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Case Report

Complete agenesis of corpus callosum with a rare neuropsychiatric presentation: A case report ☆,☆☆

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ABSTRACT

Corpus callosum agenesis is a rare phenomenon that might be associated with neuropsychiatric disorders. We present a 28-year-old woman with complete corpus callosum agenesis who presented with mood disturbance, psychosis, and delusional symptoms with schizophrenia diagnosis. She had a good response to antipsychotic therapy with risperidone.

Introduction

There has been evidence on the correlation between corpus callosum agenesis (ACC) and psychiatric disorders. The most common disorders that have been suggested are schizophrenia and neurological disturbance such as epilepsy, unipolar depression, bipolar depression, and intellectual impairment [1,2]. Andermann's syndrome is also explained as ACC, mental retardation, peripheral neuronopathy, and psychosis [3]. In the cases that are accompanied by schizophrenia, ACC is usually partial [4].

Corpus callosum (CC) is a white matter structure which connects 2 hemispheres and is involved in many higher mental functions such as memory, speech, emotion, and behavior [5]. It is in association with the formation of hippocampus, cin-

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gulate cortex, and fornix which are involved in the pathophysiology of schizophrenia [5]. The incidence of ACC is estimated to range from 0.004% to 0.7% with male predominance in different study populations; however, there is a limited data [6,7].

Considering the low incidence of ACC and also rarity of corpus callosum agenesis and schizophrenia, the coincidence of these 2 must always be noted. As of yet, there are no well-defined syndromic classification of corpus callosum pathology. In this report, we describe a case of ACC that presented with mood disturbance and schizophrenia symptoms.

Case presentation

We present a 28-year-old single, Iranian woman who had a bachelor's degree in accounting and presented with psychological symptoms. The patient had no abnormal history of gestational and birth problem or developmental delay. The patient's development was normal in school age and no remarkable abnormality was declared during her childhood. Her family history and drug use history were negative. Also, the patient did not declare any history of alcohol consumption. The patient's premorbid characteristics included anxiety, dependency, and isolation. Her parents stated she had a questionable history of seizure (generalized tonic colonic) during adolescence that was treated with Carbamazepine which discontinued from several years ago.

The early symptoms were mostly mood dominant without any previous abnormal psychological history. On her hospital admission, the patient developed with aggression, isolation, impaired communication, hypo-activity, and significant loss of function; these symptoms had started 6 months prior to admission which was considered as mood disorder impression. She suffered abolition which was a negative symptom.

Mental status examination showed anxiety, through disorders with persecutory themes and suspicious thinking such as prominent systematic persecutory delusion toward her family. She also claimed previous paranoid delusion about injury from her father. No sign of disorganized or disturbed language was found. Physical and neurological examination was unremarkable. The tests for co-ordination (finger nose test, heel-shin test, and dysdiadochokinesia) were normal. Sensory and motor systems and cranial nerves were intact. The result of her I.Q. test was 59.

Para-clinic studies did not show any significant abnormal finding. Lab data including cell blood count and kidney liver function tests were within normal limits. Serum Na and K were 145 and 4.2mmol/L, respectively. TSH level was 1.8I U/mL, and beta human chorionic gonadotropin was negative. Urine analyses and coagulation profiles as well as routine chest radiograph and electrocardiogram were also within normal limits. Urine toxicology was negative for controlled substances, illicit drugs, and alcohol.

The patient had a good response to escitalopram that started with 5 mg/daily and optimized to 10 mg/daily and risperidone that was administered 6 mg/daily. After 4 days of admission, some of the symptoms such as anxiety and insomnia were resolved, but she still had paranoid thought toward family. She was discharged from hospital after recovery of mood symptoms, while delusional symptoms were persistent with primary impression as provisional schizophrenia. In the follow-up, the patient's psychosis and delusions were persistent and gradually became dominant. She was observed closely due to the severity of symptoms in particular for persecutory delusions. Psychosis symptoms diminished gradually during the follow-ups in 2 months. Schizophrenia was confirmed as the patient's diagnosis using structural clinical interview for the Diagnosis and Statistical Manual of Mental Disorder (5th edition). The patient underwent a brain MRI considering that she developed with first episode of psychosis and also she had a history of convulsion in childhood.

Brain MRI was done for the patient which showed different signs of ACC such as dilatation of the occipital horns of the lateral ventricles (colpocephally) (Fig. 1A), concave appearance of the lateral and third ventricles named as moose head sign (Fig. 1B), special orientation of the frontal and occipital horns of the lateral ventricles named racing car sign (Fig. 1C), radiating midline sulci due to the absence of corpus callosum (Fig. 1D), and tear drop lateral ventricles and parallel parasagittal white matter (Probst bundles) (Fig. 1E).

Discussion

The relationship between brain anomalies and psychiatric manifestations has been proposed for a long time, while the exact correlation and the pathophysiologic mechanism are not fully known in many of them, in particular the correlation between ACC and schizophrenia. Although many cases of ACC might be asymptomatic, in cases who present with clinical symptoms CC is usually atrophic or absent [1,8–10]. There are growing data which show that anatomical brain abnormalities can accompany psychiatric manifestations such as delusion, psychosis, and hallucinations [2,11,12].

The clinical manifestations of ACC vary from simple learning disabilities and cognitive deficits to mood disorders and psychosis [13,14]. Previous review articles suggested that the incidence rate was seen more in males and the mean age was 33 [1]. The incidence of schizophrenia was observed either in early age or in late adulthood. Studies showed that early schizophrenia was related to thickening or fat deposition of CC, while late schizophrenia was more related to atrophy of CC; however, the findings are controversial. Also, it seems that the complete anomaly of CC correlates with positive symptoms such as delusions and hallucinations in some reports [4,6,15].

There is the possibility that abnormalities of CC correlate with the pathogenesis of schizophrenia considering that this structure is responsible for interhemispheric data transfer; as a result, disconnection may arise between two hemisphere psychiatric symptoms. Furthermore, CC has connections with other structures such as the hippocampus and cingulate cortex which are also involved in schizophrenia patients [12]. Some previous reports declared that schizophrenia and delusions usually happened in patients with partial agenesis [1,11,16], while some other reported complete agenesis of CC such as our case [1,17].

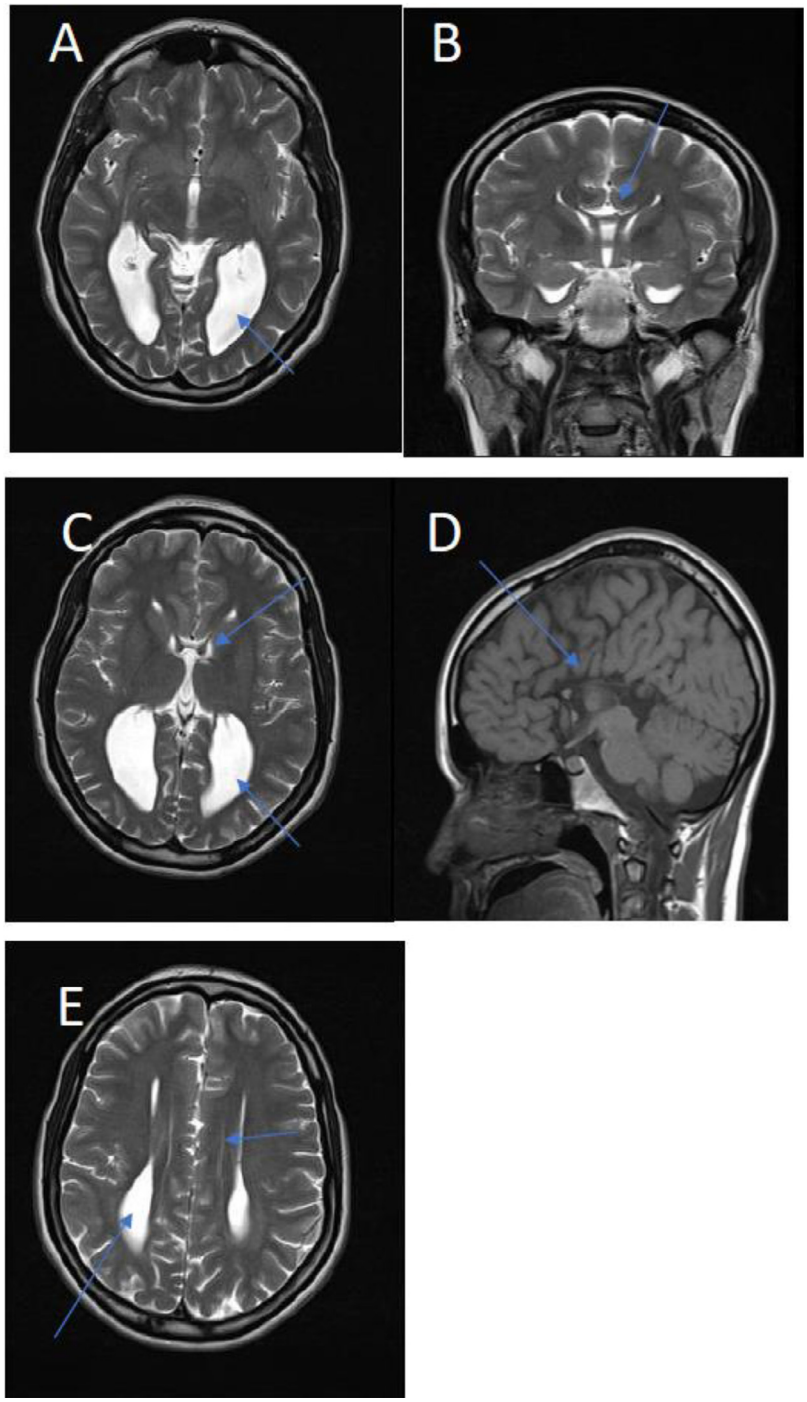

Fig. 1 – (A) Axial T2 brain MRI sequence with dilatation of the occipital horns of the lateral ventricles (colpocephally). (B) Coronal T2 sequence brain MRI with moose head sign in the lateral and third ventricles. (C) Axial T2 brain MRI sequence with racing car sign of the lateral ventricles. (D) Sagittal T1 brain MRI sequence with complete corpus callosum agenesis (CCA) and radiating midline sulci. (E) Axial T2 sequence brain MRI with tear drop lateral ventricles (long arrow) and Probst bundles (short arrow).

There is some evidence that explains the relationship between the severity of brain abnormalities and response to antipsychotic drugs [4–6]. The reason of poor response to antipsychotic agents is controversial and not fully understood, but it might be due to microscopic structural abnormalities and disconnection between the hemispheres.

Conclusion

Despite many cases of schizophrenia that may have no brain abnormalities, here we reported a case with schizophrenia impression that had complete agenesis of corpus

callosum. This case implies the importance of radiologic work-ups in patients with psychiatric problems, particularly when they present with poor response to antipsychotics or bizarre disease course. In such cases, more focus on psychopharmacological approach may be more beneficial.

Authors' contribution

The conception and design of the study was done by Leila Razeghian Jahromi and Mahsa Ghavipisheh. Material preparation and data collection were performed by Mehdi Ghaedian Jahromi and Mahsa Ghavipisheh. The first draft of the manuscript was written by Iman Ahrari and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

Ethic approval

The study was approved by ethic committee of Shiraz University of Medical Sciences with IR.SUMS.REC.1401.545 ID number.

Patient consent

Written informed consent was obtained from the patient for all the data that would be published in the manuscript.

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