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Pharmacologic management of adolescent catatonia: A dual-case series

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Abstract

Introduction: Catatonia is a syndrome characterized by psychomotor and behavioral disturbances and is associated with a substantially increased mortality risk in adolescent patients. There is a dearth of published literature describing treatment strategies for pediatric patients with catatonia. This dual-case series will describe the treatment course of 2 adolescent patients with catatonia at our pediatric inpatient psychiatric facility.

Case Series: This case series presents 2 adolescent patients (a 17-year-old male and a 16-year-old female) who initially presented with worsening agitation and paranoia, later developing catatonia. Both patients required long durations of hospitalization and were treated with high-dose lorazepam before requiring the addition of electroconvulsive therapy (ECT).

Discussion: Treatment of pediatric patients with catatonia creates a significant burden on patients, families, and the healthcare system. Treatment with high-dose benzodiazepines is high risk, while ECT is both difficult to access and comes with its own risks. Both patients discussed are transitional age, meaning they will soon be young adults who will continue to require high-level psychiatric care. Psychiatric pharmacists have a large role to play in ensuring safe medication management for these complex patients.

Conclusions: This case series of 2 adolescent patients with catatonia demonstrates marginal reduction in symptoms with high-dose lorazepam in conjunction with ECT, with minimal side effects. This case series adds to the limited available literature regarding treatment of catatonia in pediatric patients and highlights the need for further study into effective treatment alternatives.

Keywords: catatonia, pediatrics, child and adolescent psychiatry, lorazepam, benzodiazepine, ECT

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Introduction

Catatonia is a syndrome characterized by psychomotor and behavioral disturbances, ranging from unresponsiveness to agitation (Table), associated with developmental disorders, psychotic disorders, mood disorders, and medical disorders.[1,2] Malignant catatonia is a potentially life-threatening form of catatonia in which patients present with autonomic instability, including changes in temperature, blood pressure, pulse, and respiratory rate.[1] Adolescent catatonia has been associated with a substantial increase in mortality, with 1 study observing a 60-fold increased risk of premature death, including death by suicide, for adolescents treated for catatonia when compared to age- and sex-matched controls.[3] Given the scarcity of published literature describing the treatment of catatonia in the pediatric population, treatment for these patients must be determined using adult literature combined with case reports of pediatric patients and provider clinical experience.

TABLE: Diagnostic criteria for catatonia associated with another mental disorder (catatonia specifier)[*1]

The Clinical Picture is Dominated by 3 (or more) of the Following Symptoms:	
Stupor	No psychomotor activity; not actively relating to environment
Catalepsy	Passive induction of a posture held against gravity
Waxy flexibility	Slight, even resistant to positioning by examiner
Mutism	No, or very little, verbal response to instructions or external stimuli
Negativism	Opposition or no response to instructions or external stimuli
Posturing	Spontaneous and active maintenance of a posture against gravity
Mannerism	Odd, circumstantial caricature of normal actions
Stereotypy	Repetitive, abnormally frequent, non–goal-directed movements
Agitation	Notably not influenced by external stimuli
Grimacing	
Echolalia	Mimicking another's speech
Echopraxia	Mimicking another's movements
Catatonia associated with another mental disorder (catatonia specifier) may be used when criteria are met for catatonia during the course of a neurodevelopmental, psychotic, bipolar, depressive, or other mental disorder	

*Adapted from DSM-5 diagnostic criteria for catatonia (pg. 119-122).[1]

In adult patients with catatonia, benzodiazepines (BZD) are the first-line treatment, with electroconvulsive therapy (ECT) considered in severe cases or in those with inadequate response to BZD treatment alone. Lorazepam has been the most widely studied BZD for treating catatonia, but optimal dosing and titration schedules are not well-defined. A BZD challenge of 1 or 2 mg of lorazepam is often used to confirm catatonia diagnosis.[2] Lorazepam is then initiated at 3 mg/d and titrated based on response and tolerability.[4,5] There is no clearly defined maximum total daily dose, with some sources citing 24 to 30 mg/d.[4–6] Catatonia severity is assessed using the Bush-Francis Catatonia Rating Scale (BFCRS), a validated rating scale comprised of 23 items, each rated from 0 to 3 (scores range from 0 to 69, with higher numbers indicating greater severity).[7]

This dual-case series will describe the treatment course of 2 adolescent patients with catatonia treated with high-dose lorazepam and adjunctive ECT to assess the efficacy of these treatment modalities and demonstrate the importance of the pharmacist's role in care.

Case Series

Patient A

A 17-year-old male patient with no pertinent past medical history was admitted to an outside hospital for 19 days after experiencing new-onset psychosis, social withdrawal, and agitation for 1 month. Upon discharge, he was prescribed oral lorazepam 2 mg every 8 hours (6 mg/d) for catatonia, as well as fluoxetine 20 mg and aripiprazole 10 mg daily for mood and psychotic symptoms. He presented to our inpatient psychiatry unit the following day for worsening catatonic symptoms, including urinary incontinence, mutism, grimacing, immobility, negativism, and paranoia.

The patient's initial BFCRS score was 16 (Figure). Aripiprazole and fluoxetine were discontinued for concerns of worsening catatonia and mania, respectively. Lorazepam was increased to 4 mg every 4 hours (24 mg/d) over 1 week with observed improvements in mobility and speech. However, his paranoia and auditory/visual hallucinations (AVH) intensified, so aripiprazole was restarted and titrated to 10 mg daily.

After 8 days, the patient's average BFCRS score was 11. His movement and speech continued to improve with minimal improvements in AVH, so aripiprazole was transitioned to olanzapine. Catatonia assessments with BFCRS became difficult to complete because of somnolence and paranoia. His sedation improved as lorazepam was tapered to 3 mg 4 times daily (12 mg/d), but he continued to display significant anxiety, paranoia, and recurring nightmares that developed into Capgras delusions. Fluoxetine was reinitiated and titrated to 40 mg daily to target depression and anxiety. Catatonia continued to improve with BFCRS scores of 1 to 2 with minimal adjustments to lorazepam or olanzapine.

On hospitalization day 60, a significant decompensation occurred over 24 hours, with symptoms of worsening paranoia, mutism, muscle rigidity, urinary incontinence, diaphoresis, and tachycardia. Given the concern for neuroleptic malignant syndrome versus malignant catatonia, olanzapine and fluoxetine were held, and the patient was transferred to the medical unit. Upon transfer, the BFCRS score was 24. Lorazepam was retitrated to 24 mg/d over 4 days, and the patient was emergently scheduled for ECT 3 times weekly. His BFCRS score improved to 9 after 2 ECT sessions, though his mental status continued to fluctuate, with continued AVH and delusional thought content. Olanzapine was restarted and titrated to 10 mg nightly while maintaining a daily lorazepam dose of 24 mg. After 6 ECT sessions and 1 week of olanzapine 10 mg, his BFCRS score was consistently 1 to 3, so he transferred back to inpatient psychiatry.

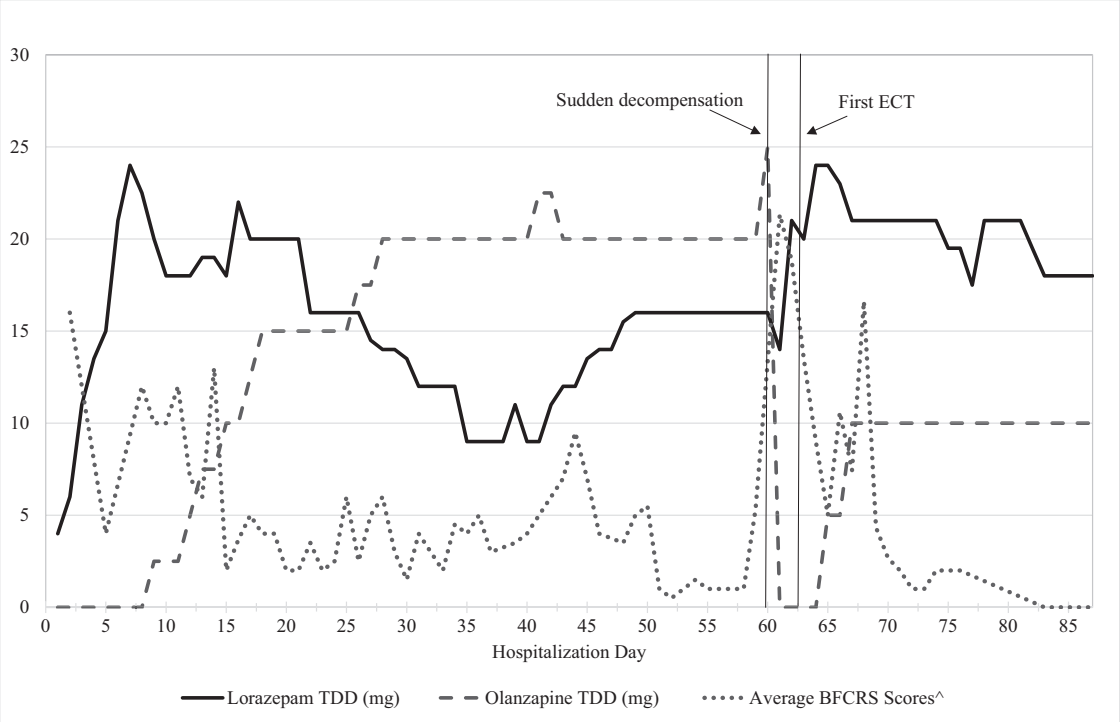

FIGURE: Patient A: Relationship between Bush-Francis Catatonia Rating Scale (BFCRS) scores and medication doses by hospitalization day. ^Average of Bush-Francis Catatonia Rating Scale (BFCRS) scores taken on that date. This number may not accurately reflect the changes throughout days as the number of data points each day may vary each day due to: (1) patient refusal, (2) inability to complete full assessment, and (3) patient's clinical status (eg, more frequent assessments during clinical decompensation).

By hospitalization day 87, the patient had 5 days of BFCRS scores of 0 and displayed improved insight into his Capgras delusions. He had responded poorly to attempts to simplify the lorazepam regimen, so 3 mg every 4 hours (18 mg/d) was continued at discharge, along with olanzapine 10 mg nightly. His guardians confirmed their ability to maintain this dosing frequency and were provided thorough medication education. Outpatient follow-up included continued ECT and participation in a partial hospitalization program.

Patient B

A 16-year-old female with a history of depression requiring inpatient treatment presented to our facility for AVH, paranoia, agitation, and a lack of attending to activities of daily living in the setting of recent trauma. She was discharged on quetiapine 25 mg nightly after a 4-day hospitalization as her guardian declined further dose titration, citing a preference for nonpharmacologic management. After 3 weeks, the patient re-presented with ongoing paranoia and AVH in the setting of medication nonadherence. Quetiapine was titrated to 200 mg nightly with notable improvements in paranoia and mood; however, her guardian declined further titration, and the patient was discharged against medical advice before complete symptom resolution.

Four days later, the patient re-presented for paranoia and disorganized behavior. Quetiapine was cross-tapered to paliperidone 6 mg nightly; however, she became more withdrawn and disorganized after several days of paliperidone therapy. Paliperidone was ultimately discontinued after she experienced an oculogyric crisis, which was effectively treated with intramuscular diphenhydramine 50 mg. The patient's presentation continued to worsen with the emergence of echopraxia, verbigeration, impulsivity, and negativity, causing suspicion of catatonia. She began repeating phrases such as "I am going to take a shower" without action to do so. She spent hours attempting acrobatics despite noticeable grimacing, labored breathing, and frequent falls. On hospitalization day 8, a BFCRS was completed with an initial score of 28. She was given 2 mg of oral lorazepam for suspected catatonia and was noted to be calm, alert, oriented, and logical 30 minutes later. Lorazepam was subsequently initiated at 2 mg orally 4 times daily (8 mg/d) and the following day, her BFCRS score had decreased to 12.

Over the next week, lorazepam was titrated to 3 mg 5 times daily (15 mg/d), though the patient continued to display scrambled speech, purposeless movement, and frequently attempted acrobatics. She was determined to be at high risk for injury or rhabdomyolysis due to her constant movement, in combination with poor fluid intake and difficulty

drawing labs or obtaining accurate vital signs. On day 17 of hospitalization, ECT was emergently initiated with a goal of 3 sessions weekly. Her BFCRS scores consistently ranged from 15 to 18 over the next several days. She appeared "stuck" at times while hyperactive at others, spontaneously screamed and disrobed, and was perseverative on topics.

By hospitalization day 27, the lorazepam dose was alternating 5 mg twice daily, and 6 mg twice daily (22 mg/d), and the patient had received 4 sessions of ECT, with BFCRS ranging from 8 to 12. After the ninth ECT treatment, she began to intermittently refuse ECT; however, her catatonia symptoms continued to improve. By hospitalization day 56, lorazepam was tapered to 5 mg 3 times daily (15 mg/d), and she had received 12 sessions of ECT, with BFCRS scores ranging from 5 to 10. She continued to present as hyperactive, paranoid, and disruptive. However, as she had turned 18 during this hospitalization, the treatment team evaluated the risk versus benefit of transferring her to an adult inpatient psychiatric unit versus discharging her home. She was discharged home on hospitalization day 57, with planned outpatient psychiatry follow-up and continued ECT.

Discussion

Limited published literature exists describing the treatment of adolescent catatonia. Our literature review was completed via PubMed using the following keywords: pediatric, adolescent, child, or children with catatonia or catatonic. A 2015 prospective naturalistic cohort study described the treatment course of 66 adolescent patients with catatonia. Fifty-one patients received a BZD, and they were considered effective for 65% of those. Most patients given a BZD received lorazepam (n = 38, 74.5%) at an average dose of 5.35 mg/d (up to 15 mg/d). Bilateral ECT was administered for 18% of the patients (n = 12) at two to three sessions per week. The mean BFCRS score decreased from 21.01 to 7.82 by discharge. Fewer side effects were associated with BZD than ECT.[8]

Case reports have also been published describing a positive treatment response with higher doses of lorazepam in pediatric patients with catatonia, consistent with adult recommendations. A 2021 case study describing a 12-year-old male with catatonia reported titrating the lorazepam dose to 24 mg/d with a positive response.[9] A 2021 case series describing 2 adolescent patients with autism spectrum disorder and catatonia reported continued benefit of lorazepam up to doses of 22 and 24 mg/d, although both patients later received ECT because of incomplete treatment response with BZD alone.[10]

Here, we add to the currently available literature, presenting 2 cases of previously healthy adolescents who required long hospitalizations and extensive medical workups and were ultimately diagnosed with catatonia. Both patients required treatment with high-dose lorazepam and ECT, with incomplete resolution of catatonic symptoms at the time of discharge.

Catatonia is not a primary diagnosis but a manifestation of an underlying psychiatric or medical disorder.[1] Treatment of catatonia takes initial priority as it can be life-threatening, with a plan to target the underlying disorder once catatonia has resolved. Case A was able to be treated simultaneously for psychosis with an antipsychotic agent; however, Case B did not tolerate the addition of an antipsychotic by the time of discharge. Both patients will likely continue to require high levels of psychiatric care for their underlying diagnoses as they transition into adulthood.

Psychiatric pharmacists play a significant role in ensuring the safe treatment of patients with catatonia, especially those requiring high-dose lorazepam therapy. Multiple, varying daily doses with frequent titrations lead to a high risk of serious medication errors. As described in the above cases, some patients will require both lorazepam and ECT. This presents a challenge as BZD raises the seizure threshold, making ECT more difficult and potentially decreasing efficacy.[11] Psychiatric pharmacists can play an important role in ensuring overall safe medication management during concurrent ECT by evaluating patients' entire medication regimens and assessing them in relation to the medications used during ECT treatment.[11]

Inpatient psychiatric pharmacists should also help bridge transitions of care for all patients, especially those discharged after hospitalization for catatonia.[12] Patient and family education are particularly important and should include action steps in the case of missed lorazepam doses, such as returning to the emergency department if 2 or more consecutive doses are missed or refused by the patient. This discussion must also be transparent about the risk of lorazepam withdrawal symptoms, including seizures. Some families feel more confident discharging home with a prescription for a seizure rescue medication, such as midazolam intranasal spray.

Access to psychiatric care continues to be a challenge for patients, especially children and adolescents.[12] Strain on patients, families, and the healthcare system is significant in these cases as they may require lengthy hospital stays. The financial and social burden of this care is significant.[13] Insurance companies may require prior authorizations for high-dose BZD therapy so psychiatric pharmacists can partner with outpatient pharmacies and providers to ensure access to medication. Access to interventional care, such as ECT, is not always available for the patient, especially in rural areas, and laws vary from state to state as to age restrictions, consent requirements from patient and guardian, and which healthcare professionals may administer ECT.[14]

Conclusion

This series of 2 adolescent patients with catatonia demonstrates a reduction in symptoms with high-dose lorazepam in conjunction with ECT, with minimal side effects. This case series adds to the limited available literature regarding the treatment of catatonia in pediatric patients and highlights the need for further study into effective treatment alternatives.

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