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Case Report

Behavioral disinhibition following corpus callosotomy done for colloid cyst excision in 15-year-old girl: A case report and literature review

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ABSTRACT

Background: There is a growing body of literature suggesting that the corpus callosum plays an important role in behavior. While behavioral deficits are a rare complication following callosotomy, they are well-documented in agenesis of the corpus callosum (AgCC), with emerging evidence reporting disinhibition among children with AgCC.

Case Description: A 15-year-old girl had undergone a right frontal craniotomy and excision of a third ventricle colloid cyst using the transcallosal approach. Ten days after the operation, she was readmitted for progressive symptoms of behavioral disinhibition. Postoperative magnetic resonance imaging of the brain showed mild-to-moderate bilateral edematous changes along the operative bed, with no other significant findings.

Conclusion: To the best of the authors' knowledge, this is the first report in literature to describe behavioral disinhibition occurring as a sequelae to a surgical procedure involving callosotomy.

Keywords: Behavioral disinhibition, Corpus callosotomy, Corpus callosum

INTRODUCTION

The corpus callosum consists of white matter tracts that connect the right and left cerebral hemispheres. It is composed of five parts from anterior to posterior: the rostrum, genu, body, isthmus, and splenium. Fibers of the rostrum and genu arch anteriorly to form the forceps minor, interconnecting the anterior parts of the two frontal lobes. The body fibers interconnect the corona radiata of each side and other white matter pathways of the posterior part of the frontal lobe, the entire parietal lobe and the superior part of the temporal lobe. Finally, fibers of the splenium arch posteriorly and form the forceps major, interconnecting the occipital lobes of each side.[6,21]

Significant advances have been made in understanding the function of the corpus callosum by studying callosotomy patients. Observations from patients who undergo callosotomy for the treatment of epilepsy revealed topographical organization of the corpus callosum, with its anterior regions involved in the transfer of motor and high-level cognitive signals and its posterior regions

involved in the transfer of somatosensory (posterior midbody), auditory (isthmus), and visual (splenium) information.[6]

Compared to adults, children seem to experience fewer neurologic complications following callosotomy. This could be attributed to enhanced neural plasticity in children that may allow development of alternate neural pathways.[11,14] While most adverse effects of callosotomy are transient, they can be permanent in some cases, and include memory deficits, neuropsychological impairment, language impairment, and seizures.[1] Disconnection syndrome, a major concern after corpus callosotomy, varies in severity and presentation depending on the site and extent of hemispheric disconnection. Acute disconnection syndrome, which improves over a period of weeks to months, can present as reduced spontaneous speech, dysphagia, non-dominant leg paresis, and incontinence. Patients with chronic disconnection syndrome can present with features of alien hand syndrome, non-dominant hand agraphia, hemispatial neglect, reduced dichotic listening, and tactile dysnomia.[20] Although neuropsychological impairment secondary to callosotomy is usually mild and difficult to detect on clinical examination, there have been reports describing development of depression, excessive self-insistence, and schizophrenic symptoms after callosotomy.[1,9]

Individuals with agenesis of the corpus callosum (AgCC) also present a unique opportunity to learn about the role of the corpus callosum in cognitive processing. AgCC has been shown to be associated with impairment in multiple cognitive domains, most particularly in problem solving skills and processing speed. Secondary to these core deficits, individuals with AgCC may present with impairment in abstract reasoning and second-order linguistic deficits, as well as social and behavioral abnormalities.[7,16]

To the best of the authors' knowledge, there is no published report of behavioral disinhibition following callosotomy and hence, we thought that it was worthwhile to describe this patient.

CASE DESCRIPTION

This patient was a 15-year-old girl who presented with a short history of progressive headache, dizziness, diplopia and visual blurring. Ophthalmology review confirmed papilloedema. Pre-operative brain MRI image demonstrated a hyperintense lesion in the third ventricle, consistent with a colloid cyst [Figure 1]. She underwent a right frontal craniotomy for excision of the cyst through an interhemispheric transcollosal approach. The post-operative period was uneventful, and she was discharged home on the fifth day. Ten days after the operation, she was readmitted for progressive disturbance in behavior that started 3 days after the operation, including inappropriate sexual actions; tearing clothes and throwing napkins at hospital staff; using inappropriate language, talking loudly and singing in public settings; voluntarily emptying her bladder in public; as well as making a mess while eating. She also showed symptoms of remarkably sparse verbal output, short-term memory deficits, easy distractibility, excessive sleepiness, and fatiguability. Apart from her behavioral changes, her preoperative diplopia and swelling at the operative site were getting progressively worse. She did not show any signs of limb weakness, primitive reflexes, gait apraxia, or paratonia.

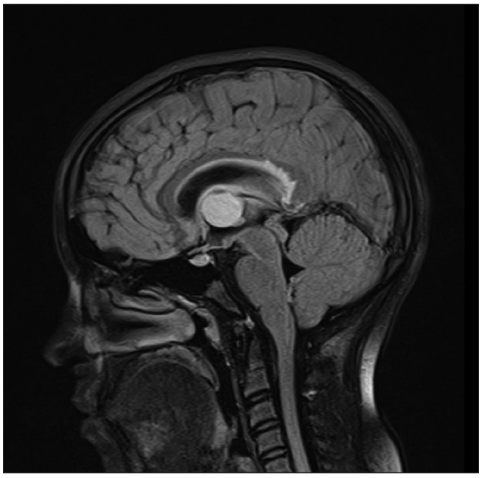

Figure 1: Preoperative magnetic resonance imaging of the brain showing a rounded and sharply demarcated lesion in the third ventricle.

Postoperative brain magnetic resonance imaging (MRI)/computed tomography (CT) were done under sedation and MRI showed evidence of an 8 mm incision in the anterior part of the body of the corpus callosum [Figure 2]. There is also bilateral frontal parafalcine gyral edematous changes along the operative bed that is more on the right side [Figure 3]. It showed mild high signal in the DWIs, FLAIR, and T2WIs without significant changes in the ADC map. A CSF collection localized in the subgaleal scalp was seen. There was also a linear rim of intraventricular subependymal hematoma collection measuring 3.8 × 0.5 cm at its maximal AP and TS dimensions with small petechial foci seen just above the anterior part of the body of the corpus callosum. Size of the lateral ventricles was only slightly prominent, and the other cerebral ventricles were of normal size. There was no evidence of infarction.

She received inpatient dexamethasone 4 mg IV injections every 6 h for a total of 2 days followed by injections every 8 h. Aspiration of the CSF scalp collection was done with removal of 20 mL of clear xanthochromic CSF. Her behavioral symptoms then steadily improved and disappeared after 2 weeks from initiation of therapy. She was discharged on dexamethasone 2 mg PO b. d. for 5 days, followed by dexamethasone 1 mg PO b. d. for another 5 days. Her short-term memory has been slower to improve (as per her parents,

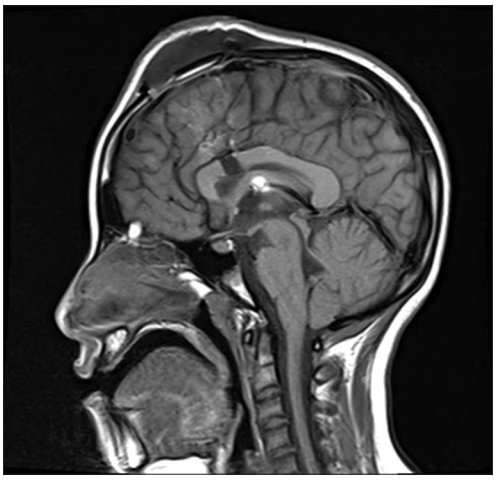

Figure 2: Magnetic resonance imaging of the brain showing evidence of an 8 mm incision in the anterior part of the body of the corpus callosum.

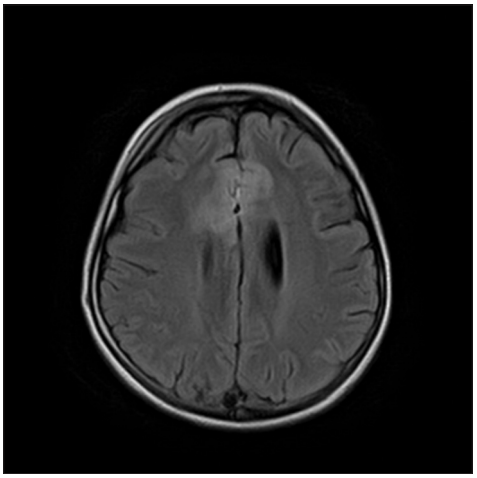

Figure 3: Magnetic resonance imaging of the brain showing bilateral frontal parafalcine gyral edematous changes along the operative bed that is more on the right side.

there has been "95% improvement in memory") and residual memory deficits are still present 6 months after the surgery, such as trouble remembering where she placed objects or memorizing word passages. According to her parents, she does not remember her episode of behavioral disturbance or most of the events that occurred during her second admission to the hospital. However, she recalls that she "was not talking much because she was afraid that she would say something wrong." Her diplopia has resolved completely. At school, she maintains good academic performance.

DISCUSSION AND CONCLUSION

Although the number of callosal fibers is determined at around birth, myelination and functional development of the corpus callosum continues into adolescence in normal children.[16] The rate of callosal growth seems to differ among males and females and is variable across different regions of the corpus callosum. Callosal maturation occurs with alternating phases of growth, reflected by an increase in callosal thickness due to increased myelination and shrinkage, which may be attributed to axonal redirection and pruning.[3,5]

There is a growing body of literature demonstrating the association between weakened integrity of the corpus callosum and behavioral abnormalities. For instance, children with AgCC have been shown to exhibit autistic-like behaviors in the realm of social interaction and communication, such as difficulty in initiating and sustaining conversation with patterns of speech that is "meaningless" or "out of place," recognition of emotion as well as interpretation of humor and non-literal language expressions.[2,18] Other well-described behavioral problems seen in children with AgCC include aggressive behavior, somatic symptoms, attention deficits, and thought problems.[14] Several reports have noted the presence of anti-social behavior, impulsivity, and emotionally unstable personality disorder as well as features of behavioral disinhibition.[12,17,19]

Further studies demonstrating the role of callosal integrity in regulating behavior show that individuals with antisocial personality disorders have underlying structural callosal abnormalities, such as increased callosal white matter volume and length and increased interhemispheric connectivity, as well as a reduction in callosal thickness.[15] Other studies have shown an association between subtle structural changes in the corpus callosum and behavioral deficits in neurodevelopmental disorders including autism, attention deficit hyperactivity disorder, and schizophrenia.[7]

In our present case, during the surgery for the third ventricular colloid cyst removal, anterior interhemispheric dissection was first done but was difficult and time consuming due to extensive thick arachnoid bands and gyral overlap. An 8 mm incision was then made in anterior part of the body of the corpus callosum to enter the right lateral ventricle. The colloid cyst was seen to be completely obstructing the foramen of Monro. The cyst was punctured and thick yellow granular fluid was released into the suction tip. Due to the complex thick attachments, the wall of the cyst was dissected off the fornix and deep cerebral veins with great care. Eventually, the cyst delivered in-toto through the callosotomy. Postoperative brain MRI/CT showed no significant pathology, with only mild frontal parafalcine gyral edema worse on the right that can be expected to occur postoperatively.

Our patient has a number of unique features. Her memory deficits are not unusual given that the fornices might have been involved as the walls of the cyst were attached to and dissected from the fornix. However, severe neuropsychological impairment, and behavioral disinhibition in particular, is unusual after such limited

callosotomy. In light of the corpus callosum's role in behavior, it is possible that transection of the anterior callosal body fibers contributed to the features of behavioral disinhibition seen in our patient, especially given that she is at the critical age for callosal maturation and may be structurally and functionally vulnerable to surgical insult.

Another factor that might have potentially contributed to her behavioral symptoms, whether in isolation or in conjunction with disruption of callosal fibers, was postoperative edema involving her prefrontal cortex, which is responsible for maintaining social appropriateness among other complex cognitive functions such as reasoning, decision-making, and personality expression, as well as the anterior cingulate cortex, which is also involved in inhibition.[4,10] This would explain her marked improvement with IV dexamethasone over the period of 2 weeks as it helped to decrease brain edema. However, postoperative edema is not uncommon and it would be very unusual for mild swelling of the frontal lobe to result in such severe behavioral symptoms.[8,13] Henceforth, considering the discrepancy between the expected clinical presentation of postoperative edema and the atypical clinical picture of this patient, it is possible that her behavioral symptoms might have been caused by a combination of etiologies involving multiple parts of her brain, including the corpus callosum, prefrontal cortex and anterior cingulate cortex.

To the best of the authors' knowledge, this is the first report in literature to describe behavioral disinhibition occurring as a sequelae to a surgical procedure involving callosotomy. Our finding emphasizes the importance of taking into account behavioral complications when considering callosotomy and ensuring that risk for such a complication is conveyed as part of the informed consent for performing callosotomy.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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