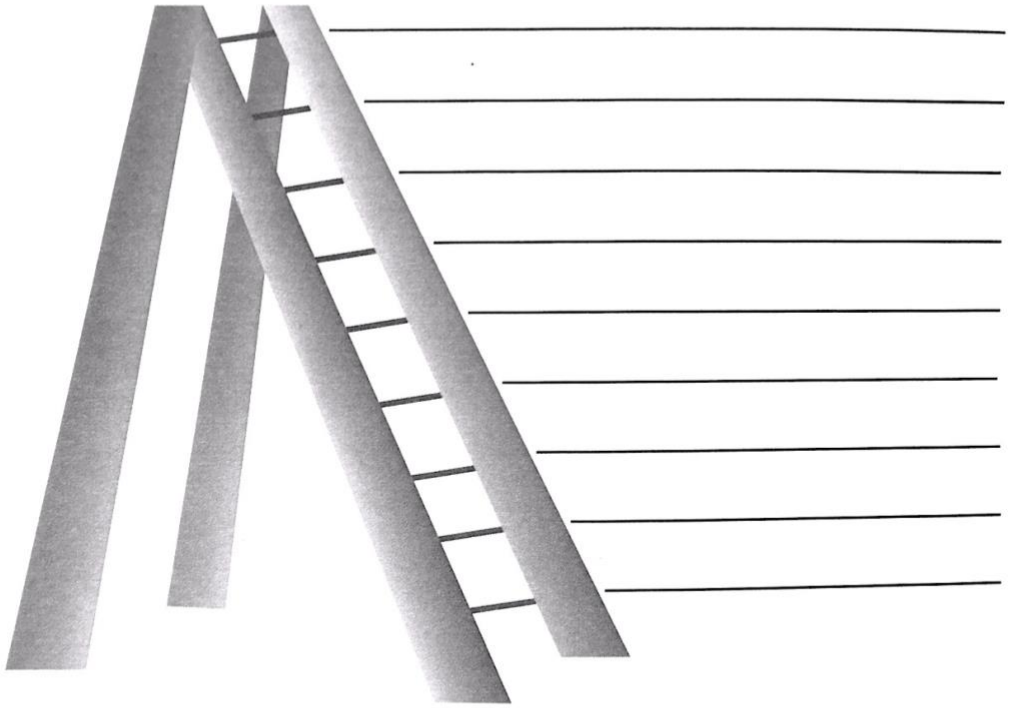
Situation: _____

APPENDIX B

Talking Ladder

from IBTSM Manual (Bergman, 2013; p. 122)

Instructions: List situations to work on, with the easiest situations at the bottom of the ladder and the hardest situations at the top.



APPENDIX C

Recruitment Flier

"INTEGRATED BEHAVIOR THERAPY FOR EXCLUSIVELY-ANXIOUS SELECTIVE MUTISM: A NONCONCURRENT MULTIPLE BASELINE DESIGN ACROSS FIVE PARTICIPANTS"

FAMILIES NEEDED

The Department of Counseling, Educational Psychology, and Special Education at Michigan State University is looking for five young children and their families to participate in a study investigating the effectiveness and acceptability of **Integrated Behavior Therapy for Selective Mutism** (IBTSM).

IBTSM is an innovative approach to SM treatment that encourages children to overcome their fear of speaking through weekly therapy sessions and speaking exercises to be completed at home, in the community, or at school. Caregivers play a key role in IBTSM as they will be facilitating a majority of these exercises.

If you are the caregiver of a child who:

- *is between the ages of 4 and 8*
- *presents with symptoms of selective mutism (SM) and anxiety when expected to speak to certain people*
- *does not have a history of speech/language delays or oppositional behaviors*
- *is not receiving other psychosocial or medication treatment for these symptoms*

your family may be eligible.

The study will consist of weekly one-hour sessions over the course of 16 weeks. Treatment sessions will be conducted by a licensed graduate student intern who has been trained in behavioral therapy for treating SM. All fees have been waived for participating families and insurance will not be billed. Families will be compensated $200 for their involvement, for some travel fees, and for rewards to support therapy.

To express interest or receive additional information, please contact the study coordinator: Allison Siroky at sirokyal@msu.edu

APPENDIX D

IBTSM Session Outlines and Adherence Checklists

***Pretreatment Session*: Caregiver and Therapist Only**

Materials Needed

- SMQ for Caregiver
- SSQ for Teacher
- Weekly Homework Form
- Two Treatment Letters (*for school principal and/or teacher*)
- Educational Handouts (*optional*)
- Home Assignment Binder (*for caregivers*)
- Teacher Binder (with assessments)

To what extent did were the following tasks completed? Please select one response per task.	***No attempt was made***	***Attempted but not successful***	***Attempted and partially successful***	***Successful***
1. Obtain and review detailed assessments of speaking behaviors (SMQ)				
2. Obtain and review detailed assessment of social anxiety symptoms.				
3. Provide information about phenomenology of SM				
4. Describe and explain cycle of avoidance				
5. Review treatment program and address any questions or concerns				
6. Explain and assign homework (weekly measures to be completed before next session)				
7. Obtain information regarding speaking behavior and social anxiety symptoms from teacher (may occur after session is complete)				
TOTAL COMPLETED = _____ + _____				
Pretreatment Session Adherence = _____ / 7 TASKS **(above sum)**				

Session 1: **Introduction and Rapport Building**

Materials Needed

- Props for rapport building activities
- Prize Brainstorming Form
- Exposure Assignment Form
- Weekly Homework Form
- Home Assignment Binder (*caregiver should bring*)

To what extent did were the following tasks completed? Please select one response per task.	***No attempt was made***	***Attempted but not successful***	***Attempted and partially successful***	***Successful***
1. Welcome child to treatment				
2. Increase the child's comfort speaking in therapy room (e.g., show them around, leave them alone to play in the room).				
3. Introduce goals and rationale for treatment (in language the child can understand).				
4. Introduce use of the reward system				
5. Develop rapport with the child through play activities (e.g., crafts, play games, puzzles).				
6. Assign homework				
TOTAL COMPLETED = _____ + _____				
Session 1 Adherence = _____ / 6 TASKS **(above sum)**				

Session 2: **Rapport Building, Reward System, Feelings Chart**

Materials Needed

- Completed Prize Brainstorming Chart
- Reward Chart
- Feelings Chart
- Situation Ratings Form
- Talking Ladder
- Playdate Form
- Classmate List
- Weekly Homework Form
- Home Assignment Binder (*caregiver should bring*)

To what extent did were the following tasks completed? Please select one response per task.	***No attempt was made***	***Attempted but not successful***	***Attempted and partially successful***	***Successful***
1. Review general events of the past week				
2. Review homework assignment				
3. Develop reward system				
4. Introduce and practice using the Feelings Chart				
5. Increase child's comfort speaking to caregiver(s) in therapy room, if necessary				
6. Continue developing rapport				
7. Introduce Talking Ladder				
8. Begin discussing child's interactions with peers				
9. Assign homework				
TOTAL COMPLETED = _____ + _____				
Session 2 Adherence = _____ / 9 TASKS **(above sum)**				

Session 3: **Class Chart, Talking Ladder, Exposure Practice**

Materials Needed

- Completed Situation Ratings Form
- Talking Ladder
- Feelings Chart
- Completed Classmate List
- Class Chart
- Other Individuals List
- Playdate Form
- Exposure Assignment
- Weekly Homework Form
- Home Assignment Binder (*caregiver should bring*)

To what extent did were the following tasks completed? Please select one response per task.	***No attempt was made***	***Attempted but not successful***	***Attempted and partially successful***	***Successful***
1. Review general events of the past week				
2. Review homework assignment				
3. Develop chart using list of peers				
4. Discuss and construct Talking Ladder (hierarchy) using the completed Situation Rating Form and Feelings Chart				
5. Practice exposure in session				
6. Discuss exposure exercises				
7. Plan for out-of-session exposure				
8. Assign homework				
TOTAL COMPLETED = _____ + _____				
Session 3 Adherence = _____ / 8 TASKS **(above sum)**				

Sessions 4, 5, 6, and 7: **Initial Exposure Sessions**

Materials Needed

- Talking Ladder
- Feelings Chart
- Copies of returned Exposure Assignment Forms (*or other relevant information*)
- Class Chart and/or Other Individuals Chart
- Playdate Form
- Exposure Assignment Form
- Weekly Homework Form
- Home Assignment Binder (*caregiver should bring*)

To what extent did were the following tasks completed? Please select one response per task.	***No attempt was made***	***Attempted but not successful***	***Attempted and partially successful***	***Successful***
1. Review general events of the past week				
2. Review homework assignment				
3. Develop, explain, and execute in-session behavioral exposures according to individual plan				
4. Devise and discuss relevant out-of-session activities				
5. Assign homework				
TOTAL COMPLETED = _____ + _____				
Session Adherence = _____ / 5 TASKS **(above sum)**				

Session 8: **Treatment Midpoint Session**

Materials Needed

- Talking Ladder
- Feelings Chart
- Copies of returned Exposure Assignment Forms (*or other relevant information*)
- Class Chart and/or Other Individuals Chart
- Playdate Form
- Exposure Assignment Form
- Weekly Homework Form
- Home Assignment Binder (*caregiver should bring*)

To what extent did were the following tasks completed? Please select one response per task.	***No attempt was made***	***Attempted but not successful***	***Attempted and partially successful***	***Successful***
1. Review general events of the past week				
2. Review homework assignment				
3. Review progress to date, problem-solve obstacles to progress (if needed)				
4. Devise and discuss relevant out-of-session activities				
5. Assign homework				
TOTAL COMPLETED = _____ + _____				
Session 8 Adherence = _____ / 5 TASKS **(above sum)**				

Sessions 9 and 10: **Intermediate Exposure Sessions**

Materials Needed

- Talking Ladder
- Feelings Chart
- Copies of returned Exposure Assignment Forms (*or other relevant information*)
- Class Chart and/or Other Individuals Chart
- Playdate Form
- Exposure Assignment Form
- Weekly Homework Form
- Home Assignment Binder (*caregiver should bring*)

To what extent did were the following tasks completed? Please select one response per task.	***No attempt was made***	***Attempted but not successful***	***Attempted and partially successful***	***Successful***
1. Review general events of the past week				
2. Review homework assignment				
3. Develop, explain, and execute in-session behavioral exposures according to individual plan				
4. Devise and discuss relevant out-of-session activities				
5. Assign homework				
TOTAL COMPLETED = _____ + _____				
Session Adherence = _____ / 5 TASKS **(above sum)**				

Session 11: **Continued Exposure Sessions and Introduction to Transfer of Control**

Materials Needed

- Talking Ladder
- Feelings Chart
- Copies of returned Exposure Assignment Forms (*or other relevant information*)
- Class Chart and/or Other Individuals Chart
- Playdate Form
- Exposure Assignment Form
- Weekly Homework Form
- Home Assignment Binder (*caregiver should bring*)

To what extent did were the following tasks completed? Please select one response per task.	***No attempt was made***	***Attempted but not successful***	***Attempted and partially successful***	***Successful***
1. Review general events of the past week				
2. Review homework assignment				
3. Develop, explain, and execute in-session behavioral exposures according to individual plan				
4. Devise and discuss relevant out-of-session activities				
5. Begin transfer of control process with explanation of concept				
6. Assign homework				
TOTAL COMPLETED = _____ + _____				
Session 11 Adherence = _____ / 6 TASKS **(above sum)**				

Sessions 12 and 13: **Continued Exposure Sessions with Additional Focus on Transfer of Control**

Materials Needed

- Exposure Ideas Form
- Talking Ladder
- Feelings Chart
- Copies of returned Exposure Assignment Forms (*or other relevant information*)
- Class Chart and/or Other Individuals Chart
- Playdate Form
- Exposure Assignment Form
- Weekly Homework Form
- Discretionary sticker (for caregivers' use)
- Home Assignment Binder (*caregiver should bring*)

To what extent did were the following tasks completed? Please select one response per task.	***No attempt was made***	***Attempted but not successful***	***Attempted and partially successful***	***Successful***
1. Review general events of the past week				
2. Review homework assignment				
3. Develop, explain, and execute in-session behavioral exposures according to individual plan				
4. Facilitate transfer of control while shaping out-of-session plans for behavioral exposures for the coming week				
5. Assign caregivers to award "discretionary sticker"				
6. Assign homework				
TOTAL COMPLETED = _____ + _____				
Session Adherence = _____ / 6 TASKS **(above sum)**				

Sessions 14 and 15: **Continued Exposure Sessions and Transfer of Control Review/Progress**
Materials Needed

- Talking Ladder
- Feelings Chart
- Copies of returned Exposure Assignment Forms (*or other relevant information*)
- Class Chart and/or Other Individuals Chart
- Playdate Form
- Exposure Assignment Form
- Weekly Homework Form
- Discretionary sticker (for caregivers' use)
- Home Assignment Binder (*caregiver should bring*)

To what extent did were the following tasks completed? Please select one response per task.	***No attempt was made***	***Attempted but not successful***	***Attempted and partially successful***	***Successful***
1. Review general events of the past week				
2. Review homework assignment				
3. Develop, explain, and execute in-session behavioral exposures according to individual plan				
4. Review progress and remaining goals for treatment				
5. Continue facilitation of transfer of control and planning behavioral exposure for coming week				
6. Address transfer of control as it pertains to the child				
7. Assign homework				
TOTAL COMPLETED = _____ + _____				
Session Adherence = _____ / 7 TASKS **(above sum)**				

Session 16: **Relapse Prevention and Graduation**

Materials Needed

- Copies of returned Exposure Assignment Forms (*or other relevant information*)
- Post-treatment SMQ for Caregivers
- Progress Chart
- Remaining Goals Worksheet
- Certificate of Achievement
- Home Assignment Binder (*caregiver should bring)*

To what extent did were the following tasks completed? Please select one response per task.	***No attempt was made***	***Attempted but not successful***	***Attempted and partially successful***	***Successful***
1. Review general events of the past week				
2. Review homework assignment				
3. Review progress and present graphic representation of improvement				
4. Continue discussion of remaining goals				
5. Discuss future speaking challenges and relapse prevention				
6. Have graduation fun, including presentation of diploma!				
TOTAL COMPLETED = _____ + _____				
Session 16 Adherence = _____ / 6 TASKS **(above sum)**				

Overall Treatment Adherence

	Cumulative Complete	***Cumulative Total***
Pretreatment Session Adherence		**7**
Session 1 Adherence		**13**
Session 2 Adherence		**22**
Session 3 Adherence		**30**
Session 4 Adherence		**35**
Session 5 Adherence		**40**
Session 6 Adherence		**45**
Session 7 Adherence		**50**
Session 8 Adherence		**55**
Session 9 Adherence		**60**
Session 10 Adherence		**65**
Session 11 Adherence		**71**
Session 12 Adherence		**77**
Session 13 Adherence		**83**
Session 14 Adherence		**90**
Session 15 Adherence		**97**
Session 16 Adherence		**103**

TOTAL COMPLETED = _____ / 103

Rate of Adherence = _____

Goal: TOTAL SCORE OF 82

APPENDIX E

Social Anxiety Scale for Children - Revised (SASC-R), FNE Subscale

Please answer each of the items regarding YOUR CHILD'S feelings and behavior. There are no right or wrong answers. Please answer as honestly as you can.

Use the numbers to show HOW MUCH each statement IS TRUE FOR YOUR CHILD:

1 = Not at all **2 = Hardly ever** **3 = Sometimes**
4 = Most of the time **5 = All the time**

Now let's try these sentences first. How much do they describe how your child feels?

a. My child likes summer vacation… 1 2 3 4 5
b. My child likes to eat spinach… 1 2 3 4 5

		Not at all	*Hardly Ever*	*Sometimes*	*Most of the time*	*All the time*
1.	My child worries about being teased	**1**	**2**	**3**	**4**	**5**
2.	My child feels that other children talk behind his/her back	**1**	**2**	**3**	**4**	**5**
3.	My child worries about what other children think of him/her	**1**	**2**	**3**	**4**	**5**
4.	My child is afraid that others will not like him/her	**1**	**2**	**3**	**4**	**5**
5.	My child worries about what others say about him/her	**1**	**2**	**3**	**4**	**5**
6.	My child worries that other children don't like him/her	**1**	**2**	**3**	**4**	**5**
7.	My child feels that other children make fun of him/her	**1**	**2**	**3**	**4**	**5**
8.	If my child gets into an argument with another youngster, he/she worries that the other youngster will not like him/her	**1**	**2**	**3**	**4**	**5**

APPENDIX F

Brief Rating of Observed Speaking Behaviors (BROSB)

1. Please mark (X) one box that best describes your child's speaking behavior at school (e.g., with his/her teacher, peers, or other school personnel).

☐	☐	☐	☐	☐	☐	☐	☐
0 Does not communicate (neither verbal/ nonverbal)	**1** Infrequently communicates nonverbally.	**2** Frequently communicates nonverbally. (but no speech)	**3** Responds verbally, but does so infrequently.	**4** Frequently responds verbally.	**5** Consistently responds verbally, but does not spontaneously initiate speech	**6** Spontaneously initiates verbal speech, but does so infrequently.	**7** Frequently and spontaneously initiates verbal speech.

2. Please mark (X) one box that best describes your child's speaking behavior at home (e.g., with his/her family, babysitter, or close family friends).

☐	☐	☐	☐	☐	☐	☐	☐
0 Does not communicate (neither verbal/ nonverbal)	**1** Infrequently communicates nonverbally.	**2** Frequently communicates nonverbally. (but no speech)	**3** Responds verbally, but does so infrequently.	**4** Frequently responds verbally.	**5** Consistently responds verbally, but does not spontaneously initiate speech	**6** Spontaneously initiates verbal speech, but does so infrequently.	**7** Frequently and spontaneously initiates verbal speech.

3. Please mark (X) one box that best describes your child's speaking behavior in public or other social situations (e.g., with unfamiliar children or adults, with his/her doctor or dentist, with store clerks).

☐	☐	☐	☐	☐	☐	☐	☐
0 Does not communicate (neither verbal/ nonverbal)	**1** Infrequently communicates nonverbally.	**2** Frequently communicates nonverbally. (but no speech)	**3** Responds verbally, but does so infrequently.	**4** Frequently responds verbally.	**5** Consistently responds verbally, but does not spontaneously initiate speech	**6** Spontaneously initiates verbal speech, but does so infrequently.	**7** Frequently and spontaneously initiates verbal speech.

Week # _____

APPENDIX G

Analog Behavioral Observation Script

1. Open book to picture. Point to page and say, **"Tell me what you see in this picture."**
2. Begin timer. Tally each word spoken in the box below.
3. After 30 seconds, if child has not spoken say, **"There is no right or wrong answer, just tell me what you see."**
4. Stop timer after 5 minutes (300 seconds).
5. Praise the child for any words spoken (e.g., **"Wow, that was a great description! Thank you for telling me what was in the picture."**).
6. Count aloud the number of tally marks. Say, **"I counted ___ words today. That means you have earned ___ stickers. Way to use your brave voice!"**
7. Give child as many stickers as words were spoken. Proceed to IBTSM session.

Please note the number of words the child speaks in response to the prompt given by his/her caregiver.	
Make tally marks here.	**Total Words Spoken**
	_____
Please note the number of instances the child <u>spontaneously initiated speech</u> (e.g., spoke verbally in the absence of a prompt to speak) with his/her caregiver or the clinician.	
Make tally marks here.	**Total Instances of Spontaneous Speech**
	_____
Date _____/ _____/ _____	Session #_____

Appendix H

Social Anxiety Scale for Children - Revised (SASC-R), Parent Form

Please answer each of the items regarding YOUR CHILD'S feelings and behavior. There are no right or wrong answers. Please answer as honestly as you can.

Use the numbers to show HOW MUCH each statement IS TRUE FOR YOUR CHILD:

1 = Not at all **2 = Hardly ever** **3 = Sometimes**
4 = Most of the time **5 = All the time**

Now let's try these sentences first. How much do they describe how your child feels?

a. My child likes summer vacation… 1 2 3 4 5
b. My child likes to eat spinach… 1 2 3 4 5

	Not at all	*Hardly Ever*	*Sometimes*	*Most of the time*	*All the time*
1. My child worries about doing something new in front of other children	**1**	**2**	**3**	**4**	**5**
2. My child likes to play with other kids	**1**	**2**	**3**	**4**	**5**
3. My child worries about being teased	**1**	**2**	**3**	**4**	**5**
4. My child feels shy around children he/she doesn't know	**1**	**2**	**3**	**4**	**5**
5. My child only talks to kids that he/she knows really well	**1**	**2**	**3**	**4**	**5**
6. My child feels that other children talk behind his/her back	**1**	**2**	**3**	**4**	**5**
7. My child likes to read	**1**	**2**	**3**	**4**	**5**
8. My child worries about what other children think of him/her	**1**	**2**	**3**	**4**	**5**
9. My child is afraid that others will not like him/her	**1**	**2**	**3**	**4**	**5**
10. My child gets nervous when talking to kids he/she doesn't know very well	**1**	**2**	**3**	**4**	**5**
11. My child likes to play sports	**1**	**2**	**3**	**4**	**5**
12. My child worries about what others say about him/her	**1**	**2**	**3**	**4**	**5**
13. My child gets nervous when meeting new kids	**1**	**2**	**3**	**4**	**5**
14. My child worries that other children don't like him/her	**1**	**2**	**3**	**4**	**5**
15. My child is quiet when he/she is with a group of kids	**1**	**2**	**3**	**4**	**5**

16. My child likes to do things by him/herself	**1**	**2**	**3**	**4**	**5**
17. My child feels that other children make fun of him/her	**1**	**2**	**3**	**4**	**5**
18. If my child gets into an argument with another youngster, he/she worries that the other youngster will not like him/her	**1**	**2**	**3**	**4**	**5**
19. My child is afraid to invite other kids to do things with him/her because they might say no	**1**	**2**	**3**	**4**	**5**
20. My child feels nervous around certain children	**1**	**2**	**3**	**4**	**5**
21. My child feels shy even with kids he/she knows well	**1**	**2**	**3**	**4**	**5**
22. It's hard for my child to ask other children to do things with him/her	**1**	**2**	**3**	**4**	**5**

APPENDIX I

Selective Mutism Questionnaire (SMQ)

Please consider your child's behavior <u>in the last week</u> and rate how frequently each statement is true for your child. Additional questions are included within each subscale to assess how frequently your child used other ways to communicate to people in those settings.

AT SCHOOL	***Never***	***Seldom***	***Often***	***Always***
1. When appropriate, my child talks to most peers at school.	**0**	**1**	**2**	**3**
2. When appropriate, my child talks to selected peers (his/her friends) at school.	**0**	**1**	**2**	**3**
3. When my child is asked a question by his/her teacher, s/he answers.	**0**	**1**	**2**	**3**
4. When appropriate, my child asks his/her teacher questions.	**0**	**1**	**2**	**3**
5. When appropriate, my child speaks to most teachers or staff sat school.	**0**	**1**	**2**	**3**
6. When appropriate, my child speaks in groups or in front of the class.	**0**	**1**	**2**	**3**
When my child communicates with someone at school, he/she speaks to a peer who responds directly.	**0**	**1**	**2**	**3**
When my child communicates with someone at school, he/she speaks in a whisper.	**0**	**1**	**2**	**3**
When my child communicates with someone at school, he/she speaks in a different/unusual voice.	**0**	**1**	**2**	**3**
HOME/FAMILY	***Never***	***Seldom***	***Often***	***Always***
7. When appropriate, my child talks to family members living at home when other people are present.	**0**	**1**	**2**	**3**
8. When appropriate, my child talks to family members while in unfamiliar places.	**0**	**1**	**2**	**3**
9. When appropriate, my child talks to family members that don't live with him/her (e.g., grandparent, cousin)	**0**	**1**	**2**	**3**
10. When appropriate, my child talks on the phone to his/her parents and siblings.	**0**	**1**	**2**	**3**
11. When appropriate, my child speaks with family friends who are well-known to him/her.	**0**	**1**	**2**	**3**
12. My child speaks to at least one babysitter.	**0**	**1**	**2**	**3**

When my child communicates at home or with family, he/she speaks to a parent or sibling who responds directly.	0	1	2	3
When my child communicates with someone at home or with family, he/she speaks in a whisper.	0	1	2	3
When my child communicates with someone at home or with family, he/she speaks in a different/unusual voice.	0	1	2	3

IN SOCIAL SITUATIONS (OUTSIDE OF SCHOOL)	***Never***	***Seldom***	***Often***	***Always***
13. When appropriate, my child speaks with other children who s/he doesn't know.	**0**	**1**	**2**	**3**
14. When appropriate, my child speaks with family friends who s/he doesn't know.	**0**	**1**	**2**	**3**
15. When appropriate, my child speaks with his or her doctor and/or dentist.	**0**	**1**	**2**	**3**
16. When appropriate, my child speaks to store clerks and/or waiters.	**0**	**1**	**2**	**3**
17. When appropriate, my child speaks when in clubs, teams, or organized activities outside of school.	**0**	**1**	**2**	**3**

When my child communicates with others in public, he/she speaks to a parent or sibling who responds directly.	0	1	2	3
When my child communicates with others in public, he/she speaks in a whisper.	0	1	2	3
When my child communicates with others in public, he/she speaks in a different/unusual voice.	0	1	2	3

Interference/Distress (*for clinical use only*)

18. How much does not talking interfere with school for your child?	**Not at all**	**Slightly**	**Moderately**	**Extremely**
19. How much does not talking interfere with family relationships?	**Not at all**	**Slightly**	**Moderately**	**Extremely**
20. How much does not talking interfere in social situations for your child?	**Not at all**	**Slightly**	**Moderately**	**Extremely**
21. How much does not talking interfere with life for your child?	**Not at all**	**Slightly**	**Moderately**	**Extremely**
22. Overall, how much does not talking bother your child?	**Not at all**	**Slightly**	**Moderately**	**Extremely**
23. Overall, how much does your child's not talking bother you?	**Not at all**	**Slightly**	**Moderately**	**Extremely**

APPENDIX J

Screen for Child Anxiety Related Disorders (SCARED)

Directions: Below is a list of sentences that describe how people feel. Read each phrase and decide if it is "Not True or Hardly Ever True" or "Somewhat True or Sometimes True" or "Very True or Often True" for your child. Then, for each statement, circle the number that corresponds to the response that seems to describe your child for the last *3 months*. Please respond to all statements as well as you can, even if some do not seem to concern your child.

	Not True or Hardly Ever True	***Somewhat True or Sometimes True***	***Very True or Often True***	
1. When my child feels frightened, it is hard for him/her to breathe.	**0**	**1**	**2**	*PN*
2. My child gets headaches when he/she am at school.	**0**	**1**	**2**	*SH*
3. My child doesn't like to be with people he/she doesn't know well.	**0**	**1**	**2**	*SC*
4. My child gets scared if he/she sleeps away from home.	**0**	**1**	**2**	*SP*
5. My child worries about other people liking him/her.	**0**	**1**	**2**	*GD*
6. When my child gets frightened, he/she fells like passing out.	**0**	**1**	**2**	*PN*
7. My child is nervous.	**0**	**1**	**2**	*GD*
8. My child follows me wherever I go.	**0**	**1**	**2**	*SP*
9. People tell me that my child looks nervous.	**0**	**1**	**2**	*PN*
10. My child feels nervous with people he/she doesn't know well.	**0**	**1**	**2**	*SC*
11. My child gets stomachaches at school.	**0**	**1**	**2**	*SH*
12. When my child gets frightened, he/she feels like he/she is going crazy.	**0**	**1**	**2**	*PN*
13. My child worries about sleeping alone.	**0**	**1**	**2**	*SP*
14. My child worries about being as good as other kids.	**0**	**1**	**2**	*GD*
15. When my child gets frightened, he/she feels like things are not real.	**0**	**1**	**2**	*PN*
16. My child has nightmares about something bad happening to his/her parents.	**0**	**1**	**2**	*SP*
17. My child worries about going to school.	**0**	**1**	**2**	*SH*
18. When my child gets frightened, his/her heart beats fast.	**0**	**1**	**2**	*PN*
19. My child gets shaky.	**0**	**1**	**2**	*PN*
20. My child has nightmares about something bad happening to him/her.	**0**	**1**	**2**	*SP*
21. My child worries about things working out for him/her.	**0**	**1**	**2**	*GD*

22. When my child gets frightened, he/she sweats a lot.	**0**	**1**	**2**	*PN*
23. My child is a worrier.	**0**	**1**	**2**	*GD*
24. My child gets really frightened for no reason at all.	**0**	**1**	**2**	*PN*
25. My child is afraid to be alone in the house.	**0**	**1**	**2**	*SP*
26. It is hard for my child to talk with people he/she doesn't know well.	**0**	**1**	**2**	*SC*
27. When my child gets frightened, he/she feels like he/she is choking.	**0**	**1**	**2**	*PN*
28. People tell me that my child worries too much.	**0**	**1**	**2**	*GD*
29. My child doesn't like to be away from his/her family.	**0**	**1**	**2**	*SP*
30. My child is afraid of having anxiety (or panic) attacks.	**0**	**1**	**2**	*PN*
31. My child worries that something bad might happen to his/her parents.	**0**	**1**	**2**	*SP*
32. My child feels shy with people he/she doesn't know well.	**0**	**1**	**2**	*SC*
33. My child worries about what is going to happen in the future.	**0**	**1**	**2**	*GD*
34. When my child gets frightened, he/she feels like throwing up.	**0**	**1**	**2**	*PN*
35. My child worries about how well he/she does things.	**0**	**1**	**2**	*GD*
36. My child is scared to go to school.	**0**	**1**	**2**	*SH*
37. My child worries about things that have already happened.	**0**	**1**	**2**	*GD*
38. When my child gets frightened, he/she feels dizzy.	**0**	**1**	**2**	*PN*
39. My child feels nervous when he/she is with other children or adults and he/she has to do something while they watch him/her (e.g., read aloud, speak, play a game, play a sport).	**0**	**1**	**2**	*SC*
40. My child feels nervous when he/she is going to parties, dances, or any place where there will be people that he/she doesn't know well.	**0**	**1**	**2**	*SC*
41. My child is shy.	**0**	**1**	**2**	*SC*

APPENDIX K

Treatment Evaluation Questionnaire (TEQ) – Parent Form

Your child recently completed an intervention in a research study on a treatment approach to selective mutism. Please evaluate the intervention by circling the number which best describes your agreement or disagreement with each statement. Please answer each question.

	Strongly Disagree	***Disagree***	***Slightly Disagree***	***Slightly Agree***	***Agree***	***Strongly Agree***
1. This was an acceptable intervention for my child's problem behavior.	**1**	**2**	**3**	**4**	**5**	**6**
2. Most parents would find this intervention appropriate for behavior problems.	**1**	**2**	**3**	**4**	**5**	**6**
3. The intervention was effective in changing my child's problem behavior.	**1**	**2**	**3**	**4**	**5**	**6**
4. I would suggest the use of this intervention to other parents.	**1**	**2**	**3**	**4**	**5**	**6**
5. My child's behavior problem was severe enough to warrant use of this intervention.	**1**	**2**	**3**	**4**	**5**	**6**
6. Most parents would find this intervention suitable for the behavior problem described.	**1**	**2**	**3**	**4**	**5**	**6**
7. The intervention did not result in negative side effects for my child.	**1**	**2**	**3**	**4**	**5**	**6**
8. The intervention would be appropriate for a variety of children.	**1**	**2**	**3**	**4**	**5**	**6**
9. The intervention was a fair way to handle my child's problem behavior.	**1**	**2**	**3**	**4**	**5**	**6**
10. I liked the procedure used in the intervention.	**1**	**2**	**3**	**4**	**5**	**6**
11. The intervention was a good way to handle my child's behavior problem.	**1**	**2**	**3**	**4**	**5**	**6**
12. Overall, the intervention was beneficial for my child.	**1**	**2**	**3**	**4**	**5**	**6**
13. The intervention quickly improved my child's behavior.	**1**	**2**	**3**	**4**	**5**	**6**
14. The intervention produced a lasting improvement in my child's behavior.	**1**	**2**	**3**	**4**	**5**	**6**
15. The intervention improved my child's behavior to the point that it would not noticeably deviate from other children's behavior.	**1**	**2**	**3**	**4**	**5**	**6**
16. Soon after starting the intervention, I noticed a positive change in my child's problem behavior.	**1**	**2**	**3**	**4**	**5**	**6**
17. My child's behavior remained at an improved level even after the intervention was discontinued.	**1**	**2**	**3**	**4**	**5**	**6**
18. Using the intervention not only improved my child's behavior in the home, but also in other settings.	**1**	**2**	**3**	**4**	**5**	**6**
19. When comparing my child with a peer before and after use of the intervention, my child's and peer's behavior was more alike after using the intervention.	**1**	**2**	**3**	**4**	**5**	**6**
20. The intervention produced enough improvement in my child's behavior so the behavior no longer was a problem.	**1**	**2**	**3**	**4**	**5**	**6**
21. Other behaviors related to the problem behavior also were improved by the intervention.	**1**	**2**	**3**	**4**	**5**	**6**

APPENDIX L

Inclusion and Exclusion Criteria Sheet

The follow form should be used when recruiting each child and his/her caregivers for participation in the current study.

Inclusion Criteria. Please mark all that apply. If a child does not meet all of the following criteria, he/she is not eligible to participate in this project.

____ Child falls within the designated age range (4-8 years old, inclusive)

____ Child exhibits symptoms of Selective Mutism, as indicated by referral to clinic

____ Caregivers rate child between 13 and 27 on the SMQ (full scale), inclusive*

____ At or above a score of 50 (boys) or 54 (girls) on the SASC-R (full scale)*

____ Caregiver and child have been oriented to the study and provided consent/assent

____ Caregiver(s), especially those indicating a history of his/her own anxiety or other psychiatric disorder, are willing and prepared to attend weekly sessions, implement homework assignments, and complete measures of SM symptoms/social anxiety levels

____ Child responded verbally to study clinician ***at least one time*** during baseline behavioral observation.

Exclusion Criteria. Please write "NO" if the statements below do not apply to this child. If one or more are left blank (i.e., not negated), the child is not eligible to participate in this project.

____ Has an existing diagnosis of a major mood disorder, psychotic disorder, or neurodevelopmental disorder (e.g., intellectual disability, communication disorder, autism spectrum disorder).

____ Failure to speak is primarily due to a lack of knowledge of, or familiarity with, the English language (e.g., child is an English Language Learner).

____ The child currently receiving some form of treatment for selective mutism or a related disorder including: other psychosocial treatments (e.g., behavioral therapy, CBT, psychodynamic therapy), medication (e.g., SSRI), or any combination of these.

____ Caregivers report a speech delay and/or existing diagnosis of a speech/language disorder during the diagnostic intake interview*

____ Caregivers report all of the following behaviors on the BCI: "Argues a lot," "disobeys parents," "Fights with other students," and "Takes things that don't belong to him/her"*

**These criteria are used to inform SM subtype, with scores on the SMQ and SASC-R confirming exclusively-anxious presentation, and caregiver-reported absence of speech/language concerns and aggressive behavior ruling out other possible subtypes.*

APPENDIX M

Research Participant Information and Parental Consent Form

You and your child are being asked to participate in a research study. This research is being done as a part of the dissertation requirements being completed by Co-Investigator Allie Siroky, MA. To protect the rights of both you and your child, investigators are required to provide a consent form to inform you about the study, to convey that participation is voluntary, to explain risks and benefits of participation, and to empower you to make an informed decision. You should feel free to ask the researchers any questions you may have. You will be given a copy of this form to keep.

Study Title: Integrated Behavior Therapy for Exclusively-Anxious Selective Mutism: A Nonconcurrent Multiple Baseline Design Across Five Participants
Researchers and Titles: Co-Investigator/Study Coordinator - Allison K. Siroky, MA, Doctoral Candidate in School Psychology; Research Supervisor - John S. Carlson, PhD, LP, Professor of School Psychology
Department and Institution: Michigan State University, College of Education, Department of Counseling, Educational Psychology, and Special Education (CEPSE)
Contact Information: Allison Siroky (sirokyal@msu.edu; 309-235-0700; 447 Erickson Hall, 420 Farm Lane, East Lansing, MI 48824) and John S. Carlson (carlsoj@msu.edu; 517-432-4856443; Erickson Hall, 420 Farm Lane, East Lansing, MI 48824)
Sponsor*:* Michigan State University, CEPSE

1. PURPOSE OF RESEARCH

The purpose of the study is to explore the acceptability, effectiveness, and feasibility of Integrated Behavior Therapy for Selective Mutism (IBTSM), a manualized behavioral approach to treating Selective Mutism. IBTSM encourages caregiver and school involvement, in addition to therapist-directed treatment sessions. In its original format, IBTSM includes 20 sessions over the course of 24 weeks. Early research supports its efficacy since 67% of children receiving IBTSM saw a removal of their SM diagnosis halfway through treatment (at 12 weeks), with 75% no longer meeting criteria for SM after the full 24-week treatment (Bergman, Gonzalez, Piacentini, & Keller, 2013). Building on results from previous research, the current study will examine whether a 16-session version of IBTSM is similarly effective for young children with SM in a community-based clinical setting. The results of this study may help researchers learn whether IBTSM leads to increased speech and lower levels of social anxiety in participating children with SM. The investigator is also interested in caregivers' thoughts about the appropriateness and effectiveness of IBTSM. *The present study will not explicitly address symptoms of other anxiety disorders (e.g., Separation Anxiety, Obsessive Compulsive Disorder, Panic Disorder).*

You and your child have been selected as potential participants in this study because your son or daughter has been diagnosed with Selective Mutism (SM), has no significant oppositional/defiant behaviors or speech/language impairments, and is between the age of 4 and 8. Your participation in this study will last about 18 weeks, with one pre-treatment session, a brief baseline period (5-9 days), and 16 weekly sessions with a licensed clinician. Weekly treatment sessions will last roughly one hour. Behavioral therapy for SM is most effective when strategies can be practiced and applied in different settings outside the clinic. In turn, IBTSM includes additional activities to encourage you and your child to practice skills at home, in school, or in the community.

Baseline Data Collection

In order to better understand how effective IBTSM is in reducing your child's SM symptoms, you will be asked to report your child's current behaviors without treatment for 1-2 weeks. This is known as the baseline phase. Baseline will last only as long as the time between the initial meeting (i.e., Pre-treatment Session) and the first treatment session (i.e., Session 1). You will complete brief measures of your child's speaking behaviors and social anxiety daily during this phase, which will last no more than two weeks. *A stable baseline is needed to determine whether IBTSM is the most appropriate treatment to address your child's individual needs. Stability means that a child's symptoms are consistent over the course of the baseline phase.* ***If behaviors and symptoms during baseline are not stable, this may suggest that your child would respond best to a more individualized treatment plan. If this is the case, you and your family will not be eligible to participate in the study and will be placed back on the clinic waitlist to receive more appropriate care.***

2. WHAT DOES IBTSM INVOLVE?

IBTSM is a manualized behavioral treatment for young children with SM. Behavioral therapy helps children to learn adaptive coping skills for managing anxiety, and it supports them in practicing these skills in controlled activities which mimic social situations where they tend to have a difficult time speaking. IBTSM is an integrative approach, so it emphasizes caregiver involvement throughout. Through IBTSM, caregivers learn about SM and how to encourage their child to apply coping skills when anxious. Caregiver responsibilities increase over time as they work with clinicians to plan and carry out practice activities outside of the clinic setting. IBTSM uses the following behavioral tools to help children overcome their fear of speaking:

- **Psychoeducation** – Learning about anxiety and how it affects the way we think, act, and feel
- **Fear Ladder –** Identifying situations where the child does not speak and developing an individualized "ladder" where children list situations from least to most challenging
- **Coping Skills –** Children learn effective ways to manage discomfort or anxious feelings
- **Applied Practices –** Starting at the bottom of their fear ladder, the child learns to use coping skills in activities which mimic situations they encounter in their daily life where they typically avoid speaking. The clinician, the child, and caregivers work together to set goals and practice before planned activities to increase the likelihood of success
- **Caregiver Skill Training** – Caregivers observe the clinician and learn to apply the same behavioral strategies during out-of-session practices to support growth in other settings
- **Reward System** – Contingency management, or a system of reinforcement, which provides rewards to children for successfully meeting goals as planned and practiced

Basic Procedures of IBTSM

Although this study follows a condensed treatment plan, no changes were made to the procedures, activities, techniques, or length of weekly sessions outlined in the 24-week IBTSM plan. In total, IBTSM will last about 18 weeks. Your child will be assigned to a clinician who is licensed in the state of Michigan, has been well trained in behavioral therapy, and has experience implementing behavioral therapy with children with SM. Your child's clinician will be supervised by Dr. Aimee Kotrba, LP, who is well-known for her experience in treating children with SM.

A pre-treatment session is meant specifically for caregivers and involves a discussion about Selective Mutism, overall treatment plans and expectations, and a pre-treatment evaluation to learn specific details about your child's symptoms. The first two treatment sessions focus on developing a warm relationship between your child and his/her clinician, particularly in cases where your child is not yet consistently speaking to them. You and your child will also learn more

about the behavioral tools and strategies used in IBTSM to increase speech and to manage anxiety (see list above).

After both you and your child are comfortable with these practices, the focus of treatment transitions to ongoing applied practices in situations where speaking is expected, guided by your child's "fear ladder" (i.e., exposure activities). The clinician will start at the bottom of the fear ladder (least challenging) and move up slowly after your child successfully uses learned coping skills and becomes more comfortable with a given "step" on the ladder. With time and continued practice, children move progressively up the ladder as they begin to effectively complete goals in various settings such as the classroom, during social gatherings with peers, or during spontaneous interactions in public. As your child successfully speaks in new situations and with new people, boosts in self-confidence and earning rewards (e.g., stickers, toys, privileges) reinforce speaking behaviors, rather than speech avoidance. Clinicians and caregivers spend the beginning of each session discussing treatment progress, practice goals, and any unexpected difficulties in applying skills or strategies outside of the clinic.

To support this ongoing discussion about the effectiveness of IBTSM, the midpoint session (Session 8) serves as an opportunity to formally assess progress in terms of verbal communication and levels of social anxiety. In the remaining weeks of treatment, caregivers begin to take on greater responsibility in planning and leading practice activities as you work closely with your child to determine appropriate rewards for their hard work. Each week, caregivers will be given additional assignments to be complete at home, in school, or in the community. Assignments typically consist of additional applied practice activities, which will be decided upon during sessions and are often paired with a reward. The final session (Session 16) focuses on providing caregivers with resources on how to maintain treatment gains and on building overall confidence by emphasizing the child's progress.

3. POTENTIAL BENEFITS OF IBTSM

Children participating in this study may experience the following potential benefits: knowledge and application of adaptive coping skills, increased verbal communication across settings, decreased levels of social anxiety in previously feared settings, increased social competence, and stronger relationships with caregivers or other relevant people who may be the focus of practices. Caregivers are also expected to learn more about Selective Mutism and will likely learn how to employ effective strategies for maintaining progress after treatment is finished. Finally, as additional case study research on IBTSM are completed, information about the effects of this treatment approach will develop an evidence base for informing future researchers, and society in general, about appropriate interventions for children with SM.

4. POTENTIAL RISKS OF IBTSM

There are no foreseeable risks to you or your children for participating in this study. Previous investigators of IBTSM found no adverse effects of this treatment (Bergman et al., 2013; Siroky, Carlson & Kotrba, 2017). Children are not likely to experience discomfort greater than what they typically encounter in daily life. In the rare and unforeseeable event that a child experiences significant discomfort, licensed study clinicians will use their training in behavioral therapy to take appropriate steps to reduce discomfort and they will have access to their supervisor, if needed.

5. PRIVACY AND CONFIDENTIALITY

Your confidentiality will be protected to the maximum extent allowable by law. Protected health information used to inform diagnosis and treatment will be stored in the clinic's HIPAA-approved, password-protect online system used with all other clients of the clinic. Please refer to the

Authorization to Use or Disclose Health Information for Research form you were asked to review and sign prior to your enrollment for further details.

Each child enrolled in the study will receive a participant number. Any communication between researchers, project assistants, and other adults involved (school teacher, clinician) will contain no more your child's designated participant number or, if needed for clarification, your child's first name. All paper documents collected for this research study (e.g., caregiver-completed questionnaires) will use the same designated participant number and will be stored in a locked file cabinet on the campus of Michigan State University for a minimum of three years after the close of the project. Electronic data (e.g., results from questionnaires) will be stored on a password-protected computer and will include your child's designated participant number. Only the appointed researchers and the Human Research Protection Program (HRPP) will have access to the research data. *Although we will make every effort to keep your data confidential there are certain times, such as a court order, where we may have to disclose your data. For example, if participants disclose to the therapist of suspected child abuse or neglected, the therapist is legally mandated to report their claim for further investigation.*

6. YOUR RIGHTS TO PARTICIPATE, SAY NO, OR WITHDRAW

Participation in this study is voluntary. Refusal to participate will involve no penalty or loss of benefits to which you are otherwise entitled. You and your child are entitled to the following rights:

- You and/or your child may discontinue participation at any time without penalty or loss of benefits to which you are otherwise entitled.
- You and/or your child have the right to say no to activities included in IBTSM.
- You and/or your child may change your mind at any time and withdraw.
- You and/or your child may choose not to answer questions or to stop participating at any time.

7. COSTS AND COMPENSATION FOR BEING IN THE STUDY

Your participation in the research project will not involve any additional costs to you or your health care insurer. All fees for treatment at Thriving Minds Behavioral Health Center (one intake session and 16 weekly therapy sessions) have been covered. Each family will receive an additional $200 compensation via money cards progressively (i.e., $100 at first visit, $50 at midpoint, and $50 at end of services) to be used toward the following:

- You will receive $50 to buy gifts, toys, etc. for the reward system component of treatment.
- You will also be compensated up to $100 for travel fees.
- Finally, you will receive an additional $50 stipend for your active participation in the study.

8. ALTERNATIVE OPTIONS

If you or your child choose to withdraw from this research project before treatment ends, or if treatment is ineffective, your child's clinician will provide alternative, evidence-based treatment options for you to consider. Consistent with effective treatments found within the literature, options may include one or more of the following:

- Referral to a child psychiatrist to discuss supplementing behavioral therapy with medication
- Referral to a clinician who specializes in treating children with treatment-resistant SM
- Intensive-dose behavioral therapy, which involves longer individual therapy sessions (3-4 hours) over a shorter period of time (4-5 days).
- A second diagnostic assessment to identify other potential conditions not identified during the study's screening procedures which may pose a barrier to treatment effectiveness

9. CONTACT INFORMATION

If you have concerns or questions about this study, such as scientific issues, how to do any part of it, or to report an injury, please contact the researchers (Allison K. Siroky, MA: *e-mail* - sirokyal@msu.edu, *phone* - 309-235-0700; John S. Carlson, PhD, LP: *e-mail* – carlsoj@msu.edu, *phone* - 517-432-4856)

If you have questions or concerns about your role and rights as a research participant, would like to obtain information or offer input, or would like to register a complaint about this study, you may contact, anonymously if you wish, the Michigan State University's Human Research Protection Program at 517-355-2180, Fax 517-432-4503, or e-mail irb@msu.edu or regular mail at 4000 Collins Rd, Suite 136, Lansing, MI 48910.

10. DOCUMENTATION OF INFORMED CONSENT

Your signature below means that you voluntarily agree to participate in this research study.

_____ _____

Caregiver Signature Date

_____ _____

Signature of Therapist Date

Videotaping for Treatment Evaluation

To ensure that treatment is carried out as intended, sessions will be videotaped and reviewed by the clinicians and researchers from this project only. Video files will be stored in a password-protected online drive. Only your child's clinician, their supervisor, appointed researchers, and the Human Research Protection Program (HRPP) will have access to these video files.

Videotaping is required to participate in the study as an important question related to this research will examine how well clinicians adhere to the treatment manual. Please indicate your preferences regarding videotaping treatment sessions.

- I agree to allow audiotaping/videotaping of my child the during treatment sessions.

  ☐ Yes ☐ No Caregiver Initials_____

Use of Results for Dissemination

The results of this study may be published in a research journal or presented at professional meetings, but the identities of all research participants, including identifying information about you and/or your child, will remain anonymous.

- I agree to allow my child's results from this study to be disclosed in reports and presentations.

  ☐ Yes ☐ No Caregiver Initials_____

Re-contacting for Follow-up Information

Although the primary goal of the present study is to examine short-term effects of IBTSM for your child with SM, we may want to learn more about long-term effects a few months after the

final treatment session. In order to obtain this data, we may re-contact you to schedule a brief follow-up interview at either three or six months post-treatment.

- I agree to allow the study coordinator to contact me in three to six months after treatment has finished to ask follow-up questions regarding the long-term effects of the treatment for my child.
  ☐ Yes ☐ No Caregiver Initials_____

APPENDIX N

Research Participant Informatin and Child Assent Form

Study Title: Integrated Behavior Therapy for Exclusively-Anxious Selective Mutism: A Nonconcurrent Multiple Baseline Design Across Five Participants
Researchers and Titles: Co-Investigator/Study Coordinator - Allison K. Siroky, MA, Doctoral Candidate in School Psychology; Research Supervisor - John S. Carlson, PhD, LP, Professor of School Psychology
Department and Institution: Michigan State University, College of Education, Department of Counseling, Educational Psychology, and Special Education (CEPSE)
Contact Information: Allison Siroky (sirokyal@msu.edu; 309-235-0700; 447 Erickson Hall, 420 Farm Lane, East Lansing, MI 48824) and John S. Carlson (carlsoj@msu.edu; 517-432-4856443; Erickson Hall, 420 Farm Lane, East Lansing, MI 48824)
Sponsor*:* Michigan State University, CEPSE

The following script is to be used to obtain non-verbal assent from children between the ages of 4 and 8 years old. The clinician should read the following script to the child without caregivers present, if possible. If a child is reluctant to respond to the clinician, caregivers may administer the form.

I want to tell you about a research study I am working on. A research study is usually done to understand how or why things happen. In this study, I want to find out more about children who have a hard time talking when they are in certain places or around certain people. I and other people who work with these kids have a good idea of what can help them feel better about talking, but we want to be sure that this plan works. This study will help me learn more about one plan for helping children feel brave when talking.

You are being asked to help us with this study because, like many other kids, you find it hard to speak around certain people or in certain places. In any study, I can only include people who want to participate. This is why I am asking if you are okay with helping me. You do not have to if you do not want to.

Let me tell you a little about this project. You will be visiting with me (*or name of therapist)* each week. You will learn more about why it is hard for you to speak sometimes, and I (*or name of therapist*) have/has some activities planned to try and make speaking easier for you. Your parents, your teacher at school, and some of your classmates will even be helping out along the way. Some of these activities will be tough, but I will never make you do anything that is too scary or too hard. You can always tell your parents or me if things feel too hard. I hope this plan will make you feel strong and brave about talking, but not everyone who takes part in this study will feel all better.

You do not have to be in this study if you do not want to be. If you decide to stop after we begin, that's okay too. No one will be mad at you if you change your mind. If you do not want to be in this research project, I will tell you what other kinds of plans there that could help you feel brave. Your parents have said it is okay for you to be in this study. If you have questions, please ask them now or at any time. Do you have any questions about the study? *(If child says yes, try*

to answer them or have them tell their caregivers so they can tell you later. Answer all questions before proceeding)

I have just a few questions for you. You can answer 'Yes' by nodding your head like this *(models nod of approval)* or you can say 'No' by shaking your head like this *(shakes head in disapproval)*. I also have a 'Yes' card and a 'No' card so you can point to your answer if you would like.

1. Are you okay with being a part of this study and working with me (*or name of therapist)* each week?

Child's/Subject's response**:** ☐Yes ☐No

(If the child answers 'Yes' to both, proceed to the following questions)

2. When I am finished with this study I will write a paper to tell other people what I have learned. This paper will not include your name, and no one will know that you were in the study. Are you okay with me writing a report about what I learn from this study?

Child's/Subject's response**:** ☐Yes ☐No

3. I also want to make sure that your therapist does a really good job following this plan. To help us do this, I want to record our weekly meetings and watch them again to remember what happened. No one else will see these recordings except the people working on this study. Are you okay with having our weekly meetings recorded on videotape?

Child's/Subject's response**:** ☐Yes ☐No

CHECK WHICH APPLIES BELOW:

☐The child/Subject is capable of understanding the study

☐The child/Subject is not capable of understanding the study

_____
Child's/Subject's Name (printed)

_____ _____
Name (printed) and Signature of Person Obtaining Consent Date

Appendix O

Visual Analysis Guides

Visual Analysis Guide: SASC-FNE Scores, Child # _____

Complete the following form for each child, using the graph of his/her SASC-FNE scores.

Preliminary Analysis. Do the data clearly depict problematic behavior (e.g., high social anxiety)? Are baseline data relatively stable?

___ Yes to both (Continue to Step 1)

___ No to either (Discontinue analysis)

Step 1: ***Level (Mean).*** Is there a noticeable change in the level (mean) between the baseline phase and the treatment phase. In other words, is the level visually *lower* than the baseline phase?

___ Yes, the level of treatment scores is visually lower than baseline (Continue to Step 2)

___ No (Discontinue analysis, mark "No noticeable treatment effect" at bottom of page)

Step 2: Trend (Slope). Is the trend (slope) distinctly more prominent during the treatment phase compared to the baseline phase? Is it trending in the anticipated direction (i.e., *downward trend over time*)?

___ Yes, the trend is more prominent in the anticipated direction during treatment (Continue to Step 3)

___ No (Discontinue analysis, mark "No noticeable treatment effect" at bottom of page)

Step 3: Variability. Are data from the treatment phase relatively stable (i.e., with minimal vertical spread and/or change in direction of slope)?

___ Yes, data from the treatment phase are stable (Continue to Step 4)

___ No (Discontinue analysis, mark "No noticeable treatment effect" at bottom of page)

Step 4: Immediacy of Effect. Are these changes noticeable within the first 3 weeks after the treatment start point? Did changes begin *after* the baseline phase (after the intervention began)?

___ Yes, observable changes occurred within 3 weeks after treatment onset (Continue to next graph or proceed to Step 5)

___ No (Mark "No noticeable treatment effect" at bottom of page)

_____ Noticeable treatment effect **_____ No noticeable treatment effect**

Visual Analysis Guide: BROSB Scores, Child # _____

Complete the following form for each child, using the graph of his/her BROSB scores.

Preliminary Analysis. Do the data clearly depict problematic behavior (e.g., low frequency of speech?) Are baseline data relatively stable?

___ Yes to both (Continue to Step 1)

___ No to either (Discontinue analysis)

Step 1*: *Level (Mean). Is there a noticeable change in the level (mean) between the baseline phase and the treatment phase. In other words, is the level visually *higher* than the baseline phase?

___ Yes, the level of treatment scores is visually higher than baseline (Continue to Step 2)

___ No (Discontinue analysis, mark "No noticeable treatment effect" at bottom of page)

Step 2: Trend (Slope). Is the trend (slope) distinctly more prominent during the treatment phase compared to the baseline phase? Is it trending in the anticipated direction (i.e., *upward trend over time*)?

___ Yes, the trend is more prominent in the anticipated direction during treatment (Continue to Step 3)

___ No (Discontinue analysis, mark "No noticeable treatment effect" at bottom of page)

Step 3: Variability. Are data from the treatment phase relatively stable (i.e., with minimal vertical spread and/or change in direction of slope)?

___ Yes, data from the treatment phase are stable (Continue to Step 4)

___ No (Discontinue analysis, mark "No noticeable treatment effect" at bottom of page)

Step 4: Immediacy of Effect. Are these changes noticeable within the first 3 weeks after the treatment start point? Did changes begin *after* the baseline phase (after the intervention began)?

___ Yes, observable changes occurred within 3 weeks after treatment onset (Continue to next graph or proceed to Step 5)

___ No (Discontinue analysis, mark "No noticeable treatment effect" at bottom of page)

_____ Noticeable treatment effect **_____ No noticeable treatment effect**

Visual Analysis Guide: ABO Words Spoken, Child # _____

Complete the following form for each child, using the graph of his/her ABO Words Spoken.

Preliminary Analysis. Do the data clearly depict problematic behavior (e.g., low frequency of speech)?

___ Yes (Continue to Step 1)

___ No (Discontinue analysis)

Step 1: ***Level (Mean).*** Is there a noticeable change in the level (mean) between the baseline phase and the treatment phase. In other words, is the level visually *higher* than the baseline phase?

___ Yes, the level of treatment scores is visually higher than baseline (Continue to Step 2)

___ No (Discontinue analysis, mark "No noticeable treatment effect" at bottom of page)

Step 2: Trend (Slope). Is the trend (slope) distinctly more prominent during the treatment phase compared to the baseline phase? Is it trending in the anticipated direction (i.e., *upward trend over time*)?

___ Yes, the trend is more prominent in the anticipated direction during treatment (Continue to Step 3)

___ No (Discontinue analysis, mark "No noticeable treatment effect" at bottom of page)

Step 3: Variability. Are data from the treatment phase relatively stable (i.e., with minimal vertical spread and/or change in direction of slope)?

___ Yes, data from the treatment phase are stable (Continue to Step 4)

___ No (Discontinue analysis, mark "No noticeable treatment effect" at bottom of page)

Step 4: Immediacy of Effect. Are these changes noticeable within the first 3 weeks after the treatment start point? Did changes begin *after* the baseline phase (after the intervention began)?

___ Yes, observable changes occurred within 3 weeks after treatment onset (Continue to next graph or proceed to Step 5)

___ No (Mark "No noticeable treatment effect" at bottom of page)

_____ Noticeable treatment effect **_____ No noticeable treatment effect**

Visual Analysis Guide: Consistency of Change Check
To be completed for all three measures.

After reviewing data for all five cases, carefully examine all of the within-phase data together for each measure (i.e., five together for BROSB, five together for SASC-FNE). Look first at the baseline data across all cases. Next, examine the treatment phases across all cases. Answer the following question:

Step 5: Consistency of Data. When analyzing all of the baseline phases and then all of the treatment phases at one time, do the observable changes appear to be *consistently occurring during the treatment phase* for all cases (i.e., a treatment effect is clearly replicate)? Mark one.

_____ Yes, there is a clear replicated effect _____ No, there was no replicated effect

_____ Mixed results because (please explain): _____

_____

_____

_____

APPENDIX P

Supplemental Tables

Table 14.

Changes in SASC-R Total and Subscale Scores at Baseline and End-of-Treatment

	Total Score	Fear of Negative Evaluation	Fear of New Situations	General Social Anxiety
Child 1				
Baseline	**58**	21	21	16
End-of-treatment	**33***	8	16	9
Child 2				
Baseline	**56**	13	29	14
End-of-treatment	**51**	8	29	14
Child 3				
Baseline	**64**	24	28	12
End-of-treatment	**27***	8	13	6
Child 4				
Baseline	**46**	14	19	13
End-of-treatment	**39***	8	20	11
Child 5				
Baseline	**67**	14	27	12
End-of-treatment	**35***	8	20	7
Average				
Baseline	**58.2**	17.2	24.8	13.4
End-of-treatment	**37***	8	19.6	9.4

*An RCI greater than 1.96 or less than -1.96 indicates a clinically significant change.

Table 15.

Changes in SCARED Total and Subscale Scores from Baseline to End-of-Treatment

	Total Score	Panic/ Somatic	GAD	Separation	Social	School Avoidance
Child 1						
Baseline	**35**	5	8	5	14	4
End-of-treatment	**15***	1	5	1	7	1
Child 2						
Baseline	**27**	2	7	0	14	4
End-of-treatment	**19**	2	3	0	13	1
Child 3						
Baseline	**40**	2	11	8	13	6
End-of-treatment	**17***	0	0	1	11	5
Child 4						
Baseline	**35**	0	6	9	14	6
End-of-treatment	**23***	2	4	10	7	0
Child 5						
Baseline	**26**	4	4	5	13	0
End-of-treatment	**17***	1	1	4	8	0
Average						
Baseline	**32.6**	2.6	7.2	5.4	13.6	4.0
End-of-treatment	**17.6***	1.2	2.6	3.2	9.2	1.4

*An RCI greater than 1.96 or less than -1.96 indicates a clinically significant change.

Table 16.

Changes in SMQ Total Scores and Item-Level Mean Scores by Subscale from Baseline to End-of-Treatment

	Total Score	School	Home	Other Social
Child 1				
Baseline	**17**	0.67	1.80	0.25
End-of-treatment	**27***	1.50	2.50	0.60
Child 2				
Baseline	**13**	0.00	1.60	0.50
End-of-treatment	**18**	0.67	1.83	0.60
Child 3				
Baseline	**16**	0.67	2.40	0.00
End-of-treatment	**36**	1.83	2.50	2.00
Child 4				
Baseline	**15**	0.17	2.40	0.20
End-of-treatment	**38***	2.50	2.67	1.40
Child 5				
Baseline	**16**	1.00	1.20	0.60
End-of-treatment	**32***	2.00	2.00	2.00
Average				
Baseline	**15.40**	0.50	1.88	0.31
End-of-treatment	**29.60***	1.70	2.30	1.32

*An RCI greater than 1.96 or less than -1.96 indicates a clinically significant change.

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