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Challenges of managing a first episode of pediatric catatonia

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To the Editors:

Pediatric catatonia is a rare and dangerous syndrome, associated with an elevated risk of premature death and poor psychosocial prognosis (Cornic et al., 2009). The underlying etiologies of pediatric catatonia are manifold, including an array of medical and psychiatric illnesses. There is an association with developmental disorders (Consoli et al., 2012).

The treatment of pediatric catatonia, as with adult catatonia, generally begins with benzodiazepines, of which lorazepam has the most evidence (Hauptman and Benjamin, 2016). Very high doses of lorazepam, as high as 14 mg per day, have been used successfully to treat pediatric catatonia (DeJong et al., 2014). Controversy surrounds using neuroleptics: some studies suggest they are effective, while others suggest they worsen catatonic symptoms. Stronger dopamine blockade has been associated with precipitation of catatonia, favoring second-generation neuroleptics over first-generation (Benarous et al., 2018).

There is scant literature on the long-term management of patients presenting with catatonia. The extended use of benzodiazepines to prevent recurrence of catatonic symptoms, for as long 14 months, has been reported (Manjunatha et al., 2007). Benarous et al. (2018) advise that neuroleptics "should be discontinue [sic] at the acute phase of catatonic episode and should be used with caution to treat the underlying psychiatric disorders when catatonic symptoms are stabilized, with regular clinical assessment for motor side effects".

We present a case of a 14-year-old male with developmental delay who was transferred to our inpatient adolescent psychiatric unit from a general pediatric unit at a children's hospital. In the two years prior to admission, the patient was noted to have periodic three-day episodes of melancholic affect, tearfulness, social withdrawal, and sleep disturbance. A year prior to admission, while visiting family in the Dominican Republic, the patient was noted to have sleep disturbance, irritable mood, and auditory hallucinations. The patient was noted to pace around and sing loudly and frantically, reportedly to drown out the hallucinated voices. He was observed at a hospital, received no psychotropic medications, and had resolution of his symptoms.

Two weeks prior to admission, he developed progressive depressed mood, social withdrawal, and recurring auditory hallucinations, culminating in tremulous rigidity and verbal unresponsiveness. The patient was admitted to a general pediatric unit for catatonia, where he was treated with lorazepam. His treatment team at this outside hospital resolved his symptoms with a total daily dose of 18 mg of lorazepam. An extensive work-up for medical etiologies of catatonia was negative.

On initial assessment on our adolescent psychiatry unit, the patient was noted to be drowsy but had no signs or symptoms of catatonia. His Bush Francis Catatonia Rating Scale score was 0 (Bush et al., 1996). Lorazepam was gradually tapered. We initiated olanzapine for treatment of a presumed underlying mood disorder with psychotic features, and gradually increased the dose. See Fig. 1.

On day 12, the patient was found rigid, tremulous, tachycardic, and hypoxic with an oxygen saturation of 82% on room air. He was agitated and aggressive. He was brought to the emergency department and given 0.5 mg of lorazepam intramuscularly, and his symptoms rapidly improved. He returned to the adolescent psychiatric unit. Subsequently, the total daily dose of lorazepam was increased and the dose of olanzapine was decreased. He remained stable leading up to and following discharge on this regimen.

There are multiple diagnostic and treatment challenges inherent in a case of pediatric catatonia. The syndrome itself is recognizable, but clarifying the underlying etiology is critical to inform treatment. We faced two distinct diagnostic scenarios: first, determining what precipitated the initial episode of catatonia; second, determining what precipitated the re-emergence of presumed catatonic symptoms during the hospitalization.

On admission, medical illness had been ruled out, and the patient did not appear to have active symptoms of a mood disorder or catatonia. Diagnosis had to be made from the history provided by the parents, the patient, and report from the initial hospital's clinicians. Psychotic symptoms were described, but other aspects of the patient's presentation

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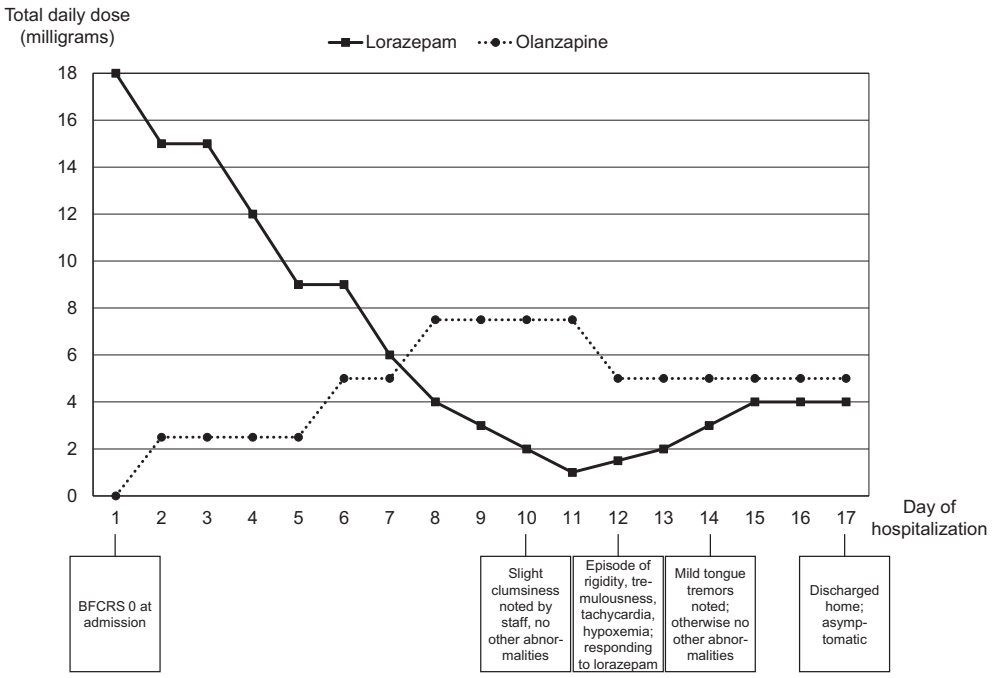

Fig. 1. Symptoms and total daily doses of lorazepam and olanzapine over the course of the patient's hospitalization.

were more ambiguous. The patient's melancholic affect and social withdrawal appeared to represent depression, but could have represented negative symptoms of schizophrenia. (The patient's family clarified that this withdrawn behavior did not represent his developmental delay.) The patient's sleep disturbance, expansive affect, and singing could have represented mania, but could have represented an excited catatonia or psychosis.

Thus, unless a very precise history is available, distinguishing between an underlying unipolar depression with psychotic features, bipolar disorder with psychotic features, or primary psychotic disorder may not be possible. Despite the risk of worsening or re-precipitating catatonia, a second-generation neuroleptic is the best treatment option in this situation, as it is an effective treatment for any of these three potential underlying etiologies. We chose olanzapine, as it is a strong 5-HT2 antagonist and moderate D2 antagonist (Kapur et al., 1998).

The re-emergence of catatonic symptoms occurred in the setting of lowering the benzodiazepine dose and increasing the neuroleptic dose. We considered three etiologies: catatonia due to insufficient treatment of the underlying psychiatric illness, neuroleptic-induced catatonia, and withdrawal from benzodiazepine. The first and second explanations were directly at odds with one another. The third could have explained the episode independent of catatonia and did not exclude the first two hypotheses (i.e., we considered a multifactorial explanation). Furthermore, benzodiazepine withdrawal has been a reported trigger of catatonia, even after use duration shorter than two months (Lauterbach et al., 2009).

Each etiology would call for a different intervention. Insufficient treatment of psychotic disorder would require a dose increase of olanzapine; neuroleptic-induced catatonia would require a dose reduction (and extra-pyramidal symptoms would require an anticholinergic); benzodiazepine withdrawal would require additional lorazepam, as would recurrence of catatonia due to rapid lorazepam taper. Our patient had not demonstrated active symptoms of underlying mood or psychotic disorder on the unit, whereas the dose of olanzapine had been increased and the dose of lorazepam had been tapered relatively quickly. Thus, we surmised the most likely explanation was a combination of the second and third hypotheses. It was not possible to be certain about the explanation; in clinical practice, such certainty is rare. The patient's response to pharmacologic titration was the best evidence supporting our hypotheses.

In summary, we recommend the following: 1) perform an exhaustive medical work-up has for pediatric catatonia; 2) consider both underlying mood and psychotic disorders; 3) clarify the patient's history, including if there is any baseline developmental delay, but recognize there may be limited specificity provided; 4) second-generation neuroleptics may be the best choice for maintenance medication, and should be titrated slowly and independent of other major treatment changes; 5) tapering of benzodiazepines should be performed slowly and independent of other major treatment changes.

Conflict of interest

The authors report no known conflicts of interest.

Contributors

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