



Treatment Complexity in a Twin Adolescent Pair with Selective Mutism: A Case Report

Özlem Sireli^{1,a,*}, Seyda Müberra Pakoz^{1,b}, Merve Soyhan^{1,c}, Cansu Mercan Işık^{1,d}, Elif Abanoz^{1,e}, Ayla Uzun Çiçek^{1,f}

¹ Department of Child and Adolescent Psychiatry, Sivas Cumhuriyet University, Faculty of Medicine, Sivas, Türkiye

*Corresponding author

Case Report

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ABSTRACT

Selective Mutism (SM) is a disorder characterized by the individual's persistent inability to speak in specific social situations (e.g., school) where speech is expected, even though the individual is able to speak in other situations. It is a rare anxiety disorder and is often seen in children between the ages of 4-8. The presence of SM in family members is important in terms of both etiology and clinical course. While family burden is a factor that increases the incidence of SM, according to some research results, the clinical symptoms of twin cases diagnosed with SM are more severe than those of non-twins. A positive family history negatively affects the prognosis of SM and increases resistance to treatment. In this article, the diagnosis and treatment process of 14-year-old monozygotic twin adolescents who have been followed up with the diagnosis of SM for a long time will be discussed. The main treatment methods for SM are medication and psychotherapy. It is known that psychotherapeutic interventions in particular vary depending on individual differences. The most important purpose of this article is to draw attention to the differences in treatment interventions of twin SM cases and to evaluate the clinical features of the cases in the light of the literature.

Keywords: Selective mutism, monozygotic twin, adolescent

Selektif Mutizimli Ergen İkiz Çiftte Tedavi Karmaşıklığı: Bir Olgu Sunumu

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ÖZET

Selektif mutizm (SM), bireyin başka durumlarda konuşuyor olmasına karşın, konuşmasının beklendiği özgül toplumsal durumlar (örn. okul), sürekli bir biçimde, konuşamıyor olması ile karakterize bir bozukluktur. Nadir görülen bir anksiyete bozukluğu olup sıklıkla çocuklarda 4-8 yaş aralığında görülmektedir. Aile bireylerinde SM varlığı hem etyoloji hem de klinik gidişat açısından önemlidir. Aile yükünlüğü SM'nin görülme sıklığını artıran bir faktör iken bazı araştırma sonuçlarına göre, SM tanısı olan ikiz olguların klinik semptomları ikiz olmayanlara göre daha şiddetlidir. Aile öyküsünün pozitif olması SM'de klinik gidişatı olumsuz etkilemekte, tedaviye direnci artırmaktadır. Bu yazıda, 14 yaşlarında, uzun süredir SM tanısı ile takip edilen monozygot ikiz ergenlerin tanı ve tedavi süreci ele alınacaktır. SM'de başlıca tedavi yöntemi ilaç tedavisi ve psikoterapidir. Özellikle psikoterapötik müdahalelerin bireysel farklılıklar doğrultusunda çeşitlilik gösterdiği bilinmektedir. Bu yazının en önemli amacı, ikiz SM olgularının tedavi müdahalelerindeki farklılığa dikkat çekmek ve olguların klinik özelliklerini literatür eşliğinde değerlendirmektir.

Anahtar Kelimeler: Selektif mutizm, monozygotik ikiz, ergen

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^a ozlemsireli@cumhuriyet.edu.tr
^c merve.soyhan1996@gmail.com
^e elifabanoz_17@hotmail.com

^b 0000-0002-5549-4154
^d 0009-0001-9238-5256
^f 0000-0002-9214-4735

^b pakozseyda48@gmail.com
^d dr.cansumercan@gmail.com
^f aylauzun@cumhuriyet.edu.tr

^b 0009-0004-4665-0962
^d 0000-0001-9437-3024
^f 0000-0003-2274-3457

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Introduction

Selective Mutism (SM) is an anxiety disorder that is relatively rare compared to other anxiety disorders and it is characterized by the inability to speak in specific settings and situations where speech is anticipated, despite having the ability to speak¹. Although precise percentages have not been established, research has reported that the frequency of selective mutism ranges from 0.11 to 2.2% and it is more common in girls than in boys^{2,3}. Psychiatric comorbidities, particularly social phobia, specific phobia, and separation anxiety disorder, are extremely prevalent among SM patients^{3,4}.

Long-term follow-up research on the clinical trajectory of SM is scarce. In a study, the average disease duration for SM was found to be 9 years⁵. In general, it is suggested that in most untreated SM cases, mutistic symptoms gradually go away and that during adolescence and early adulthood, the majority of cases do not fulfill the diagnostic criteria for SM⁶. A poor prognosis is linked to advanced age, initial disease severity, and a family history of SM⁷.

Although it has been reported that SM is more common in twins, it is unclear whether this is a referral bias⁸. Furthermore, it has been noted that the clinical symptoms of SM in twins are more severe than those in singletons^{8,9}. This article aims to discuss the follow-up and treatment process of a pair of female monozygotic adolescent twins who have had SM for a long time. Written informed consent for the presentation and publication of the case was obtained from the patients and their parents on the condition that the patients' anonymity must be preserved.

Description and Presentation of Cases

Fraternal twin female patients, aged 14 years and 9 th-grade students, applied to our clinic accompanied by their mothers and at their mother's request, not speaking in social situations like school, friends, and foreign environments, and extreme shyness and timidity. In one-on-one interviews with different psychiatrists, nonverbal communication (making eye contact, using gestures and facial expressions, shaking the head) was achieved with both adolescents, but verbal communication was not established. Due to the patients' limited verbal communication, the mother provided the majority of the anamnesis information.

It was learned that the patients spoke on time and fluently, without any delay in speech and other developmental stages, and their "not talking to strangers" problems were initially noticed in the first grade of elementary school. The mother stated that until that date, as parents, they had observed that children did not talk to anyone when there were strangers at home, when they went out for a visit, or in other social environments, but they did not consider this as an abnormal situation because they spoke normally and fluently when there were no outsiders present at home (except for their grandmother, who they saw a lot). Without any preschool instruction, when the children entered elementary school together, in the same classroom, at the same desk, they

did not experience separation anxiety. The mother were the children's primary caretakers, and they grew up in an isolated household setting with no friends. During the first half of first grade, the teacher observed that the sisters talked only to each other and did not talk to anyone including the teacher herself and their classmates; when they asked them questions or wanted to communicate with them, they acted as if they could not hear them at all; they did not even make eye contact; they did not participate in games with their classmates, and they were remarkably shy and introverted, and thus recommended that they receive a psychiatric evaluation from child and adolescent psychiatry. They had no triggers, stressful/traumatic life events, or stressors linked to their "not talking to strangers" problems. The children who were evaluated for the first time by a child psychiatrist in the second half of the first grade, it was reported that their mental development was normal, and considering the diagnosis of selective mutism, they were started on fluoxetine 5 mg/day medication. However, family members did not start drug treatment and did not go to child and adolescent psychiatry follow-ups because they thought that these problems would spontaneously resolve when they grew up, and they were worried about the side effects of drug treatment.

When the children started the second grade of elementary school, their classroom teacher changed and a male teacher whom the mother described as authoritarian, rude, rather harsh, and emotionally detached took over. The mother claimed that at that time, her children were more timid than ever at school and were reluctant to attend school. The classroom teacher again referred the children for a psychiatric evaluation due to their lack of verbal and nonverbal communication with him and their classmates. They next saw a different child and adolescent psychiatrist, who once more started them on 5 mg of fluoxetine per day. The dosage of fluoxetine was gradually increased to 20 mg/day during the follow-ups, and risperidone 0.25 mg/day was added to the treatment regimen. The mother mentioned that they received treatment with fluoxetine 20 mg/day and risperidone 0.25 mg/day for a year and that during this period, the children communicated only non-verbally with the classroom teacher and classmates with whom they were not close, and spoke with one or two classmates, albeit in whispers. Later, with the arrival of summer vacation, this treatment regimen was stopped by the parents, and the children were left without treatment for approximately 6 months. Throughout the subsequent processes, various medication therapies were selected for the children who were assessed by numerous child and adolescent psychiatrists before applying to our clinic (including selective serotonin reuptake inhibitors [fluoxetine, sertraline, escitalopram, fluvoxamine], imipramine from tricyclic antidepressants, mirtazapine from atypical antidepressants, antipsychotics [risperidone, aripiprazole, paliperidone, amisulpride] and hydroxyzine from antihistamines). Furthermore, even though many psychiatrists recommended that siblings attend separate classes, the children never attended separate classes because they refused to do so. In summary, as a result of the approximately 8-year

treatment process, which was not seriously interrupted, it was learned that the children could only communicate verbally and non-verbally at a limited level with very a few friends (one or two) with whom they were close, outside the home and family environment, and that they still did not talk to other people.

It was reported that children who were born as fraternal twins via planned and cesarean section had no health issues during or after birth and completed the neuromotor development stages on schedule. Except for selective mutism, the children's medical histories were free of significant mental or medical illnesses, and their neurological examinations revealed no pathology. According to family history, the mother was extremely quiet, timid, and introverted at a young age, and she became very excited when speaking in foreign environments, as well as having personality traits such as shyness and timidity. Apart from this, there was no feature in their family history, including speech-related disorders. Their 19-year-old brother and father were described as talkative, energetic, and outgoing.

When patients applied to our clinic, they had not received treatment in about 9 months. Follow-up and treatment of both cases continued with weekly meetings in the first month of treatment, and every two weeks thereafter. The therapy was implemented in the form of individual and child-focused parent interviews. Two different child psychiatrists conducted separate psychiatric interviews with the cases, and both noted that both cases were extremely anxious. During the psychiatric examination, only non-verbal communication was possible in both cases, and written communication was also provided in one case. Clinically, the intelligence of the cases was within normal limits and it was ascertained that their academic performance was good. The sentence completion test's contents were mostly about the "inability to speak"; there were no depressive items. Neither of them was persuaded to draw. The home video recordings yielded they were fluent in oral language and spoke freely. Autism spectrum disorders were excluded because the patients maintained normal communication and interaction within the family and did not have limited or repetitive thoughts and behaviors, and psychosis was excluded because they did not exhibit delusions, hallucinations, or other psychotic symptoms. Both cases were diagnosed with selective mutism and social phobia, and they were started on fluoxetine 20 mg/day and aripiprazole 5 mg/day, which had previously been shown to be the most effective for them. Two different child psychiatrists separately conducted behavioral treatment-oriented interviews with the cases, which included practice/exposure-based tasks, social skills acquisition, and new tasks. Furthermore, patients were now asked to attend separate classes at school, and 3 weeks of lorazepam treatment was added when their classes were separated. During the follow-ups, after the patients separated their classes at school, they established effective nonverbal communication with their teachers and classmates, as well as effective verbal and nonverbal communication with their close friends, began to hang out with their close friends during breaks, and were able to participate individually in group activities such as music

and role-playing. Family members also commented positively on the cases' ability to form social relationships. During the outpatient clinic follow-up, verbal communication was possible in both cases, albeit in a low voice and whisper, but although the patients' social functionality improved, the expected symptomatic improvement had yet to be achieved. It appeared that the severity of selective mutism symptoms decreased as classes were separated at school. The cases were followed up and treated for about 8 months, but then the mother voluntarily ended the psychiatric interviews, stating that her children had recovered, albeit not completely.

Discussion

In this article, a pair of monozygotic female adolescents diagnosed with SM were presented, in whom psychotherapeutic and behavioral interventions were inadequately applied and the disease process was protracted, even though they received sufficient treatment support in the form of medication assistance. This case report underlines the importance of social isolation and a family history of SM and anxiety disorders, which contribute to treatment resistance and reinforcement and maintenance of mutism, despite strong drug support.

SM is an uncommon anxiety disorder that is strongly associated with other anxiety disorders, especially social anxiety disorder^{3,4}. Most studies agree that girls are more likely than boys to develop SM, and some research has shown that SM is more common in twins and that their clinical characteristics are more severe^{2,3,8,9}.

The frequency and clinical presentation of SM in twins are poorly understood. However, studies indicate that in addition to strong genetic characteristics in twins, there are also environmental factors such as social isolation that strengthen the continuation of mutism and other clinical characteristics^{8,9}. There are surprisingly very few studies or case reports in the literature regarding the occurrence of SM in twins⁸⁻¹¹, and according to some research, twins are disproportionately common in SM sufferers¹¹. It is asserted that the twins struggle to grow apart and develop their own identities and that this is made worse by the fact that neither the parents nor the larger community can recognize the twins as two distinct people. In addition to the inability to develop an individual identity, it has been emphasized that twins exhibit mutual mirroring, which results in social isolation, the persistence of SM symptoms, and resistance to treatment⁹. We observed that our twin pair with SM had difficulty separating from each other, although they were encouraged to separate, that they could not demonstrate social skills because they could not gain an individual identity, and that they continued to see each other as the same unit. Thus, it was thought that their close interaction with each other and the use of the language they created among themselves hampered the individualization and socialization processes and disrupted ego development, so they were asked to have separate classes. Indeed, once our cases were separated into different classes and the twins' unity was broken, their verbal and nonverbal communication skills improved probably due to their social isolation

barriers decreased. Furthermore, the fact that the mother of the twin pair we present exhibits personality traits such as shyness, introversion, and timidity, although not severe enough to meet social phobia, may contribute to a lack of verbal communication in the family, social isolation, and behavioral inhibition. It is also clear that the mother's personality traits can cause a genetic predisposition for both SM and social phobia diagnoses in her twins. These factors can lead to the severity of SM symptoms remaining unabated and/or an inadequate response to treatment. In addition, negative prognostic factors such as the relatively older age of our cases and their high disease severity from the beginning may also explain the persistence of symptoms and poor response to treatment. Again, previous studies have shown that SM is strongly associated with other anxiety disorders, especially social anxiety disorder, and that almost all cases of selective mutism are diagnosed with social phobia^{2-6,11}. Similarly, our cases were diagnosed with social phobia along with SM, and we think that this might contribute to the relatively poor prognosis.

Several approaches are used in the treatment of selective mutism, and it is generally recommended that pharmacological therapy and psychotherapeutic interventions be applied together. Psychotherapeutic interventions should involve the patient, his/her family, and his/her surroundings. It is advised that mental health professionals, school counselors, and speech therapists be part of the therapy team^{3,4}. However, there is a dearth of evidence-based information regarding the care and treatment of twins with SM, and this data mostly originates from case reports⁸⁻¹⁰. There is an argument that suggests twins should be separated for treatment purposes to promote and enhance individuality, even if this is not supported by study findings⁸⁻¹⁰. It is highlighted that separating not only twins with SM but also twins with speech difficulties in environments such as schools will have extremely positive effects on speech and language skills¹². We implemented behavioral therapy-based interventions to shatter the twins' unity, promote their socialization and individualization, assist their self-differentiation, and further their ego development. For each twin pair's assessment, treatment, and follow-up, we selected different therapists. We observed that various behavioral strategies we used had generally positive outcomes. In addition to behavioral interventions, pharmacological treatments, especially selective serotonin reuptake inhibitors, also have an important place in the treatment of SM. Because of this, we started our twin pair cases on fluoxetine and aripiprazole, two psychotropics that were most beneficial in the past and we continued drug treatments without significant side effects. In conclusion, observing rapid changes in their communication and interactions as a result of the "separating their classroom and social environments" technique applied for treatment purposes to increase the individuality of our cases, as well as drug therapy suggests that separating the environments of twins with SM during their treatment is an extremely important and necessary approach.

Conclusion

This study adds to the scant body of literature on twin pairs with SM therapy. The treatment method of the twin pair with SM that we present here demonstrates the complexity of SM treatment in twins, and points out the value of separating their social settings —like the school environment— to strengthen their individual unique identities and individuality. It was thought that our patients' interaction with each other and the use of the language they created among themselves led to the development of a symbiotic relationship over a long period, which contributed to the persistence of symptoms and the emergence of treatment resistance. Indeed, meaningful progress was made by arranging the twin pair's classroom and social surroundings to be separate and providing them with chances for separation and individualization.

References

1. American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders (DSM-5). 5th ed. Arlington (VA): American Psychiatric Association; 2013.
2. Hua A, Major N. Selective mutism. *Curr Opin Pediatr*. 2016;28(1):114-120. doi:10.1097/MOP.0000000000000300
3. Dogru H, Ucuz I, Uzun Cicek A, Comertoglu Arslan S. Clinical characteristics according to sex and symptom severity in children with selective mutism: a four-center study. *Nord J Psychiatry*. 2023;77(2):158-164. doi:10.1080/08039488.2022.2146748
4. Viana AG, Beidel DC, Rabian B. Selective mutism: a review and integration of the last 15 years. *Clin Psychol Rev*. 2009;29(1):57-67. doi:10.1016/j.cpr.2008.09.009
5. Remschmidt H, Poller M, Herpertz-Dahlmann B, Hennighausen K, Gutenbrunner C. A follow-up study of 45 patients with elective mutism. *Eur Arch Psychiatry Clin Neurosci*. 2001;251(6):284-296. doi:10.1007/pl00007547
6. Smith-Schrandt HL, Ellington E. Unable to Speak: Selective Mutism in Youth. *J Psychosoc Nurs Ment Health Serv*. 2018;56(2):14-18. doi:10.3928/02793695-20180122-04
7. Oerbeck B, Overgaard KR, Stein MB, Pripp AH, Kristensen H. Treatment of selective mutism: a 5-year follow-up study. *Eur Child Adolesc Psychiatry*. 2018;27(8):997-1009. doi:10.1007/s00787-018-1110-7
8. Segal N. 'Two' quiet: monozygotic female twins with selective mutism. *Clin Child Psychol Psychiatry*. 2003;8(4):473-488. doi:10.1177/13591045030084005
9. Sharkey L, Mc Nicholas F. Female monozygotic twins with selective mutism—a case report. *J Dev Behav Pediatr*. 2006;27(2):129-133. doi:10.1097/00004703-200604000-00008
10. Tachibana R, Nakamura K, Schichiri K, et al. Elective mutism in twins. *Jpn J Child Adolesc Psychiatry*. 1982;23:277-286.
11. Dummit ES, Klein RG, Tancer NK, Asche B, Martin J, Fairbanks JA. Systematic assessment of 50 children with selective mutism. *J Am Acad Child Adolesc Psychiatry*. 1997;36(5):653-660. doi:10.1097/00004583-199705000-00016
12. Mora G, Devault S, Schopler E. Dynamics and psychotherapy of identical twins with elective mutism. *J Child Psychol Psychiatry*. 1962;3:41-52. doi:10.1111/j.1469-7610.1962.tb02037.x