

CASE REPORT

Recurrent catatonia in a patient with bipolar disorder and polycythemia: A case report

Tiffany Eatz^{1*}, Jessica Mikolowsky¹, and Lisa Oliveri-Naidu^{1,2}¹Department of Psychiatry, University of Miami Miller, School of Medicine, Miami, Florida, United States of America²Department of Psychiatry, Miami VA Healthcare System, Miami, Florida, United States of America

Abstract

We report the diagnostic and management challenges encountered in a case of a 57-year-old male patient with bipolar I disorder and comorbid polycythemia secondary to obstructive sleep apnea, who was treated in our inpatient psychiatric unit for suicidal ideation and recurrent catatonia. He was found to have catatonia with a Bush–Francis Catatonia Rating Scale score of 18. He subsequently exhibited a positive response to the lorazepam challenge test. Before discharge, his daily lorazepam dose was titrated to 6 mg, which led to resolution of catatonic symptoms, and was tapered to discontinuation. During the second hospitalization, the patient's catatonia returned, but due to lorazepam-induced bradycardia, he refrained from taking more than 1 mg of the medication. Divalproex sodium and memantine were initiated as off-label catatonic treatment plus lurasidone for bipolar depression. The patient was also found to have a positive antinuclear antibody titer (1:160). The hemato-immunological abnormalities present in our patient complicated his catatonic condition, underscoring the need to explore medical and neuropsychiatric catatonic factors for accurate diagnosis of this neuropsychiatric syndrome.

***Corresponding author:**Tiffany Eatz
(t.eatz@med.miami.edu)

Citation: Eatz T, Mikolowsky J, Oliveri-Naidu L. Recurrent catatonia in a patient with bipolar disorder and polycythemia: A case report. *J Clin Basic Psychosom.* 2024;2(4):4140. doi: 10.36922/jcbp.4140

Received: July 4, 2024**Accepted:** September 25, 2024**Published Online:** November 4, 2024

Copyright: © 2024 Author(s). This is an Open-Access article distributed under the terms of the Creative Commons Attribution License, permitting distribution, and reproduction in any medium, provided the original work is properly cited.

Publisher's Note: AccScience Publishing remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

Keywords: Catatonia; Bipolar disorder; Polycythemia; Neuropsychiatry; Antinuclear antibody

1. Introduction

Catatonia is a neuropsychiatric condition characterized by various motor and behavioral abnormalities, such as immobility, reduced verbal communication, unusual actions, and impaired volition.^{1,2} It is crucial to differentiate catatonia from other psychiatric disorders, including different types of mood disorders, psychotic conditions with significant negative symptoms, and selective mutism.^{3,4} Catatonia presents in different forms, including stuporous, excited, malignant, and neuroleptic-induced types.^{3,5} Originally classified under schizophrenia, catatonia is now recognized in association with various medical and psychiatric conditions. Initially, in the 1800s, Kahlbaum described catatonia as a psychomotor syndrome associated with different underlying disorders.⁶ Shortly thereafter, Kraepelin instead defined catatonia as a manifestation of dementia praecox (an obsolete term for schizophrenia) rather than a separate illness.⁷ Approximately one century later, this erroneous classification was rectified.⁷ Today,

approximately 2.7 – 17% of acute psychiatric patients demonstrate catatonia, a setting-dependent statistic.⁸

2. Case presentation

A 57-year-old Caucasian male with a history of bipolar I disorder (BD1) and polycythemia was evaluated by our psychiatric team for depressive symptoms and suicidal thoughts after being admitted to hospital due to loss of consciousness. Initial neurological assessments, including magnetic resonance imaging of the brain and electrocardiogram for syncope and stroke, were normal. Laboratory results showed elevated hemoglobin (18.9 g/dL) and hematocrit (57%), with decreased erythropoietin levels (2.5 mIU/mL). He tested negative for a *JAK2* V617F mutation. Arterial blood gas analysis showed a partial pressure of carbon dioxide ($p\text{CO}_2$) of 35.0, a partial pressure of oxygen ($p\text{O}_2$) of 79.7, and pH of 7.432. Following therapeutic phlebotomy, he was transferred to the psychiatric unit and started on lurasidone 40 mg daily for bipolar depression. The patient had stopped receiving psychiatric treatment for 30 years and had recently experienced social and economic stressors, including a career change and moving in with family. Notably, the patient had never undergone any preventative psychotherapy or counseling.

During his psychiatric stay, he was diagnosed with catatonia, with a Bush–Francis Catatonia Rating Scale score of 18. His symptoms included immobility, mutism, echolalia, as well as abnormal willpower and behavior. He exhibited a positive response to the lorazepam challenge test. A daily 6 mg lorazepam led to the significant resolution of the catatonic symptoms, and lorazepam was tapered before discharge after 2.5 weeks of hospitalization.

Two days later, the patient returned with worsening bipolar depression. We increased lurasidone to 60 mg, but catatonia reappeared 1 week thereafter. Lorazepam was reintroduced into the treatment regimen, but due to bradycardia, he could not tolerate doses of lorazepam above 1 mg. No rigidity, fever, or leukocytosis was observed in the patient, and normal levels of ferritin and creatinine phosphokinase were recorded. Due to logistic and administrative obstacles, electroconvulsive therapy (ECT) was not performed due to inability; otherwise, he may have been a candidate. Divalproex sodium and memantine were initiated as alternative treatments.

The second hospital admission lasted 3 weeks, during which the patient's condition stabilized following the administration of tolerable doses of lorazepam. During the second hospital stay, other potential medical causes were explored given the recurrence of catatonic signs after a prolonged hospital stay and stabilization. In addition,

the patient initially presented with a syncopal-type event and had been psychiatrically stable and functioning in the community without medication for years before this presentation. Further investigation revealed a positive antinuclear antibody (ANA) titer of 1:160, which warranted a rheumatology consultation. No additional systemic issues were identified, and the patient's hemoglobin level and hematocrit remained stable. The patient showed signs of gradual improvement and was discharged with plans for follow-up care. A timeline of the patient's overall clinical course is shown in [Figure 1](#).

3. Discussion

Catatonia can arise from various medical conditions, and benzodiazepines are used as the primary treatment approach for this syndrome.⁹ The polycythemia described in this case may be related to obstructive sleep apnea and a positive ANA titer, but its implications for the development of catatonia in this patient remain unclear. This patient did not display delirium, which often coexists with catatonia,⁹ and his laboratory results were otherwise negative for metabolic derangements.

Alternative treatments for catatonia include memantine, anti-epileptic drugs, and amantadine.⁹ Memantine and amantadine are used primarily in schizophrenia spectrum disorders, while anti-epileptics such as carbamazepine and valproic acid may be beneficial in mood disorder cases.¹⁰ It has been reported that patients with catatonia and underlying mood disorders positively responded to carbamazepine in a daily dose of 100 – 1,000 mg, without taking benzodiazepines concurrently.¹⁰ Valproic acid, in a daily dose of 600 – 4,000 mg, has also been used as an effective monotherapy in patients with excited catatonia complicated by schizophrenia spectrum illnesses.¹⁰ Daily topiramate at a dose of 200 mg, together with benzodiazepine as an adjunct, has also been used to treat catatonia in four patients, as described in one case series.^{10,11} The application of levetiracetam¹² and zonisamide¹³ for the treatment of catatonia has also been conducted and described in the literature.¹⁰

ECT is a viable option for refractory or severe cases involving malignant features, malnourishment, and severe depression, but it was not accessible in this instance.⁹ We believe that our patient would have been a potentially suitable candidate for ECT if it was available in our facility.

Regarding further pharmacological treatments, antipsychotics are generally avoided in patients with catatonia due to the heightened risk of neuroleptic malignant syndrome or worsening catatonia. Despite this, if needed, a second-generation antipsychotic should be used. Clozapine is the recommended antipsychotic to be

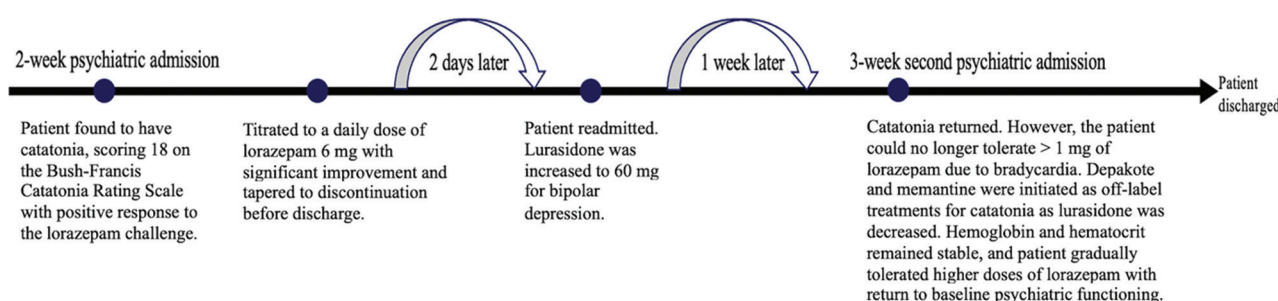


Figure 1. Timeline of inpatient clinical course

used in catatonia owing to the highest levels of efficiency and efficacy demonstrated in previous research.¹⁴⁻¹⁷ Coupling of the increased dopamine release in the striatum through 5-HT_{1A} receptor stimulation and low D2 receptor occupancy may produce a net effect on overall dopamine neurotransmission, which is potentially responsible for the anticatatonic effects of clozapine.¹⁴ Despite advantages in treating catatonia, the use of clozapine requires a baseline evaluation, slow titration, and close monitoring.¹⁴ Lurasidone and cariprazine may be useful for mood disorder-related catatonia,¹⁸ which justifies its use in our patient.

Hypoxia, induced by obstructive sleep apnea, may exacerbate catatonia. One case in the literature involved a 20-year-old male patient with central hypoventilation presenting with resistant catatonia.¹⁹ He had a history of hypoventilation at birth and was supported by 24-h mechanical ventilation for the first 5 years of his life.¹⁹ During hospitalization, laboratory result indexes, including complete blood count, of this patient were normal, as opposed to polycythemia accompanied by an increased hemoglobin and hematocrit identified in our patient. A fluorodeoxyglucose positron emission tomography scan during hospitalization showed a hypometabolic distribution, which is characteristically consistent with hypoperfusion.¹⁹ Increased mechanical ventilation successfully resolved his psychiatric symptoms.¹⁹ Despite the rarity, hypoxia from (central or obstructive) apnea is possibly linked to catatonic symptoms in our patient. This association may imply that increased PCO₂ and decreased PO₂ (the latter of which was seen in our patient) may have pathophysiological implications for catatonic symptoms. Phlebotomy can increase oxygenation and thus serve as a treatment for polycythemia, which holds the potential, in addition to lorazepam, to resolve catatonia symptoms.

In patients with positive ANA value of 1:160, autoimmune causes and/or comorbidities should be explored. Immune dysregulation is the mechanistic driver for a variety of neuropsychiatric disorders, such

as narcolepsy, dementia, depression, and psychosis.³ Autoimmune disorders such as anti-N-methyl-D-aspartate (NMDA) receptor encephalitis and hyponatremia with subsequent extrapontine myelinosis secondary to Addison's disease have been cited as triggers of catatonia.³ In some cases, autoimmune disorders appeared to be the proximate cause of catatonia (NMDA receptor encephalitis); in other cases, the autoimmune disorder was a more distal cause (Addison's disease).³

The major limitations of the current case were patient's loss to follow-up and non-attendance at scheduled mental health, rheumatology, and hematology outpatient visits. In addition, there is limited literature on catatonia with comorbid polycythemia and hematological-immunological disorders. More case reports and studies are needed to enhance our understanding of these associations.

4. Conclusion

Managing recurrent catatonia in a patient with BD1, polycythemia, and a positive ANA titer presents complex challenges. The presence of hemato-immunological abnormalities adds a layer of complexity to the detection and management of catatonic symptoms, underscoring the need for comprehensive exploration of the neuropsychiatric and systemic factors of catatonia to help strengthen our understanding of the neuropsychiatric condition.

Acknowledgments

None.

Funding

None.

Conflict of interest

The authors declare that they have no competing interests.

Author contributions

Conceptualization: All authors

Investigation: All authors

Writing–original draft: Tiffany Eatz
Writing–review & editing: All authors

Ethics approval and consent to participate

Verbal consent of the patient was obtained for participating in this case study.

Consent for publication

Verbal consent of the patient was obtained for releasing his data in this paper. No photographs or images of the subject were included for publication. All data of the subject were deidentified to safeguard his privacy and anonymity.

Availability of data

Data are available from the corresponding author upon reasonable request.

Further disclosure

The current case has been presented at The Academy of Consultation-Liaison Psychiatry annual meeting on November 10, 2022.

References

1. Brar K, Kaushik SS, Lippmann S. Catatonia update. *Prim Care Companion CNS Disord.* 2017;19(5):16br02023.
doi: 10.4088/PCC.16br02023
2. Walther S, Stegmayer K, Wilson JE, Heckers S. Structure and neural mechanisms of catatonia. *Lancet Psychiatry.* 2019;6(7):610-619.
doi: 10.1016/S2215-0366(18)30474-7
3. Rogers JP, Pollak TA, Blackman G, David AS. Catatonia and the immune system: A review. *Lancet Psychiatry.* 2019;6(7):620-630.
doi: 10.1016/S2215-0366(19)30190-7
4. Peralta V, Cuesta MJ, Serrano JF, Mata I. The Kahlbaum syndrome: A study of its clinical validity, nosological status, and relationship with schizophrenia and mood disorder. *Compr Psychiatry.* 1997;38(1):61-67.
doi: 10.1016/S0010-440X(97)90055-9
5. Rasmussen SA, Mazurek MF, Rosebush PI. Catatonia: Our current understanding of its diagnosis, treatment and pathophysiology. *World J Psychiatry.* 2016;6(4):391-398.
doi: 10.5498/wjp.v6.i4.391
6. Hirjak D, Foucher JR, Ams M, *et al.* The origins of catatonia-systematic review of historical texts between 1800 and 1900. *Schizophr Res.* 2024;263:6-17.
doi: 10.1016/j.schres.2022.06.003
7. Fink M, Shorter E, Taylor MA. Catatonia is not schizophrenia: Kraepelin's error and the need to recognize catatonia as an independent syndrome in medical nomenclature. *Schizophr Bull.* 2010;36(2):314-320.
doi: 10.1093/schbul/sbp059
8. Francis A, Fink M, Appiani F, *et al.* Catatonia in diagnostic and statistical manual of mental disorders, fifth edition. *J ECT.* 2010;26(4):246-247.
doi: 10.1097/YCT.0b013e3181fe28bd
9. Denysenko L, Sica N, Penders TM, *et al.* Catatonia in the medically ill: Etiology, diagnosis, and treatment. The academy of consultation-liaison psychiatry evidence-based medicine subcommittee monograph. *Ann Clin Psychiatry.* 2018;30(2):140-155.
10. Beach SR, Gomez-Bernal F, Huffman JC, Fricchione GL. Alternative treatment strategies for catatonia: A systematic review. *Gen Hosp Psychiatry.* 2017;48:1-19.
doi: 10.1016/j.genhosppsych.2017.06.011
11. McDaniel WW, Spiegel DR, Sahota AK. Topiramate effect in catatonia: A case series. *J Neuropsychiatry Clin Neurosci.* 2006;18(2):234-238.
doi: 10.1176/jnp.2006.18.2.234
12. Muneer A. Aripiprazole in the treatment of refractory mood disorders: A case series. *Clin Psychopharmacol Neurosci.* 2014;12(2):157-159.
doi: 10.9758/cpn.2014.12.2.157
13. Nakagawa M, Yamamura S, Motomura E, Shiroyama T, Tanii H, Okada M. Combination therapy of zonisamide with aripiprazole on ECT-and benzodiazepine-resistant periodic catatonia. *J Neuropsychiatry Clin Neurosci.* 2012;24(3):E9.
doi: 10.1176/appi.neuropsych.11080191
14. Tabbane K, Halayem S, Joobar R. Clozapine for the management of persistent catatonia. *J Psychiatry Neurosci.* 2016;41(6):E81-E82.
doi: 10.1503/jpn.150352
15. Dursun SM, Hallak JE, Haddad P, *et al.* Clozapine monotherapy for catatonic schizophrenia: Should clozapine be the treatment of choice, with catatonia rather than psychosis as the main therapeutic index? *J Psychopharmacol.* 2005;19(4):432-433.
doi: 10.1177/0269881105053313
16. England ML, Ongür D, Konopaske GT, Karmacharya R. Catatonia in psychotic patients: Clinical features and treatment response. *J Neuropsychiatry Clin Neurosci.* 2011;23(2):223-226.
doi: 10.1176/jnp.23.2.jnp223
17. Chattopadhyay S, Saha I, Dan A, Bhattacharyya K. Clozapine

responsive catatonia: A series of five cases. *Ind Psychiatry J*. 2012;21(1):66-68.

doi: 10.4103/0972-6748.110955

18. Baldessarini RJ, Vázquez GH, Tondo L. Bipolar depression: A major unsolved challenge. *Int J Bipolar Disord*. 2020; 8(1):1.

doi: 10.1186/s40345-019-0160-1

19. Dranovsky A, Needleman JP, Sylvester J, VanHeertum R, Muskin PR. Progressive paranoid psychosis in a 20-year-old with central congenital hypoventilation syndrome. *Pediatrics*. 2014;134(3):e900-e902.

doi: 10.1542/peds.2013-2740