

Pediatric Obsessive-Compulsive Disorder Exacerbation and Obstructive Hydrocephalus: A Case Report

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We present the case of an 11 year-old boy with a previous history of obsessive-compulsive disorder (OCD), who experienced a dramatic and acute worsening of OCD symptoms in temporal association with obstructive hydrocephalus secondary to a tectal low-grade glioma. Management and resolution of the hydrocephalus was temporally associated with an improvement in his OCD compulsion symptoms. The present case does not establish proof of cause and effect, but highlights potential multifactorial influences on OCD onset and clinical course. Cortico-striatal-thalamic-cortical pathways, physically distorted by hydrocephalus in this case, have long been implicated in OCD etiology. Clinical implications include the importance of conducting an appropriate neurologic work-up to rule out biological causes for acute and dramatic OCD exacerbations with neurologic signs, even in the context of preexisting OCD. Given that neurologic lesions may exist in the absence of typical signs and symptoms, that they may further disrupt OCD circuitry, and that treatment may lead to resolution of associated psychiatric symptoms, it is important to remain cognizant of these differential diagnoses.

BACKGROUND

Obsessive-compulsive disorder (OCD) is a common and debilitating neuropsychiatric disorder affecting between 0.5% and 4% of the population.^{1,2} Despite its childhood onset in 30% to 50% of cases,³ appropriate diagnosis and treatment are often delayed by years, worsening long-term prognosis. The etiology of OCD is multifactorial and remains to be fully elucidated, including complex genetic underpinnings and potential environmental influences, such as stress and behavioral modeling.^{1,2,4} Specific brain regions implicated in this disorder include the prefrontal cortex, basal ganglia, and cortico-striatal-thalamic-cortical pathways.⁵

Obsessive-compulsive symptoms have been reported as presenting symptoms of neurologic diseases in

adults, including normal pressure hydrocephalus and, less commonly, central nervous system tumors.⁶⁻⁸ Given that such neurologic conditions may exist in the absence of typical signs and symptoms and that treatment may lead to resolution of associated psychiatric symptoms,⁶ it is important to remain cognizant of these differential diagnoses.

In this report, we describe the first reported case of an acute pediatric OCD exacerbation as the presenting complaint for obstructive hydrocephalus and a midbrain, low-grade glioma, followed by temporally associated OCD improvement after ventriculostomy and drainage.

CASE REPORT

An 11 year-old boy with previous diagnoses including OCD,

abstract

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Dr Leung drafted the initial manuscript; Dr Bleakley collected data and reviewed and revised the manuscript; Dr Low coordinated data collection and reviewed and revised the manuscript; Dr Saran was involved in clinical assessment and literature review; Dr Stewart conceptualized and designed the study, supervised data collection, and critically reviewed and revised the manuscript; and all authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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attention-deficit hyperactivity disorder (ADHD), and Tourette syndrome presented to the emergency department with a 2-week history of dramatically worsened obsessions and compulsions. The family endorsed a past history of safety obsessions beginning at age 8 years, followed by impairing and compulsive skin cream use at 10 years old, which led to an OCD diagnosis and initiation of fluoxetine at a dose of 10 mg daily. Before his exacerbation, his OCD had been stable for several months.

In the 2 weeks preceding emergency department presentation, the boy abruptly began to obsess about themes related to morality and religion, including a fear of going to hell. Compulsions included verbalization of specific religious convictions, which, when interrupted, resulted in verbal aggression and violence. He concurrently demonstrated worsening severity of motor and verbal tics, including tongue thrusting and coughing. He engaged in self-harm behavior related to his OCD distress (in the form of hitting himself) and developed passive suicidal ideation. Given the dramatic combination of symptoms described above, he was initially brought to a community-based mental health team. His OCD severity, as rated by the Children's Yale-Brown Obsessive-Compulsive Scale (CY-BOCS) score, was 18/20 for obsessions, 16/20 for compulsions, and 34/40 in total at that time, indicating severe to extreme OCD. After assessment, his fluoxetine dose was increased to 30 mg daily and 1 mg of risperidone daily was added in treatment augmentation.

Before the OCD exacerbation described above, identified life stressors included the death of a distant relative and the temporary absence of the patient's father from the home because of work commitments. His past medical history was unremarkable. His

paternal family history was notable for severe OCD in an aunt, and his maternal family history included an unspecified anxiety disorder in an aunt and unspecified substance use disorders in an uncle.

MANAGEMENT

To manage the extreme OCD exacerbation, the boy was admitted to the inpatient psychiatric unit. Fluoxetine was titrated upwards from 10 to 60 mg daily, and risperidone was replaced with quetiapine, which was titrated to 300 mg daily. Given the intensely dramatic OCD presentation, efforts were made to address potential organic triggers. Given the presence of an elevated anti-streptolysin O titre level, a 2-week course of clindamycin was initiated, in addition to ibuprofen at a dose of 200 mg 3 times a day. There was no clear change noted after antibiotic or ibuprofen trials.

Regarding nonpharmacologic OCD treatment, the patient was given 8 sessions of cognitive behavioral therapy (CBT) as an inpatient, incorporating exposure and response prevention (ERP), mindfulness, and OCD externalization strategies. ERP success was limited by the patient's agitated state and by the relative absence of replicable triggers. At discharge after 6 weeks, safety and violence concerns had resolved, although ongoing daily obsessional thoughts and compulsive behaviors persisted. Despite intentions to continue CBT efforts in the community after discharge, no sessions were conducted.

INVESTIGATIONS

Routine serology identified a slightly elevated potassium level of 5.1 mmol/L with normal values for thyroid stimulating hormone, prolactin, liver function tests, antinuclear antibody, C-reactive protein, anti-double-stranded DNA

antibody, extended electrolytes, cholesterol panel, amylase, and serum copper. He had an elevated anti-streptolysin O titer equal to 593 IUs. On physical exam, neurologic findings were reported as unremarkable, with the exception of potential ptosis and vertical nystagmus noted in his right eye. These ophthalmologic signs were not found on subsequent exams, which reported normal fundoscopy with no reported evidence of papilledema.

Despite nonlocalizing neurologic findings, the lack of response to CBT, ERP, high-dose SSRI, and atypical antipsychotic augmentation led to requesting an MRI of the brain. MRI of the brain identified a small superior tectal mass, demonstrating high T2/ fluid attenuated inversion recovery (FLAIR) and low T1 signal intensity with central hypointensity, suggestive of punctate central hemorrhage or calcification (see Fig 1A). The mass measured 11 × 11 × 8.5 mm (transverse × anterior-posterior × craniocaudal), causing proximal aqueduct obstruction and resulting in hydrocephalus with dilated lateral and third ventricles. The floor of the third ventricle was displaced inferiorly with anterior displacement of the optic chiasm and lateral terminalis. No other abnormalities were noted.

SURGERY

The neurosurgery team was consulted during admission subsequent to the MRI findings, and surgery was conducted 3 months postdischarge. On the day before this procedure, the patient's CY-BOCS score was reported to be 20/40, with an obsession subscore of 10/20 and compulsion subscore of 10/20, in keeping with moderately severe OCD. This represents an improvement of 41% compared with initial admission. A right endoscopic third ventriculostomy was performed to manage his hydrocephalus. The

tectal mass was not removed. He recovered postoperatively without complications.

OUTCOME AND FOLLOW-UP

The patient was managed 4 months postoperatively by neurology and neurosurgery services, at which time he had a normal neurologic exam. MRI at that time demonstrated improved ventricular size and motion compared with the previous scan, with no visible defect from the ventriculostomy (see Fig 1B). The patient and family reported decreased OCD symptoms, despite the fact that his fluoxetine dose had been tapered to 40 mg daily in the first 2 months postoperatively (because of suspected behavioral activation side effects). When reassessed by the pediatric OCD program at 4 months postventriculostomy, he had a CY-BOCS score of 10/40 and no compulsions (0/10), indicating a very mild, and nearly subthreshold, level of OCD, and an additional 50% improvement in severity. There was no noted improvement of tics or ADHD symptoms, with ongoing impulsivity, motoric hyperactivity, and distractibility, and fluoxetine was further decreased to 20 mg daily. In written communication 1 year after surgery, the mother reported that the patient, "...is doing well. He still has minor OCD thoughts but is functioning at a level so much higher than before."

DISCUSSION AND CLINICAL RELEVANCE

This is the first pediatric report of obstructive hydrocephalus presenting with an acute OCD exacerbation. The temporal association of OCD severity improvement after ventriculostomy and drainage is intriguing, yet only partially fulfills proof of causality. In pediatric populations, there are case reports of patients with brainstem

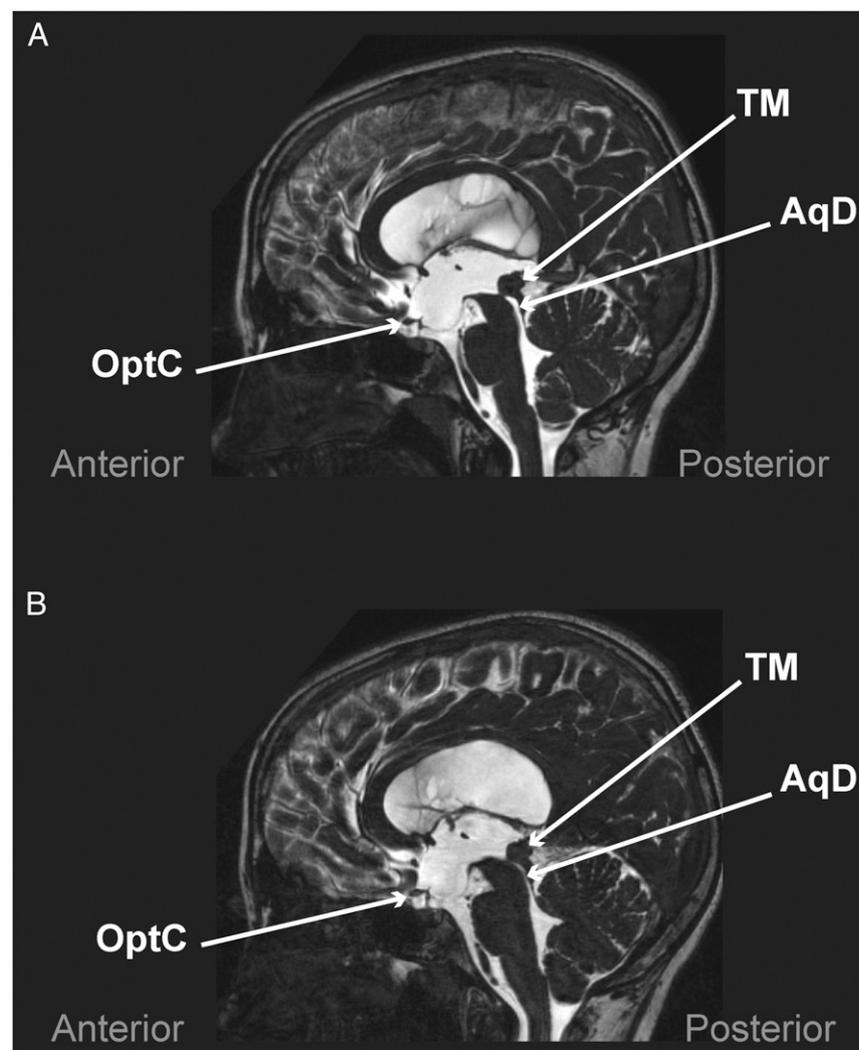


FIGURE 1

T2 weighted brain MRI identifying a small superior tectal mass (TM), demonstrating high signal intensity with central hypointensity, suggestive of punctate central hemorrhage or calcification. A, Pre-ventriculostomy: The mass measures 11 × 11 × 8.5 mm (transverse × anterior–posterior × craniocaudal), causing proximal aqueduct (AqD) obstruction and resulting in hydrocephalus with dilated lateral and third ventricles. The floor of the third ventricle is displaced inferiorly with anterior displacement of the optic chiasm (OptC) and lateral terminalis. No other abnormalities were noted. B, Post-ventriculostomy: Demonstrating less displacement of the third ventricle and OptC.

gliomas presenting with obsessive-compulsive symptoms.^{9,10} Ventricular shunting was performed in only one of these patients, but this was done prior to the onset of OCD and in the absence of hydrocephalus. In adult populations, obsessive-compulsive symptoms have also been reported in patients with normal pressure hydrocephalus, with subsequent resolution of psychiatric symptoms after ventricular shunting.^{6,11,12} The present case highlights the complex and multifactorial potential

influences on OCD clinical course. The patient's OCD initially presented in the context of a positive OCD family history and typical comorbidities, including a tic disorder and ADHD.

Putative mechanisms underlying OCD symptomology include functional and structural distortions of regions involving the prefrontal and parietal cortices and cortico-striatal–thalamic–cortical circuits. For this particular case, initial OCD onset was likely the product

of multiple complex genetic and environmental factors. Although the role of the undiagnosed brainstem glioma with respect to OCD onset is unclear, the concurrence of its resultant obstructive hydrocephalus with an OCD exacerbation, and concurrence of drainage with OCD improvement, are worthy of discussion. Although OCD exacerbation is rarely the presenting symptom to mark the presence of hydrocephalus, neurohistologic changes associated with the latter have been noted to extend throughout the entire cortex and result in behavioral and cognitive deficits.¹³ Despite these deficits at presentation, drainage of hydrocephalus has led to long-term improvements in neuropsychological outcomes in children.¹⁴

The goal of this case report is to report on clinical observations of temporal changes and to contribute to the literature in the domain related to hydrocephalus and OCD, rather than to provide conclusive evidence of cause and effect. OCD severity scores were collected at initial admission (34/40 reflecting severe to extreme illness), 16 weeks later at the time of surgery (20/40 reflecting moderate severity), and 16 weeks subsequent to ventriculostomy (10/40 reflecting mild OCD). This may reflect natural disease course, a result of therapeutic interventions, unknown factors, or a combination of these.

In considering potential contributors to the improvement between initial admission and surgery 16 weeks later, the only consistent, empirically supported OCD treatment intervention was fluoxetine, which was gradually increased from 10 mg to a therapeutic dose of 60 mg daily. In contrast to medication management of anxiety and depression, OCD has a linear dose-response curve, indicating improved likelihood of response at the high

end of dose ranges.¹⁵ Moreover, the time required for an adequate trial is between 8 and 12 weeks, which could account for ongoing improvement between discharge and surgery.¹⁶ Despite attempts, only limited patient engagement in CBT was achieved during admission, and CBT community-based follow-up did not occur as planned on discharge.

Multiple potential contributors to the observed improvement between surgery and follow-up 4 months later should also be considered. These include empirically proven treatment effects, potential impacts of the surgery, and unknown factors. Because of concern over potential activation side effects, fluoxetine was decreased from 60 to 40 mg during this period, which is unlikely to have led to OCD gains and may have logically been expected to cause symptom worsening. Moreover, ADHD symptoms (which resemble behavioral activation) did not resolve with the dose decrease. No CBT sessions were conducted during this period, negating the potential for this influence. Regarding surgical impact, postoperative MRI did demonstrate resolution of hydrocephalus, although the relationship between this change and OCD symptom improvement is not proven via a single case study.

The marked improvement of CY-BOCS scores by 50% in the 4 months after surgery is remarkable, with standard definitions of clinically significant treatment response ranging between 25% to 35% on this measure.¹⁶ The differential postoperative improvement of compulsions (100%) but not obsessions (0%) is subject to speculation. Brain structure differences have been correlated with obsession (parietal myelination and axonal density) versus compulsion (cingulum and basal ganglia) Yale-Brown Obsessive-Compulsive Scale scores.¹⁷ Intriguingly, the latter deep brain structures are more proximal to

ventricles and thus more vulnerable to effects of hydrocephalus, which could hypothetically account for preferential improvement in compulsion scores in this situation.

In conclusion, this patient presented with an acute, dramatic OCD exacerbation in the context of previously undetected astrocytoma and hydrocephalus. He experienced partial improvement with inpatient admission, an adequately dosed SSRI trial, and an attempt at CBT, shifting his OCD from the severe/extreme to the moderately severe range. In the months after ventriculostomy and hydrocephalus drainage, with an SSRI dose decrease and no additional CBT sessions, his OCD severity continued to improve to a mild level. Although this may reflect natural OCD disease course or other unconsidered factors, it is possible that the emergence and resolution of hydrocephalus played a role in impacting OCD severity. Hence, even in the context of a known OCD diagnosis, dramatic or atypical exacerbations warrant consideration of previously undiagnosed structural pathology.

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ABBREVIATIONS

ADHD:	attention-deficit hyperactive disorder
CBT:	cognitive behavioral therapy
CY-BOCS:	Children's Yale-Brown Obsessive-Compulsive Scale
ERP:	exposure and response prevention
OCD:	obsessive compulsive disorder
SSRI:	selective serotonin reuptake inhibitor

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