Psychiatria Danubina, 2024; Vol. 36, No. 2, pp 259-261 https://doi.org/10.24869/psyd.2024.259 *Case Report*

EROTOMANIC DELUSIONS AND TACTILE HALLUCINATIONS IN A LATE ADOLESCENT GIRL WITH SCHIZENCEPHALY

Zeynep Nur Demirok

Department of Psychiatry, Kartal Dr. Lutfi Kirdar City Hospital, Istanbul, Turkiye

received: 17. 12. 2023; revised: 2. 3. 2024; accepted: 2. 4. 2024

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INTRODUCTION

Schizencephaly, a rare congenital disorder which is examined in cortical developmental malformations, is characterized by unilateral or bilateral clefts lined with dysplastic gray matter, extending from the pia surface to the ventricles (Patwal et al. 2022). It is known that structural and developmental disorders of the brain pose a risk for the development of neuropsychiatric symptoms (Borgwardt et al. 2007). To this day, a total of 9 cases have been reported in which psychosis is concurrent with schizencephaly (Patwal et al. 2022). In reported cases thus far, symptoms such as auditory and visual hallucinations, religious, grandiose and persecutory delusions, thought insertion and withdrawal have been documented. However, to our knowledge there have been no reports of such cases occurring with erotomanic delusions and tactile hallucinations. Here, we report a case of tactile hallucinations and erotomanic delusions in a patient who has been followed up with a diagnosis of schizencephaly since the age of 2.5 and developed psychotic symptoms in late adolescence. By sharing this case presentation, we aimed to contribute to our knowledge about the psychotic manifestations of schizencephaly.

CASE PRESENTATION

A 19-year-old girl was admitted to the outpatient clinic along with her mother. In the 3 month-period before their visit, she had difficulty in sleeping, irritability, crying, and thoughts of having been infested with lice. Furthermore, she exhibited ambivalence and frequently and repeatedly asked questions. At the same time, she began to hear the voice of a boy from her school. His voice was telling her that he was in love with her most of the time, but also saying insulting words to her from time to time. She frequently felt his kisses on her lips and neck and his hands on her breasts and other parts of her body. She often told her family that he was in love with her and that she needed to reach him. For that reason, she was sending him a lot of messages on social media, requesting to communicate with him. Her mother stated that sometimes she answered to the voices that she heard and acted as though she was trying to wipe something off her neck and body. Her mother explained that the boy was a student with mild intellectual disability, and the patient had last seen him a year ago. There was no history of contact between the patient and him that could traumatize the patient psychologically or physically. According to her medical history, she had epileptic seizures since infancy. At the age of 2.5, her first brain MRI revealed cortical dysplasia on bilateral frontotemporal areas and a cleft at the right anterior frontal lobe extending from cortex to ventricle, suggesting open lip schizencephaly (Figure 1). After the diagnosis, she initiated pediatric neurologic follow-up. Her last epileptic seizure was about 1 year before

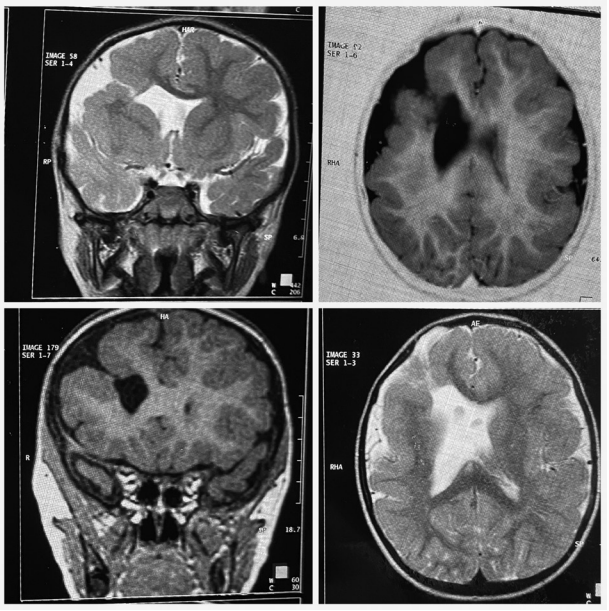

Figure 1. MRI scans showing a cleft opening to ventricle at right anterior frontal lobe

her psychiatric complaints emerged. She was treated with lamotrigine 150mg daily for a couple of years. In addition, previous medical records, at the age of 4-5, reported that she had velopharyngeal deficiency and left spastic hemiplegia. For this, speech therapy and physiotherapy were initiated. To the history ascertained by her mother, we concluded that the patient had psychomotor developmental delay. She received special education for 15 years. She was able to acquire literacy skills. The patient had no prior history of psychiatric treatment or medication use before this current application. She had no history of alcohol and substance use. The family history was free of psychiatric conditions.

Neurological examination revealed a muscle strength of 4/5 in the left upper and lower extremities and dysarthria, consistent with her previous medical records. During the mental status examination, the patient was cooperative and oriented in time and place. In the first interview, she was willing to explain her complaints, especially her discomfort with the conflicting messages the boy gave her. Her speech was within normal limits in all aspects, except for some articulation problems. Her mood was dysphoric, and her affect was congruent with her mood. Perseveration was detected in her thought process. The content of her thoughts revealed delusional beliefs of erotomania. As a perceptual disorder, auditory and tactile hallucinations were detected. She lacked insight about her psychotic symptoms.

She scored 59 on the Kent EGY Test and 63 on the Porteus maze test. Complete hemogram, blood sugar, electrolytes, liver function tests, kidney function tests, thyroid function test, lipid profile tests were within normal limits. The patient was consulted with a neurologist. Her complaints were not associated with seizures, and it was recommended to continue the current antiepileptic treatment.

We established a probable causal relationship between the patient's premorbid neurodevelopmental condition, schizencephaly, and her clinical presentation. We concluded the admission with a diagnosis of "mild intellectual developmental disorder" and "psychotic disorder due to another medical condition". She was initiated on aripiprazole 5mg and fluoxetine 20mg daily. Because of its gastrointestinal side effects, aripiprazole was switched to olanzapine, and was gradually increased to 20 mg daily, a therapeutic dosage for psychosis. Fluoxetine was increased to 40mg daily. During the follow-up, the patient fully recovered from her affective symptoms, ambivalence, sleep problems and thoughts of having lice. She experienced partial remission of erotic delusional beliefs. Although she continued to believe that the boy whose voice she heard had feelings for her, they were less intense and more open to being challenged. Her stalking behavior persisted, albeit with a lower frequency. A decrease in intensity of tactile and auditory hallucinations was recorded. Besides, she no longer presented further acting on the hallucinations. Written informed consent was obtained from the legal guardian of the patient for the publication of this case presentation

DISCUSSION

In this report, a patient who was diagnosed with epilepsy comorbid with schizencephaly since the age of 2.5 years and developed psychotic disorder in late adolescence is discussed. In the literature review, a total of 9 cases in which the presence of psychosis with schizencephaly was reported, 2 of which were associated with bipolar disease, and the others were cases in which the first psychotic episode or chronic psychosis was detected.

It has been understood that in the majority of cases reported so far, hallucinations are predominantly auditory and visual in nature. In cases where delusions are present, it is observed that paranoid, grandiose, and jealous delusions are found (Tylš et al. 2019). The case mentioned here differs from other cases in the literature with the presence of erotomanic delusions and tactile hallucinations. Negative symptoms such as flattening of affect and decreased speech, which are reported in some of the cases in the literature, are not present in this case (Tylš et al. 2019, Tibrewal et al. 2015). Of the 9 cases available, only one is similar to the case here, which is open lip schizencephaly (Melo et al. 2013).

In the mentioned case, neurological anomalies such as low cognitive capacity, comorbid epilepsy, motor weakness, and speech abnormalities were present as in the other cases in the literature. In the cases reported so far, patients were diagnosed with schizencephaly after neuroimaging tests were performed accidentally or upon clinically suspecting organicity after the development of psychotic symptoms. In this case, however, a patient followed up with schizencephaly showed psychotic symptoms after a long delay- during a risky period of life for the development of psychosis. In addition to the fact that the onset of the patient's symptoms occurred at a typical age range for psychosis, the onset of psychotic symptoms during a period when the patient had not had a seizure for over a year and the electroencephalography recordings were normalized, could suggest the phenomenon of "forced normalization". This phenomenon has been described as the electroencephalography findings after the emergence of psychotic symptoms in an epileptic patient being more normal or completely normal compared to previous findings,

or the onset of psychotic symptoms in a patient after controlling epileptic seizures with medication (Kawakami & Itoh 2017). During the 1.5-year follow-up period, the fact that the patient did not have another epileptic seizure and the partial remission of psychotic symptoms despite the antipsychotic therapy applied at a therapeutic dose may support this phenomenon (Carazo Barrios et al. 2020).

The relationship between abnormal brain structure and psychosis has long been debated. The neurodevelopmental hypothesis of schizophrenia suggests that the foundations of a condition that emerges as a frank disorder in adolescence or early adulthood may be events that occur in early development (Owen et al. 2011). Schizencephaly, which can be defined as a neuronal migration anomaly during the third to fifth gestational months, is a neurodevelopmental disorder that has been argued to be related with psychosis (Melo et al. 2013). However, our knowledge about the association between schizencephaly and psychosis is limited due to the low prevalence of schizencephaly. It has been suggested that disruption in intracortical connections may be a possible mechanism in the tendency of psychosis development in later stages of life in this malformation (Tylš et al. 2019). The hypothesis has been proposed that the disruption of the focal activation mechanism in cortical activity is associated with the schizophrenic cortex (Weinberger 1996).

Our patient also had intellectual disability. In a previous case in which erotomanic delusions and somatic sexual hallucinations were observed in a woman with intellectual disability, it was argued that these types of psychotic symptoms may develop through a compensatory mechanism due to the patient's lack of social skills and barriers to building a wide social network (McGuire et al. 1994).The manifestation of similar symptoms in our case can be related to the patient's similar difficulties.

We believe that the case mentioned in this article is an important contribution to our limited knowledge about psychotic manifestations in schizencephaly, a rare neurodevelopmental anomaly. Systematic studies are needed to better understand the relationship between schizencephaly and the development of psychosis.

Ethical Considerations: Does this study include human subjects? NO

Conflict of interest: No conflict of interest

Funding sources: The author received no funding from an external source.

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Correspondence:
Zeynep Nur Demirok
Kartal Dr. Lutfi Kirdar City Hospital Cevizli,
D-100 Guney Yanyol, Cevizli Mevkii No:47,
34865 Kartal/İstanbul, Turkiye
zeynepnurdemirok@gmail.com / ++905418512775