A Case of Corpus Callosum Agenesis Presenting with Recurrent Brief Depression

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ABSTRACT

Agenesis of corpus callosum can have various neuropsychiatric manifestations. Following case report highlights the case of a young man presenting with features of recurrent brief depressive disorder, each lasting for about 3 to 7 days, for over a year. He had history of occasional headache and episodes of swooning attack in between, usually precipitated by emotional events. His neuroimaging revealed agenesis of corpus callosum. He was experiencing swooning attacks as he became aware that some 'unusual' findings were present in his reports. Recurrent brief depression can be a manifestation of this congenital anomaly, and conversion disorder can be present as comorbid diagnosis perhaps due to ignorance and fear of this apparently innocuous congenital malformation.

Key words: *Conversion disorder, corpus callosum agenesis, recurrent depression*

INTRODUCTION

Agenesis of corpus callosum (ACC) is mostly a congenital anomaly that occurs predominantly in males, either isolated or in combination with other CNS or systemic malformations. Corpus callosum develops from the lamina reuniens in the telencephalon and begins to appear at around 10.5 weeks, till it reaches adult form by 17 weeks. The occurrence of ACC is usually sporadic. Disturbance of embryogenesis in the first trimester of pregnancy by an unknown insult is sometimes put forward as a probable antenatal cause. Once thought to be a rare condition, ACC has been turning up with increasing frequency ever since the widespread clinical use of MRI. ACC can occur in other congenital anomalies, including Arnold-Chiari malformation, Dandy-Walker syndrome, schizencephaly (clefts in brain tissue) and holoprosencephaly (failure of the forebrain to divide into lobes), Aicardi's syndrome, fetal alcohol syndrome and several trisomies.[1] ACC can also be associated with malformations in other parts of the body, such as midline facial defects, or can occur as one of the developmental abnormalities in schizophrenia. The effects of the disorder range from subtle to severe, depending on associated brain abnormalities. Intelligence may be normal with mild compromise of skills requiring matching of visual patterns. But children with the most severe brain malformations may have intellectual disability, seizures, hydrocephalus and spasticity.[2,3]

Previous literatures have mentioned about various neuropsychiatric presentations of this congenital anomaly. Depressive spectrum disorders have been mentioned in various literatures to be present with this anomaly, but recurrent brief depression has not been mentioned in any of them.[3,4] This congenital anomaly has also been found in bipolar disorder,[5] schizophrenia[6] and can interfere with psychomotor development.[7] In addition, individuals with ACC are reported to have more sensory deficits and abnormalities, as well as altered patterns of feeding/eating, elimination and sleep.[8]

CASE REPORT

A 25-year-old man residing in a suburban region was brought to the Psychiatry OPD at the tertiary care

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hospital with biparietal dull-aching headache lasting for about an hour, occurring 3 to 4 times per month for the past 2 years. The headache, to start with, was spontaneous in onset, without nausea or vomiting, diurnal variation, fever, photophobia, phonophobia, and subsided with analgesics (tablet ibuprofen 400 mg) only. He had been prescribed tab Flunarizine 10 mg OD and Amitriptyline 25 mg OD for the last 1 year. On careful history-taking from the informant (father) and the patient himself, it was known that the patient had suffered from episodes of depressed mood and irritability every month lasting for 3 to 7 days on an average, diminished interest in pleasurable activities, hypersomnia, psychomotor retardation, fatigue and loss of energy for the past 1 year with recurrent thoughts of death but without any history of suicidal attempt so far. There was no positive family history of any major medical or psychiatric illnesses in the family up to three generations. He had been advised to have a CT scan followed by an MRI of brain for the persistent headache, 1 month back. Also there was a history of palpitation, breathlessness, clenching of teeth followed by unresponsiveness, each episode lasting for 30 minutes to 1 hour, four times in the past 2 weeks. There had been no tongue bite, incontinence, loss of consciousness during the episodes, and the patient was occasionally verbalizing during these periods. He had been brought to the emergency room 1 week back by his family members and neighbors in a state of unresponsiveness. The reported symptoms had lasted for 2 hours, along with jerky movements of all four limbs and clenching of teeth but without any injury, incontinence or fall. According to his parents, his problems started suddenly after he came to know his MRI brain findings. He was able to adjust his body parts when placed in awkward position during these periods of unresponsiveness. After gaining consciousness, he refused to talk or answer but tried to use gestures and sign languages to answer questions asked to him. His vocalization was normal as tested by cough reflex. He had a fall when he was asked to walk a few steps, but the fall was guarded without any injury.

During mental status examination (MSE), the patient was found to be guarded initially, with increased reaction time, monotonous speech and depressed affect. Neuropsychological testing revealed subtle differences in higher cortical function. He suddenly had an attack mimicking a swooning attack in the presence of his family members while the interview was in progress, which subsided on negative reinforcements and suggestions. There were no clinical features suggestive of an epileptic seizure, which was confirmed by routine EEG and subsequently by video-EEG.

DISCUSSION

Agenesis of corpus callosum is a rare condition (with US frequency rates reported to be 0.07% to 5.3%) which usually presents with clinical features such as seizures, feeding problems, developmental delay, impaired hand-eye coordination, impaired visual and auditory memory and hydrocephalus. In this patient, there were few delayed milestones of development only without any gross physical or mental disability. (IQ was 77 as per WAIS-IV version.) No other congenital anomalies were found. The uniqueness of the case was that it presented with recurrent depressive disorder with swooning attacks. The patient responded well to a trial of benzodiazepines (Clonazepam) followed by maintenance with SSRIs. The diagnosis has been done on the basis of appendix B of the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR), which includes criteria and axes provided for further study. The diagnosis remained stable on

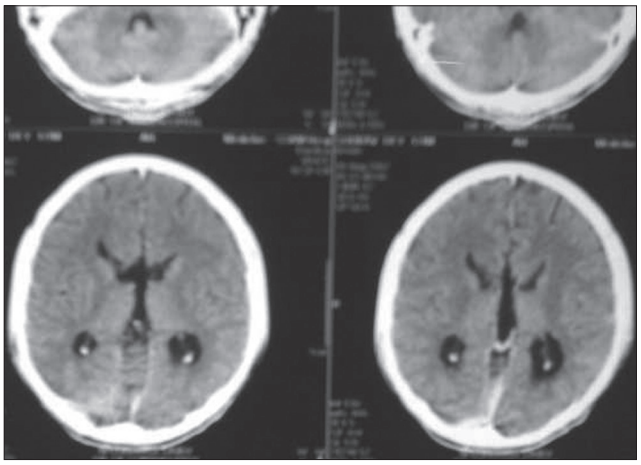

Figure 1: Contrast-enhanced CT scan showing parallel ventricles with continuation of interhemispheric fissure with the third ventricle; however, there was no evidence of any interhemispheric cyst

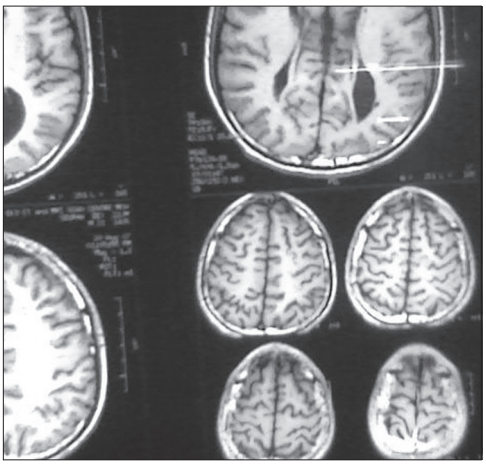

Figure 2: Axial T1 MRI showing prominent interdigitations of gyri

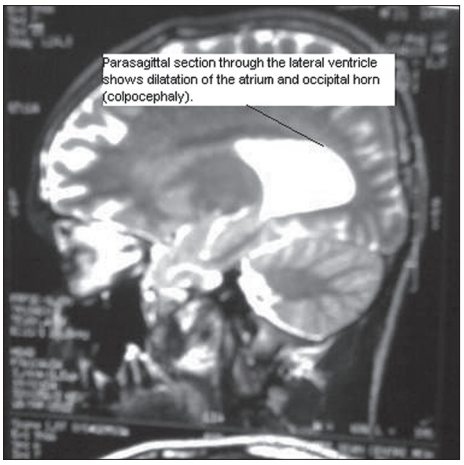

Figure 3: The sagittal T1 image in this patient showing absent anterior genu and splenium. Third ventricle was high riding interposed between the two lateral ventricles. There were also dilatations of the atrium and occipital horn (colpocephaly)

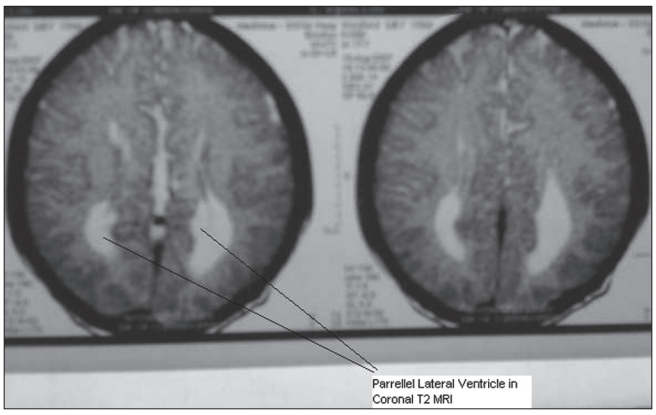

Figure 4: Coronal T2 MRI of the patient showing agenesis of corpus callosum with parallel lateral ventricles

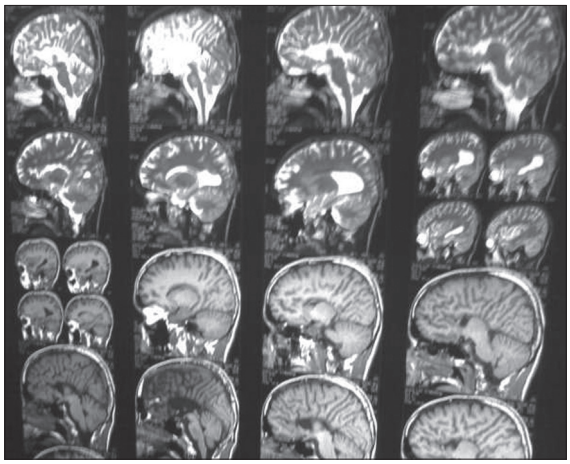

Figure 5: The sagittal sections of the T2-weighed imaging replicating earlier findings

subsequent follow-up visits and did not turn out to be any other major mood disorders. Though the condition can have wide range of manifestations, in our case we noticed only recurrent brief depression *de novo* and conversion disorder as a consequence of reaction when the diagnosis was disclosed to the patient. He never met the criteria of major depressive disorder during the entire period of his illness.

CONCLUSIONS

The clinical manifestations of callosal agenesis can be described under two headings, viz., nonsyndromic and syndromic. Nonsyndromic forms are the most common. An unknown, though probably small, proportion of patients are completely asymptomatic, or more commonly, their condition is incidentally discovered during neuroimaging.[9] The recurrent brief depression is an entity mentioned in ICD-10 (International Classification of Mental and Behavioral Disorders) in the section of other recurrent mood (affective) disorders (F 38.10). The DSM-IV-TR demands further study to formulate a more valid diagnosis of this entity. The questions of stability and validity of its diagnosis are always there since its nosology. In this case, it has been diagnosed with another rare congenital CNS anomaly, the agenesis of corpus callosum (ACC). Psychiatric antecedents and comorbidities in agenesis of corpus callosum (ACC) need to be explored more in future case reports. Similarly comorbid general medical conditions need to be studied for getting more information on the diagnosis of recurrent brief depressive disorder.[10]

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