Side Effects, Treatment Resistance and Reverse Intermetamorphosis Syndrome: A Case Report and Theoretical Review

Fatih Yığman, MD,[1] Süheyla Doğan Bulut, MD,[1] and Canan Efe, MD[2]

[1] *Health Sciences University, Dışkapı Yıldırım Beyazıt Research and Training Hospital, Psychiatry Clinic, Ankara, Turkey*
[2] *Gölbaşı Şehit Ahmet Özsoy State Hospital, Psychiatry Clinic, Ankara, Turkey*

ABSTRACT

Background: Delusional misidentification syndromes include a group of beliefs that the person or objects around the patient change or have changed. There are four main types of delusional misidentification syndromes: Capgras, Fregoli, intermetamorphosis, and the syndrome of subjective doubles. Intermetamorphosis is a delusional misidentification syndrome associated with agnosia and characterized by the patient believing that a person or people around him/her have changed both physically and psychologically. Reverse intermetamorphosis is a subtype characterized by delusional misidentification that occurs in the person himself.

Case report: This case report presents a patient with schizophrenia additionally diagnosed with reverse intermetamorphosis syndrome and who had side effects and resistance to many treatment options. She finally responded positively to electroconvulsive therapy (ECT).

Conclusion: Recognizing delusional misidentification syndromes is difficult for many clinicians. In this case, we evaluated the approach to cases of reverse intermetamorphosis. There are no guidelines in clinical practice regarding the treatment and diagnosis of DMS. With this study, we sought to present a perspective by reviewing the theoretical information.

INTRODUCTION

Delusional misidentification syndrome (DMS) is an umbrella term that covers four main variables, including Capgras, Fregoli, intermetamorphosis, and syndrome of subjective doubles (1). Courbon and Tusques (2) first used the term intermetamorphosis syndrome in 1932. Affected people believe that other people change both their physical and psychological identity. While physical identity defines appearance and physical characteristics, psychological identity is related to the concept of "self," that is, it interprets "who" the patient is. For example, in a case described in the literature, it was reported that a male patient was physically turning into his stepfather, not just psychologically. The patient stated that his face had turned into the face of his stepfather and that his extremities, penis and stomach were similarly the same as his stepfather's (3). Since intermetamorphosis syndrome is rarely seen, there are few studies about it. In intermetamorphosis, people believe that other people's identities, both physical and psychological, are transformed into someone else. Reverse intermetamorphosis is a rare subtype. In the case of reverse intermetamorphosis syndrome, the patient believes that he/she has gained a new identity after radical changes in both the physical and psychological identity (4).

This case report presents and discusses treatment of a patient with synesthesic hallucinations in addition to delusions of reverse intermetamorphosis, who developed side effects to various antipsychotic treatments but responded positively to electroconvulsive treatment.

Address for Correspondence: ✉ Dr. Fatih Yığman, Health Sciences University, Dışkapı Yıldırım Beyazıt Research and Training Hospital, Psychiatry Clinic, Şehit Ömer Halisdemir Street, No:1, Altındağ, Ankara, 06110 Turkey dr.yigman@gmail.com

PATIENT INFORMATION

We describe this 31-year-old, single, high school graduate, and unemployed female patient as S. S. lives with her mother in Ankara. According to her story, her first symptoms began when she was 18 after acceptance into university. At that time, her belief that she would suffer from her environment began to form. She believed that people had terrible plans for her. She was unable to attend university due to her symptoms; she was never employed and was hospitalized six times in a psychiatric clinic within 13 years. Exacerbation of illness was always provoked by psychomotor agitation, suspicion, anxiety, hallucinations, and arguments with her mother. She had been receiving I.M. risperidone depot injection 50mg every two weeks and oral tab 4mg daily, and olanzapine 20mg daily for the past year. However, over the last six months the patient had developed beliefs that she was a robot named B. that had changed places with the real S., and was hospitalized due to increasingly violent behavior at home. It was reported that she was unresponsive to quetiapine and sertindole treatment from her previous hospitalizations.

CLINICAL FINDINGS

A mental state examination showed that the patient was conscious, cooperative and had complete orientation. She was defensive and suspicious throughout the interview. She believed she was a robot named B. and those unknown individuals kidnapped the real S., and that Robot B. had replaced the real S. by the same individuals. According to her statements, S. was a beautiful and polite child; however, Robot B. was ugly and not even human. The treatments given to Robot B. were actually parts of S.'s brain. Since her mother could not understand this difference, S. told her that she was angry. She also stated that her hospitalization was a plan by some mysterious people. She said that she heard voices and that she was able to smell the scent of battle with men. She stated that she could smell the blows that were dealt when people fight. Her family history was unremarkable. There was no additional disease in her clinical history. She had no history of substance or alcohol use. She was a 6 pack/year smoker.

DIAGNOSTIC ASSESSMENT

Neurological examination and EEG tests were given to exclude other underlying causes yielded normal results. MRIs were attempted three times; however, the patient was unable to remain stationary, and the MRIs were unsuccessful. There was no pathology in her brain imaging from previous hospitalizations. Her laboratory tests taken at admittance (thyroid function, vitamin B12, folate, ferritin, complete blood count, biochemistry parameters) were in the normal range.

THERAPEUTIC INTERVENTION

The patient's clinical state was evaluated as reverse intermetamorphosis and schizophrenia with synesthesia hallucinations. The BPRS score of the initial evaluation was 47. Since the patient had been unresponsive to antipsychotics such as quetiapine, sertindole, olanzapine and risperidone from previous hospitalizations and outpatient follow-ups, she was considered treatment-resistant and initiation of clozapine was planned. When clozapine reached 300 mg/day dose, the patient developed agranulocytosis, and the treatment was discontinued. When agranulocytosis subsided, amisulpride was initiated. Tachycardia developed at 400 mg/day dosage, and the treatment was discontinued. Due to the patient's continuous psychomotor agitation towards her relatives and medical staff, and the development of side effects with antipsychotics, ECT was planned. Complications developed under general anesthesia during the fourth session of ECT, and ECT was discontinued.

FOLLOW-UP AND OUTCOMES

The Brief Psychiatric Rating Scale score decreased to 27 after the fourth session of ECT. Her psychomotor agitation subsided and she ceased speaking about synesthetic hallucinations. It was decided to continue her treatment with monthly depot haloperidol decanoate injections.

DISCUSSION

Our case presented with paranoid delusions towards relatives, synesthetic hallucinations, and psychomotor agitation during exacerbation periods, and had been followed up after a diagnosis of schizophrenia for 13 years. For the last six months, the patient believed she was both physically and psychologically Robot B., and that the real S. had been kidnapped and replaced by unknown people. Her case was evaluated as reverse intermetamorphosis in addition to her existing schizophrenia diagnosis. According to the literature, 75% of DMS cases are female; the age of onset ranged from 12 to 78 years, with an average age of 40 years. In more than 4/5 of the patients, the age of onset was 30 years (5). Our 31-year-old female patient was consistent with the literature in terms of sociodemographic data.

Delusional misidentification syndromes are commonly associated with psychiatric disorders, including schizophrenia, bipolar disorder, and schizoaffective

disorder. However, it has also been known to occur in organic neurological manifestations such as dementia, brain tumor and Parkinson's disease (4). In a study by Kirov et al. (6) conducted with 195 patients diagnosed with psychosis they found eight patients who fit the "classic" delusional misidentification syndrome. They reported that six patients were diagnosed with Capgras, one patient with Capgras and syndrome of subjective doubles, and one patient with a syndrome of subjective doubles. According to this study, it was reported that 4.1% of patients diagnosed with psychosis had delusional misidentification syndromes (6). Our case is the only report in the literature to combine reverse intermetamorphosis syndrome and synesthetic hallucinations, to the best of our knowledge. Therefore, we believe our case report is important as it will contribute to the literature by raising awareness of the syndrome and treatment methods.

Studies on etiopathogenesis have focused on the right frontal lobe and right hemisphere, and have emphasized the frontal and temporal lobes (7). In our case, brain imaging was not performed in the patient's last hospitalization, although normal results of the EEG and neurological examination gave us reservations about an underlying neurological etiology.

Field reports should examine these cases in more detail to better understand DMS and to create a clearer diagnosis and treatment algorithm for future studies. Thus, it may be possible to recognize the diagnoses of the DMS group and reach the correct treatment approaches. This case allowed us to clarify the definition and classification of intermetamorphosis syndrome and its relation to the patient's perception of identity. In reverse intermetamorphosis syndrome, these two identities are perceived differently both psychologically and physically, and the patient's explanation of the relationship between the two identities included a different concept of delusion, involving a radical physical and psychological change within the self. The recommended model for an accurate definition of delusional misidentification should comprise three main stages: First, the real patient S. should be carefully identified. According to the patient's belief, S. was a beautiful girl, a polite child, and loved her mother. S. had been kidnapped by some evil people. Secondly, we must characterize the changed identity, Robot B. According to the patient's beliefs, Robot B. was an ugly woman who was not even human. The cigarettes she smoked were produced from S.'s brain. These two steps allow systematic evaluation and comparison between the two identities. Finally, the psychological relationships between the two identities should be evaluated. The patient thinks she has someone else's identity and also thinks that she has the characteristics of the identity she is physically thinking about, such as hair, face, stomach or genitals. According to the patient, Robot B. was created by the same evil people who had kidnapped S. S's mother did not realize S. had been kidnapped and therefore Robot B. was angry with her mother.

There are no guidelines on the treatment and diagnosis of delusional misidentification syndromes in clinical practice. They are generally associated with exacerbated periods of comorbid psychiatric disorders and regress along with the regression of underlying illness. According to many researchers, DMS symptoms are very resistant to treatment despite various neuroleptic treatments. The literature reports a limited number of misidentification syndrome cases treated with antipsychotics such as olanzapine, risperidone, quetiapine, trifluoperazine and pimozide; however, syndromes generally have an inadequate response to neuroleptic treatments and are treatment-resistant (8). Case reports and information on treatment options of reverse intermetamorphosis are limited. In our case S. was unresponsive to treatments for schizophrenia, including quetiapine and sertindole. The patient's delusions continued despite a one-year combined treatment of risperidone and olanzapine. Clozapine and amisulpride treatments were discontinued due to side effects. Our patient underwent ECT and had a significant response after four sessions. There are few studies in the literature on the effectiveness of ECT treatment for delusional misidentification syndromes (9). The fact that our patient's delusions regressed with ECT shows that ECT may be preferred as an option in the treatment of delusional misidentification syndromes.

Recognizing the delusional relationship between the patient's actual identity and the changed identity and developing a more precise and objective definition of the illness is necessary for understanding delusional misidentification syndromes and evaluating the treatment process. A clear definitive system for delusional misidentification syndromes will help clinicians in accurately evaluating these syndromes. There is, moreover, a need for further studies on the phenomenology and etiology, including organic pathologies, and its relationships with other psychiatric disorders. This case report may help contribute to other studies in this field.

PATIENT PERSPECTIVE

Following ECT, S. now knew she was herself and stopped thinking about Robot B. She did not want to talk about her past delusions, but her relationship with her mother became more positive.

Informed Consent

Patient confidentiality has been strictly observed. Written informed consent for the publication of these cases was obtained from the patient or their legal guardian.

Disclosure

The authors report no conflict of interest, and they alone are responsible for the content and writing of this paper.

References

1. Christodoulou G, Malliara-Loulakaki S. Delusional misidentification syndromes and cerebral "dysrhythmia." Psychopathology 1981;14:245-251.
2. Courbon P, Tusques J. Identification délirante et fausse reconnaissance. Ann Med Psychol 1932;190:1-12 (French).
3. Silva J, Leong G. Syndrome of "reverse" intermetamorphosis. Can J Psychiatry 1996;41:260.
4. Barrelle A, Luauté J-P. Capgras syndrome and other delusional misidentification syndromes. Neurologic-Psychiatric Syndromes in Focus-Part II. 42. Freiburg: Karger Publishers, 2018: pp. 35-43.
5. Kandeğer A, Tekdemir R, Selvi Y. Delusional misidentification syndromes: A case of intermetamorphosis. J Clin Psychiatry 2017; 20:150-154.
6. Kirov G, Jones P, Lewis S. Prevalence of delusional misidentification syndromes. Psychopathology 1994;27:148-149.
7. Darby R, Prasad S. Lesion-related delusional misidentification syndromes: A comprehensive review of reported cases. J Neuropsychiatry Clin Neurosci 2016;28:217-222.
8. Yalin Ş, Varol Taş F, Gunevır T. The coexistence of Capgras, Fregoli and Cotard's Syndromes in an adolescent case. Noro Psikiyatr Ars 2008;45.
9. Rapinesi C, Kotzalidis GD, Casale AD, et al. Treatment-resistant, five-year long, postpartum-onset Capgras episode resolving after electroconvulsive therapy. Int J Psychiatry Med 2015;49:227-234.