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When Behavioral Intervention Failed: A Single Case Report of a Successful Treatment of Selective Mutism Using Fluoxetine

Poling Bork[1,*], Donna Snyder[2]

[1]Department of Graduate Studies, Brock University, St. Catharines, L2S 3A1, Canada
[2]Pediatrician, Niagara Falls, L2E 6G1, Canada

Abstract This article demonstrates the remediation of selective mutism (SM) with fluoxetine treatment on a male (9 years 2 months), following unsuccessful behavioral and psychosocial interventions. Although it is premature to consider pharmacological treatment as a more effective treatment than behavioral or other psychotherapeutic modalities for persistent SM, the results from this study are consistent with prior research demonstrating the efficacy of pharmacological intervention, specifically fluoxetine, in the treatment of SM.

Keywords Selective Mutism, Behavioral, Psychosocial, Pharmacological Intervention, Fluoxetine

1. Introduction

Selective mutism (SM) is a childhood condition that is characterized by the persistent failure to speak in specific social situations such as school, despite speaking to immediate family members at home [1]. This failure to speak may persist into adolescence and adulthood if left untreated.[2] The onset age for SM is between 2.7 years and 4.1 years,[3] and more females are affected by SM than males.[4] Due to its relatively low prevalence rates of 0.71%,[5] many health care professionals are unfamiliar with SM and therefore unable to provide proper help.[6,7] As a result, the lag time can be as long as four years before SM sufferers receive treatment/intervention.[8]

Although the etiology of SM remains controversial,[8,9] many researchers believe SM is an anxiety disorder[5,8,10] that closely relates to social phobia[6,11] and specific phobia – the fear of expressive speech.[12] The proposal of SM as a variant of anxiety disorder provides a pragmatic theoretical approach for assessment, treatment and research. Indeed, literature documents the successful treatment of SM as an anxiety disorder using behavioral[13] and pharmacological modalities.[14-16] Specifically, applied behavioral techniques such as self-modeling, shaping, contingency management, systematic desensitization, and stimulus fading have become the most frequently used method of intervention for SM.[17,18]

* Corresponding author:
mentor@brocku.ca (Poling Bork)
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SM has been found to be resistant to treatment with documented interventions that extend over several years.[17] Early intervention is of paramount importance, to avoid the mutism behavior strengthening, and becoming entrenched over time.[6, 19] In fact, few children receive intervention during early primary school years. Often appropriate treatment is delayed until after the child turns 7. As well, Schwartz and Shipon-Blum[20] contends that only 30% to 40% of children older than 12 who are diagnosed and treated appropriately will speak to a wide circle of schoolmates. Despite intervention, there are many SM cases that are intractable to conventional psychosocial intervention.[17] As such, medications that are efficacious for anxiety-related disorders may be needed to target the serotonergic dysfunction (an underlying neurobiological deficit) in children.[3]

2. Pharmacological Treatment for Selective Mutism

Selective serotonin reuptake inhibitors (SSRIs) are effective in treating children with selective mutism because SSRIs increase brain levels of the synaptic neurotransmitter serotonin.[20] Thus, such increases in serotonin have been found to have considerable beneficial effects on various forms of anxiety.[21,22] Schwartz et al.[6] predicates that some parents and physicians are reluctant to include medication in the treatment of SM due to its possible adverse effects, yet, consideration of pharmacological therapy is warranted if a child does not make sufficient improvement with behavior therapy after 3 to 6 months, or the mutism causes significant impairment.[3,6] Pharmacological treatment has

been found to be an effective intervention for persistent SM, which has been unresponsive to behavioral and psychosocial interventions. Specifically, fluoxetine, a type of SSRI, appears to be the most promising pharmacological agent for SM.[6, 23]

2.1. Rationale for Treatment with SSRIs for Individual with Selective Mutism

Fluoxetine (Prozac©), a type of SSRIs, is frequently selected for child and adolescent anxiety disorders. [23-25] Since SM is conceptualized as an anxiety disorder, it is most often treated pharmacologically with SSRIs.[6] As a group, SSRIs offer a more tolerable side effect profile than the tricycle antidepressants and Monoamine Oxidase Inhibitors.[3] Schwartz et al. [6] noted that SSRIs are not only effective for SM, but are considered by some child psychiatrists to be safe if used carefully. Research Unit on Pediatric Psychopharmacology Anxiety Study Group[26] has documented the safety and efficacy of SSRIs in treating children with various anxiety disorders, depression, and paediatric obsessive-compulsive disorder.[3, 27]

Among SSRIs, fluoxetine (i.e., Prozac) has been found to be effective for SM.[23,28]Carlson et al. [23] identified 12 of 21 (57%) pharmacological treatments of SM used fluoxetine. Most patients tolerated the medication well, and their speaking (and or social) behavior improved significantly following the treatments. Fluoxetine does not need to be tapered in small decrements over several weeks to avoid withdrawal syndrome (e.g., dysinhibition, emotional lability, nausea, and headaches) that is caused by an abrupt discontinuation of a typical SSRI medication.[20]

Further, not all SSRIs (e.g., citalopram, escitalopram, fluoxetine, fluvoxamine, paroxetine, and sertraline) are targeted specifically for use with children. Although literature has documented treatments of SM with paroxetine,[15] and citalopram,[29] only fluoxetine (Prozac©), fluvoxamine (Luvox©), and sertraline (Zoloft©) have been approved by the Food and Drug Administration (FDA) for use in children. [30]

It is important to point out that the success of interventions in SM with an SSRI is inversely correlated with age, as it is confirmed that a younger age predicts a better outcome.[31] As well, although fluoxetine need not be tapered in small decrements to avoid withdrawal symptoms, evidence has suggested that this period of time "allows children to consolidate their progress and to expose themselves to the anxiety-provoking situations that previously elicited the symptom".[3]

3. The Case Study Overview

This study reports on a child diagnosed with SM who was nonresponsive to behavioral and psychosocial intervention, but was successfully treated with fluoxetine. The patient exhibited symptoms of SM (only whispered to two peers) when he entered pre-school at 2 years 10 months. At the preschool level, the child was not diagnosed by the family physician with SM. Subsequently, the child entered grade one in a different school at the age of 5 years 10 months and became completely mute inside the school. The child was diagnosed as suffering form SM at 6 years 3 month. A referral was made by the pediatrician, to a clinician who had experience in treating children with SM using behavioral therapy. Using a framework similar to Dow, Sonies, Scheib, Moss, and Leonard's[32] school-based multidisciplinary intervention, an outpatient treatment strategic plan was set up using predominately behavioral techniques (i.e., systematic desensitization, stimulus fading, contingency management), and was implemented in and out of school involving a management team consisting of parents and teachers.[32]

3.1. Behavioural Intervention

The boy's supportive peers in school were identified. Classroom seating arrangements were made so that the child was situated away from the teacher – the most anxiety provoking figure, and was sitting next to someone he was comfortable with to foster speaking opportunities. During the first year, the boy's mother came in each day as a "teacher's helper" to encourage the child to whisper to her inside the classroom. Then, due to work, instead of spending every day at school, the mother and father took turns and came in each morning before the class to conduct activities with the boy that would allow the opportunity for the child to practice speech inside school; starting in a private location (i.e., empty library) and slowly moving into the classroom. In addition, the peers whom the boy felt comfortable with were invited 2 to 3 times weekly for after school play dates at the boy's home. Using the self-modeling approach, a tape recorder was used to tape the child's speech to enable him to "talk" to his friends, for "show and tell" (while the boy squeezed his mother's hand and hid behind her), and for reading evaluation purposes.

Although walkie-talkies were also made available inside school and at home, the boy was unable to speak through them after numerous attempts. After 12 months of behavioral intervention, the boy seemed to be more comfortable around his peers, but was still unable to use walkie-talkies or other medians (e.g., mother, telephone), to communicate with his friends. The mother consulted the clinician and family physician about adding fluoxetine into the treatment, which both promptly rejected due to possible adverse affects. The behavioral treatment continued for another 6 months and the boy was still not progressing. Frustrated, the mother also enrolled the child in art/play therapy, psychodynamic therapy; even hypnosis sessions were added. After 24 months of failed behavioral and psychosocial intervention, the boy began to receive pharmacological treatment with fluoxetine by the pediatrician who diagnosed him with SM.

3.2. Pharmacological Treatment

The introduction of fluoxetine treatment (1.25mg/day) was well tolerated and was gradually increased to 10 mg/day at week 21. At that time the boy was able to speak to a friend outside of school without eye contact (behind friend's back) and communicate comfortably with 2 classmates through a walkie-talkie, in and out of school. In addition, the boy was also able to use a walkie-talkie to communicate with his classroom teacher. Significant improvements in other social situations (i.e., spoke directly to a younger cousin, ordered food at restaurants, spoke to swimming instructor) were also observed. At week 28, the boy continued to make further progress and spoke directly to 5 more friends (2 from school) at home, but only whispered to them inside the school. As the dosage increased to 12 mg/day at week 31, so too did the number of people the boy spoke to. Again, the boy tolerated the medication well, and no side effect was reported. At week 36, the boy reportedly spoke into the microphone giving farewell wishes to the retiring principal in front of the whole school. Evidently, the boy no longer met the criteria for SM and was talking freely to everyone at that point. Treatment effects were maintained at the 1-year follow-up. At the 2-year follow-up, according to teachers, the boy reportedly was too talkative in the class, and was able to speak to everyone. The3-year follow-up showed the boy was no longer as talkative inside a new school, and was adjusting well while SM remained in remission.

4. Implications for Pharmacological Treatment

Despite successful treatment of pharmacotherapy in SM, treatment with medication alone is ill-advised.[28] Medication should be considered when behavioral therapy shows inadequate response, when the mutism is severe and has comorbid symptoms, or if the child is older.[3, 6, 29]As mentioned, medication is associated with possible adverse effects. Although no side effect was reported in this case study, common adverse effects of SSRIs include nausea, abdominal pain, sleep disturbance, change in appetite and weight, lightheadedness, and fatigue. Other adverse effects, although uncommon, include serious depression, irritability, temper tantrums, and mania,[20] allergic reactions to the drug and serotonin syndrome (confusion, muscle spasms, profuse diaphoresis, hyperthermia) and autonomic nervous system instability. In addition, there have been reports of epistaxis or bruising in children,[33] suicidal ideation, [30, 34, 35]growth attenuation in children[36] as well as sudden cardiac deaths among children who are taking SSRIs.[37]As such, the American Psychiatric Association has published a guideline that children who are taking SSRIs must be monitored closely and should be seen by their primary physician weekly for 1 month, then every other week for another month.[6]

5. Limitation of the Study

This study has several methodological limitations: 1) Instead of using standardized instruments to measure changes, this study used descriptions to elucidate behavioral change; 2) Pharmacological treatment was conducted along with concurrent behavioral and psychological interventions, therefore it is possible that pharmacological treatment "enhanced" the treatment effect of the psychosocial and behavioral interventions or vice versa; 3) Due to the rarity of this disorder, this study joins the majority of pharmacological treatments of SM that are case studies.[23] Although a controlled study using a larger sample of children with SM can be difficult to conduct, a well designed study using standardized instruments to measure change across multiple settings, and placebo-controlled, double-blind design studies that are more methodologically sound are needed to further establish the efficacy of pharmacological treatment of SM.

6. Discussions

Findings in this case study is consistent with the majority of pharmacotherapy treatments of SM in several ways: 1) The onset age of SM was within the range of 2.7 years and 4.1 years as reported by Garcia et al. in 2004;[3] 2) A lack of public awareness of SM resulted in the boy being diagnosed 3 years later; 3) The boy was unresponsive to conventional behavioral and other psychosocial interventions for 2 years; 4) Due to possible adverse effects the family physician and the SM clinician (a child psychologist) rejected the option of pharmacological treatment after 2 years of failed intervention; 5) Fluoxetine provided a rapid resolution of SM when other methodologies failed; 6) The patient tolerated fluoxetine well with no reported side effects.

Because the pediatrician was unfamiliar with treating SM with fluoxetine, the medication was initiated at the lowest dosage (1.25 mg/day) and increased very gradually throughout treatment. This is consistent with other pediatricians who administer fluoxetine beginning with 1.25 mg per day and gradually increasing the dose to a maximum of 20 mg per day (Schwartz & Shipon-Blum, 2005). In contrast, psychiatrists who have experience with SSRIs often prescribe higher doses of fluoxetine, up to 60 mg per day,[20] because children metabolize the drug faster than adults and thus require full doses of antidepressants.[38] Indeed, studies have demonstrated successful fluoxetine treatments of SM with an initial dosage of 20 mg/day[16] and increased up to 60 mg/day at week 9[31] with minimal side effects. Due to the fact that our patient was treated very cautiously and gradually, whether or not he could have overcome SM much sooner with a stronger dosage of medication remains unknown. Further methodology and sound research is needed to explore this approach.

Despite limitations, this study adds to the current knowledge base in the efficacy of medication treatment for SM. While pharmacological treatment was conducted along with behavioral and psychosocial treatments, and given that

the chronic symptom of mutism persisted several years and failed to respond to multiple intervention approaches, it is safe to conclude that the medication was at least partly, if not wholly, responsible for the changes in speech.

7. Conclusions

This study demonstrates the importance of medication treatment for children who have persistent SM and are unresponsive to psychosocial and behavioural interventions. It also demonstrates fluoxetine is an effective and well-tolerated treatment option for SM for this child. However, there are clear implications to treating SM with medication due to the possible adverse effects, as well as the stipulation of using medications that are not approved by the Food and Drug Administration for use in children. Therefore, it is important to make accurate judgments to weigh the long-term benefit against the magnitude of side-effects caused by medication, and to monitor children closely when treating them with medication.

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