

Critical Review:

The efficacy of recent intervention techniques (from 2016-2018) used to treat selective mutism

Leighanne Chan, Julia Walker
M.Cl.Sc SLP Candidates

University of Western Ontario: School of Communication Sciences and Disorders

This critical review examines the evidence regarding recent techniques used in the treatment of selective mutism (SM). Types of interventions for SM include behavioural, psychodynamic, pharmaceutical, systems, and multimodal approaches. Overall, the evidence gathered from this review suggests that the strongest degree of support is for behavioural approaches and multimodal approaches that included a behavioural component. This review revealed that speech-language pathologists (S-LPs) are not represented in the recent research or the treatment of SM. Recommendations for future research and implications for the role of S-LPs in the treatment of SM are provided.

Introduction

Selective mutism (SM) is listed in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) as an anxiety disorder that occurs in childhood. It is characterized by the consistent failure to speak in specific social contexts that require speaking (e.g., school), when the child is able to speak in other settings (e.g., at home) (Zakszeski & DuPaul, 2017).

There are five different categories of treatment approaches for SM described in the literature: behavioral, psychodynamic, psychopharmacological, systems, and multimodal approaches. Behavioural approaches indirectly or directly examine and treat the reason behind the failure to speak in certain circumstances (e.g., to escape demands or to gain attention) based on theories from applied behaviour analysis, cognitive behavioural approaches, and/or social learning theories (Zakszeski & DuPaul, 2017). Psychodynamic approaches usually employ play or art therapies, as they attempt to understand the origins of SM through the child's unconscious. Pharmacological approaches utilize medication to relieve SM systems. Systems approaches provide education, counselling, and skills training to individuals who are significant to the child with SM, such as parents, family members, and teachers (Zakszeski & DuPaul, 2017). Finally, multimodal approaches combine methods from multiple approaches (Muris & Ollendick, 2015).

As SM poses long term effects on language, communication, academic and occupational performance, as well as psychiatric well-being (Muris & Ollendick, 2015), it is important that children receive effective and timely intervention. However, there is still no specific or universally accepted treatment for SM (Esposito et al., 2016). Since children with SM are often referred for S-LP services, there is a need to

examine the current intervention approaches and efficacy of these different treatment methods.

Objectives

The objective of this paper is to critically review the literature in order to discover the efficacy of recent intervention methods used to treat selective mutism.

Methods

Search Strategy

Several computerized databases, including PubMed, Psych Info, Google Scholar, were searched using the following terms:

(treatment) OR (intervention) OR (therapy) AND (selective mutism) OR (selective mute child).

The search was limited to articles written in English between 2016-2018.

Selection Criteria

Studies selected for inclusion in this review paper were required to investigate any type of treatment for selective mutism between the years of 2016 and 2018. Limits included were English studies with human research subjects

Data Collection

This search yielded eight articles that fulfilled the above requirements. The results of the literature search yielded the articles with the following treatment types harmonious with the selection criteria mentioned above: behavioural therapy (shaping hierarchy, cognitive behavioural therapy), pharmacology (Fluoxetine, Pramipexole), psychodynamic therapy (psychomotor therapy, Ericksonian Hypnotherapy), and multimodal approach (S-CAT: systems and behavioural approach).

Results

Behavioural Therapy:

Lang, Gothelf, Domachevsky, Ginton, Kushnir, and Gothelf (2016) completed a single group study without controls to examine the long-term outcomes of cognitive behavioural therapy (CBT) for children with SM. Twenty-four children who were between the ages of 5 to 15 at the time of follow-up, at least one-year post-treatment, participated in this study. All children had social anxiety disorder (SAD) at baseline, met DSM-IV criteria for SM, and received modular SM-focused CBT. Outcomes were assessed using several measures, including the Anxiety Disorders Interview Schedules for DSM-IV: Life-Time Version (ADIS-IV-L) and the Selective Mutism Questionnaire (SMQ). Results were analyzed through paired *t*-tests, the McNemar test, and independent sample *t*-tests. Lang et al. (2016) found that children who completed the modular CBT showed improvements in SM and SAD symptoms based on clinician and parent rating scores. There was more significant improvement in those who completed the program compared to those who did not, which demonstrates the efficacy of modular CBT, given that completers and non-completers were similar at baseline. Lang et al. (2016) also reported significant decline in rate of SM, SAD, and specific phobia from baseline to the end of the study.

The study had a satisfactory sample size, and though only 24 of the 36 children who participated in the original study also participated in this follow-up study, the authors noted that those who did not participate in the follow-up study were similar to those who did in terms of severity of SM symptoms and rates of comorbid psychiatric diagnoses. However, there were no exclusion criteria mentioned for participants, and comorbid psychiatric diagnoses were noted to be present in some children. An additional confound was that some children were also taking medication at the time of the original study. Modules for CBT were well-described in terms of goals, but not detailed enough for a clinician to fully carry the treatment out. Given that SM symptoms are often most severe in school settings, the authors acknowledged that they should have used an outcome measure from teachers' perspectives. Lang et al. (2016) also noted the lack of a control group.

Overall, Lang et al. (2016) showed that SM-focused CBT is feasible in children with SM, and that there are possible long-term effects in reducing SM and comorbid anxiety symptoms. However, there needs to be randomized control trials, with participant characteristic confounds removed, as well as an outcome measure with teacher reports of SM symptoms to further and more accurately assess the efficacy of this approach.

Oerbeck, Overgaard, Stein, Pripp, and Kristenson (2018) completed a follow-up study for a randomized control trial to examine long-term effects of school-based cognitive behavioural therapy for SM completed five years prior. Thirty of the 32 children who participated in the original study also participated in this follow-up study. The authors noted that both children of the two families who did not reply to the invitation to participate in the follow-up study had SM and social phobia at the one-year follow-up study. All children were treated with weekly sessions of school-based CBT lasting up to 6 months by local clinically experienced therapists who followed a detailed manual, under the supervision of the first or last author, but with no further adherence measures. Outcome measures included diagnostic status, teacher- and parent-rated questionnaires, child-rated quality of life and speaking behaviour. At the five-year follow-up, Oerbeck et al. (2018) found that 70% of participants were in full remission, 17% were in partial remission, and 13% still had selective mutism. The authors noted that most children showed continuous progress, except for three children who had regressed since the 1-year follow-up. Oerbeck et al. (2018) also reported more prominent improvement in younger children aged 3-6 in the original study. In addition to improvements seen across all outcome measures, there was a reduction of comorbid anxiety disorders.

This study has a strong level of evidence, with a representative sample for the population in question. The sample population, inclusion, and exclusion criteria for the study were well described by the authors. Explanations of possible confounds were discussed, and inclusion and exclusion criteria were valid. Across the original study, the 1-year follow-up study, and this 5-year follow-up study, outcome measures were consistently used. The CBT training and manual could have been more explicitly explained, as a new clinician would not be able to implement this treatment based only on this study. Results may have been positively skewed as the number of participants in the follow-up study differed from the number of participants in the original study, especially since the two families who did not participate in the current study had children who continued to have SM and social phobia at the previous follow-up study. Besides this, detailed statistical analysis was completed.

Overall, this study provided compelling evidence of the long-term efficacy of CBT in the treatment of SM for children ages 3-9, but especially so in younger children ages 3-6.

Bunnell, Mesa, and Beidel (2018) utilized a between groups design to assess the behavioural change during the implementation of a two-session hierarchy

for shaping successive approximations of speech in children with SM. Fifteen children with SM (ages 5-7) were randomly assigned to one of 3 behavioural groups: shaping using mobile apps (i.e., Apple iPad), shaping using other therapeutic tools, or shaping using reinforcement. Children participated in two 55-minute treatment sessions. Treatment outcomes were measured using behavioural responses of the children (galvanic skin conductance), time taken to complete the shaping hierarchy, child and caregiver reports of the child's social anxiety, and caregiver report of the child's speaking. Results showed that 93.33% of the participants completed the hierarchy and were speaking to unfamiliar adults by the end of the second session, regardless of the shaping modality used in the session.

Participants with comorbid diagnoses and on a stable dose of antidepressant medication were not excluded from this study. The authors use of flexible inclusion criteria was a strength, since the results can be generalized to individuals with multiple diagnoses. Even with the wide inclusion criteria, this study had a small sample size, which limits the generalizability of the results. The small sample size also restricted the authors' ability to test group differences statistically, and as a result, authors compared the data descriptively among groups using 95% confidence intervals.

There were several strengths included in this study methodology. Authors randomly assigned participants to treatment groups, which eliminated potential treatment bias. A variety of assessment measures were used, including physiological measures, time measurement, child and caregiver reports. This increases confidence in the results obtained, as they do not rely exclusively on unreliable caregiver report. Overall, the study methodology and intervention techniques were well described, which would allow the study to be easily replicated.

As pointed out by the authors, this shaping hierarchy could be used as a useful tool during the initial stages of SM treatment, but it is not a treatment in itself. Treatment was provided to the participants by the authors following this study; however, these treatment approaches were not standardized, and no data was reported on maintenance and generalization of speaking behaviors during this follow-up. Therefore, this study does not provide evidence to support the effectiveness of this technique as a first step in SM treatment. Due to these factors, this study offered a suggestive level of evidence, and requires further research to support the use of these methods in conjunction with a SM intervention. The results of the study show promise, as this technique may be helpful in allowing children with SM to begin to speak in order to make effective gains in intervention.

Pharmacology:

A study conducted by **Barterian et al. (2018)**, used a single-case design to examine the effects of the use of fluoxetine for the treatment of SM. Six children with SM, who had not benefited from previous psychosocial treatment, were randomly assigned to different fluoxetine treatment schedules. Treatment schedules varied by the length of time children received the placebo before receiving fluoxetine. Parents were asked to observe their child in social interactions with an unfamiliar adult 3 times per week and rate their child's social engagement. Results showed that children experienced improvement in social anxiety, responsive speech, and spontaneous speech with medium to large effect sizes; however, children still met DSM-V criteria for SM at the end of the study.

This study included a small sample size, which makes the results difficult to generalize to the greater population. The exclusion criteria used in this study were extensive, which also limited the population of children to which these results can apply.

The researchers designed a sound treatment methodology that complemented a small sample size. Participants were randomly assigned to a treatment schedule, which increases confidence in the effectiveness of the treatment. Researchers also used a placebo, allowing each participant to act as their own control when comparing post-fluoxetine behaviour to baseline measurements. The researchers ensured that the placebo and treatment were the same flavor, color, consistency, and quantity, to reduce the chance that behaviour of the participant would change simply by knowing they were receiving the treatment. Authors accounted for possible placebo effect by building in a week of no medication into all participants' treatment schedules. The treatment schedules and methods were well-described in the paper, which will make the results easy to replicate in future studies.

Due to the nature of this study design and the small number of participants, no analytical statistical measures were completed. The appropriate descriptive statistics (i.e., Cohen's *d*, SD) were used to measure difference in engagement between baseline and post-therapy levels.

The level of evidence offered by this study is moderately suggestive due to the appropriateness of the study design, the measures used, and the analysis completed. However, based on the findings of the study, this type of therapy may result in modest gains only, as all children still met criteria for SM at the end of the study.

Naguy (2017) examined the efficacy of pramipexole medication for the treatment of a 9-year-old girl with chronic SM, who was unresponsive to other medications and behavioural treatment. In the

current study, pramipexole was used over 6 weeks, improvement was demonstrated by week 3, and response was maintained at week 6, 8, and 12. Booster sessions were noted to help with maintenance of results. The medication was reported to result in transient nausea initially, but generally had high tolerability.

The study sample consisted of only one participant, which decreases the power of the study. Authors argued that SM was a motivational deficit, and since pramipexole had been shown to boost dopamine and increase motivation, it was a logical medication for SM. However, explanations were not provided on how SM and motivation were related. Authors argued that the child's previous failure with other drug trials is evidence against the placebo effect, but a randomized control trial with a larger sample size would have been stronger evidence against the placebo effect. Outcome measures were not clearly defined beyond the child reportedly being more engaged with the therapist and other adults, so statistical analyses were not performed for outcome data.

Overall, Naguy's (2017) study provides an alternative pharmacological option for treatment of chronic SM, but the study needs to be reduplicated with clearer outcome measures and a larger sample size to provide stronger evidence for this approach.

Psychodynamic:

Researchers **Cavarra, Brizio, and Gava (2016)** presented a single case study in which Ericksonian Hypnotherapy was used as main treatment for SM. The participant, a 7-year-old girl with SM, completed five sessions of Ericksonian Hypnotherapy over the course of three months. The results, as reported by the client's mother, showed significant changes in the child's behaviour immediately following treatment, including the resolution of symptoms and the diagnosis of SM being no longer applicable.

The intervention techniques used by the therapist were well-described in a narrative format. Despite these description, these methods would be impossible to recreate, as the therapy is individualized and not standardized. This study also contained several methodological limitations. The author acknowledged the inherent limitations of this study due to the nature of a single case design. The researcher noted that the generalizability of results to other patients is extremely limited from this study, and that the effectiveness of the intervention must be tested in more methodologically sound studies. Another limitation of this study was the lack of formalized assessment measures. The researcher could have utilized a standard measure of SM at the beginning and end of treatment in order to quantify the change in the child's behaviour, instead of relying completely on caregiver report. As a result, all findings from this study are based on parent report, which is not

an objective measure, as the mother may be trying to please therapist. As there were no formal assessments given, there was no use of descriptive or analytical statistical measures. Assessment measures and statistical analyses would increase the reader's confidence in the validity of this therapeutic approach.

Due to these limiting factors, this study offered an equivocal level of evidence. Further research is needed to clarify these methods and generalize this therapy approach. The results of the study show promise, as this technique may be helpful in treating children with SM in a timely manner who have not responded to other intervention types.

Esposito et al., (2016) investigated the efficacy of a 6-month standard psychomotor treatment in children with SM. Participants included 138 children with SM who were randomly assigned to one of two treatment groups: psychomotricity or behavioral and educational counseling. Psychomotricity treatment was delivered by trained child therapists three times per week during 45 minute sessions. After 6 months of treatment, the Selective Mutism Questionnaire (SMQ) and the Child Behavior Checklist (CBCL) were administered, and results were compared with pre-treatment results. After 6 months of psychomotor treatment, results showed that children showed a significant reduction among CBCL scores, SM symptoms in all situations (school, family, and social situations), and in SMQ total score.

The sample size of the study was adequate, but a large number of children (79) were excluded from the study. Due to the extensive exclusion criteria, it would be difficult to generalize results to children with comorbid disorders. In addition to participants being excluded, researchers also lost a number of participants during the course of the study due to follow up, medical trouble, and failing to correctly complete the therapy. Researchers chose to exclude these individuals from the study analysis, though it would have been a better effectiveness measure to include all participants in the analysis.

The study methodology included many strengths. The participants were randomly assigned to treatment groups, which eliminated potential treatment bias. The authors included detailed participant characteristics including the participants' age range, sex distribution, ethnicity, and socioeconomic class. The researchers found the two treatment groups to be comparable for age and sex, which increases the reader's confidence that any differences in results between groups are due to the intervention and not participant characteristics. The intervention goals were well described, but not the intervention protocol itself. The methods specified that all of the clinicians performing the intervention shared the same protocol,

but this protocol was not included in the article. This makes the results difficult to replicate, challenging for other clinicians to learn from the paper and apply it to their work, and difficult for clinicians of other disciplines to know what is involved in the therapy sessions.

Due to these factors, this study offered a moderately suggestable level of evidence. This study supports the effectiveness of psychomotricity as a therapy for SM, but further research is needed to clarify the long term effects of this treatment.

Multimodal Approaches:

Klein, Armstrong, Skira, and Gordon (2017) conducted a clinical trial that investigated Social Communication Anxiety Treatment (S-CAT) as an intervention for children and families with SM. Forty children with SM attended 3 therapy sessions focusing on cognitive behavioural strategies and parent education held once every 3 weeks. Several assessment measures, including the SMQ, CBCL, and family compliance ratings, were completed at baseline, at each treatment session, and at the completion of treatment. Following 9 weeks of treatment, children showed significant gains in speaking frequency on all SMQ items and decreased levels of anxiety and withdrawal as reported by parents on the CBCL.

The authors utilized a number of methodologically sound measures to increase the validity of this study. The researchers were not involved in treatment development or therapeutic delivery, which enabled them to act as blind and unbiased reviewers during data collection, analyses, and interpretation. The study included fidelity checks (i.e., progress notes and video recordings reviewed by research assistants), which helps increase the confidence in the accuracy of the results. The goals and procedures used in the S-CAT intervention were well described, and the authors also provided a website link where the reader could read further about the S-CAT treatment sessions if desired. This would make it easy for future studies to replicate, or for other therapists to use this approach in practice with patients.

When analyzing the study results, researchers completed appropriate statistical measures, including ANOVAs and *t*-tests. The researchers collected a variety of measures, including coded progress notes written by the therapist, and ratings of anxiety, withdrawal, and language use in different environments by the parent and teacher pre- and post-treatment. Collecting outcome measures from a variety of individuals in multiple contexts helps to support the effectiveness of the treatment and the ability for treatment effects to generalize to everyday situations.

This study utilized a medium sample size; however, the population of SM children and their

parents was geographically limited to the Mid-Atlantic region and only included families seeking free services offered at a private clinic. A second limitation of the study was the absence of a control group, which would have been an effective way to control for possible confounds.

Overall, this study provided a highly suggestive level of evidence based on the type of design, the baseline established, and the statistics used. This treatment should be studied further, specifically in a randomized control trial, to demonstrate the generalizability of S-CAT to other patients and therapists.

Discussion

Critical review of recent literature (2016-2018) revealed that the strongest evidence for efficacy of SM treatment was through behavioural approaches, such as CBT studied by Oerbeck and colleagues (2018). Additionally, multimodal approaches, which in Klein et al.'s study (2017) combined behavioural and systems techniques, have shown promising results.

However, in all of the studies reviewed, S-LPs were not involved in the research or the implementation of treatment approaches for SM. Behavioural, psychodynamic, and pharmaceutical approaches are out of S-LPs' scope of practice. Systems and multimodal approaches can be within S-LPs' scope of practice, but this was not shown in the recent literature. Nonetheless, children with SM appear on S-LPs' caseloads, as S-LPs are trained to address the social communication difficulties that accompany SM (Johnson and Wintgens, 2015). Therefore, it is important that S-LPs be involved in the treatment of SM (Johnson and Wintgens, 2015) and must be actively involved in research to support the effectiveness of their intervention.

Few studies have investigated the efficacy of speech-language therapy in the treatment of SM, and fewer have compared this method to existing intervention approaches. Research prior to 2016 has compared the efficacy of speech-language therapy to pharmacology to the multimodal approach of combining speech-language therapy and medication for treatment of SM (Manassis & Tannock, 2008). Results indicated that medication alone showed encouraging gains for participants with SM, but that the sample was too small to rule out the possibility of combining medication and therapy resulting in even greater reduction of SM symptoms (Manassis & Tannock, 2008). As S-LPs continue to provide treatment, more research similar to that of Manassis and Tannock (2008) needs to be conducted to evaluate the efficacy of speech-language therapy compared to other approaches. Without this research of randomized control trials comparing different treatment approaches and varying

combinations of treatment approaches, gold standard treatment for SM cannot be established.

In other parts of the world, such as the United Kingdom, S-LPs play an essential role in the treatment of SM. According to Johnson and Wintgens (2015), S-LPs and therapists play an essential role, either directly or consultatively, in the assessment, diagnosis, and treatment of SM. Johnson and Wintgens (2015) describe how speech and language services across the UK have put in place, or are in the process of developing, evidence-based SM Care Pathways which emphasize early intervention and home-school liaison, leading to an excellent prognosis for children with SM. As S-LPs play such an important role in SM care in other countries, it is important that S-LPs in Canada advocate for the valuable role they could play on the SM intervention team.

Conclusion

Recent evidence supports the behavioural approach as an effective intervention for SM. One of the three behavioural approaches analyzed in this review offered a compelling level of evidence. However, this approach, along with psychodynamic and pharmacological approaches, are outside an S-LP's scope of practice. It is recommended that further research be conducted to confirm the most effective treatment methods for SM. S-LPs need to be actively involved in research in order to determine the effectiveness of their role in the treatment of SM. In future studies, a between-groups design should be utilized in order to compare existing treatment approaches (i.e., pharmacological, behavioural, psychodynamic) to multimodal approaches that include S-LP service and with independent S-LP intervention.

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