CATATONIA IN ADOLESCENCE: A Case Report

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ABSTRACT

The pathogenesis of catatonia is poorly understood and it can be fatal without effective treatment. Therefore, a swift diagnosis is necessary to treat this condition. It has been rarely described in children and adolescents. In a literature search, we have found only one reported case of excited catatonia described in a 16-year-old girl. In the following case report, we discuss a 16-year-old boy who presented with bipolar disorder-manic with catatonia. Through this case report, we hope to highlight some key points in the diagnosis and management of catatonia.

INTRODUCTION

Catatonia has been rarely described in children and adolescents.[1] Cohen, et al., estimates the incidence of catatonia (all syndrome types) in children and adolescents to be 0.16 million per year.[2] In psychiatric populations, this frequency differs greatly, ranging from 0.6 to 17 percent.[2,3] This, however, is significantly lower than estimated adult frequencies of 7.6 to 38 percent.[4] Literature reviews in child and adolescent literature have revealed only a limited number of reported catatonia cases, allowing the treatment of catatonia to remain vague.

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We report the case of a 16-year-old boy who presented with characteristics of excited catatonia. When conducting a thorough search on Pubmed, we found only one additional case report of excited catatonia in adolescence.[5] This report emphasizes the need for more data about the etiology and treatment of catatonia, especially in the pediatric population.

CASE REPORT

Presentation. A 16-year-old boy with a history of attention deficit hyperactivity disorder (ADHD) and a mild learning disability diagnosed at age 12 presented to our emergency department with altered mental status. The patient exhibited symptoms of disorganization and was talking in word salad. He had auditory and visual hallucinations as well as paranoid delusions. His gait was festinating and unsteady and he exhibited muscle stiffness without cog-wheeling. The patient also presented with mild hypertension and tachycardia.

Past medical history. The patient had been treated for ADHD at age 12 with atomoxetine for three months. The medication was discontinued following successful treatment, as the family felt he had "outgrown his ADHD." There was no prior history of mania or psychosis and the patient was functioning well until a month before the current presentation.

A month before the current presentation, the patient described to his parents an incident at school of someone putting a sexually explicit note in his book. The school teacher denied the incident ever took place. A few weeks after this, he called the police while his parents were out. The police found the patient to be incoherent and disorganized, and he was taken for psychiatric assessment. This led to his first psychiatric admission.

He was admitted for two weeks to the psychiatric facility. He presented with subacute onset of rapid speech, grandiosity, and reduced sleep. He was initially tried on a low dose of olanzapine, which was switched to quetiapine (300mg twice daily) and divalproex sodium (500mg twice daily) because of extrapyramidal symptoms (EPS) (severe dystonia). He again developed mild neck stiffness on quetiapine, which improved upon adding benztropine (1mg twice daily). The patient improved on these medications and was discharged in a stable state on quetiapine (30mg twice daily), divalproex sodium (500mg twice daily), and benztropine (1mg twice daily). He was diagnosed with bipolar disorder, manic episode.

After discharge, he did well for 4 to 5 days and returned to school. However, his condition gradually deteriorated again, and his family found him increasingly confused with muscle stiffness leading to unstable gait. He was then evaluated by his outpatient psychiatrist who referred him to our emergency room with concern of neuroleptic malignant syndrome (NMS).

Upon this current presentation to the emergency room at our facility, the psychiatry team evaluated him and found him to be floridly psychotic (paranoid delusions along with auditory and visual hallucinations) and disorganized. He suffered from severe stiffness along with rapid eye blinking, unusual head and neck movements, and peculiar behavior (simulating guitar playing). His parents additionally reported depressed mood and reductions in sleep, interest, energy, appetite, and concentration, as well as psychomotor retardation.

Background. The patient was enrolled in the tenth grade and was doing well in classes. There was no history of use or abuse of illicit drugs or alcohol by the patient. This was confirmed by a negative urine toxicity screening. There was no history of legal issues. The patient was supported by both parents.

Family history was remarkable for maternal bipolar disorder. No history of chronic or recent medical illnesses were present.

Mental status exam. Upon presentation, the patient was drowsy and disheveled. He had disorganized speech and talked in word salad, which made it very difficult to understand him. He maintained poor eye contact and did not establish rapport. His mood was anxious and fearful, while affect was constricted and flat. He exhibited disorganized thoughts, paranoid delusions, and auditory and visual hallucinations. The patient denied suicidal and homicidal ideations. Based on his vocabulary and educational background, the patient's intellectual function appeared to be below average. Insight was impaired, and judgment was poor.

Formulation and differential diagnosis. A thorough medical workup, including imaging in the emergency room, was unremarkable. A neurology consult indicated that neurological etiology was unlikely.

The patient was administered lorazepam 1mg intravenously, and there was a significant improvement in stiffness and gait that lasted for approximately 30 minutes. Rapid cognitive and motor response to intravenous lorazepam suggested a working diagnosis of catatonia with bipolar disorder, manic subtype, influenced by his past history of mania.

We additionally considered NMS as a possible diagnosis because of his history of EPS on an antipsychotic medication. However, absence of fever and presence of normal creatine kinase went against this diagnosis. Other laboratory tests, including serial blood counts, metabolic panels, amylase, lipase and valproic acid levels, were also normal.

Our differential diagnosis on admission was delirium, neurological disease, NMS, bipolar disorder with catatonia, and psychosis not otherwise specified (NOS).

Hospital course. The patient was admitted to the psychiatric unit. Vital signs included heart rate of 105/minute and a blood pressure of 130/55mmhg. His tachycardia was attributed to dehydration, but the etiology of the hypertension was uncertain at this time. We continued hydration and administered lorazepam 1mg intramuscular every six hours as needed.

Catatonic symptoms mildly responded to lorazepam, so the dosage was increased to 1.5mg every six hours intravenously (IV) on Day 3 and 2mg every six hours IV on Day 4. However, even after a week of admission, the patient continued to be disorganized, manic, and psychotic. Thereafter, we initiated aripiprazole 5mg twice daily to help with the mood lability and psychosis.

There was only a mild improvement in psychosis after starting aripriprazole; however, we were very cautious in increasing the dose as the patient had experienced severe side effects (dystonic reactions) on antipsychotics in the past. Benztropine 0.5mg p.o. b.i.d was therefore administered. Psychosis slowly improved by Day 10, but the mood lability continued so we added lithium carbonate 150mg twice daily to help with his manic state. Gradually the medications were increased for maximum therapeutic benefit. He was now on lithium carbonate (controlled release formulation) 450mg twice daily and aripiprazole 10mg twice daily. This further led to improvement in both psychosis and mania.

During this time, ziprasidone 20mg intramuscularly (IM) every eight hours and benztropine 0.5 IM every eight hours had been suggested as needed in case of agitation; however, after titration of lithium and aripiprazole, he did not require any PRN medications. By Day 16 of admission, the catatonia had improved to the point of remission, thus allowing us to discontinue the lorazepam. The patient was discharged shortly thereafter from the unit on aripiprazole 10mg b.i.d., lithium carbonate (controlled-release formulation) 450mg p.o. b.i.d., and benztropine 0.5mg b.i.d.

TABLE 1. Associated features of catatonia and NMS[6]

CLINICAL SIGNS	CATATONIA	NMS
Hyperthermia	Yes	Often
Motor Rigidity	Yes	Yes
Mutism	Yes	Yes
Negativism	Often	Yes
Altered Consciousness	Yes	Yes
Stupor or Coma	Yes	Yes
Autonomic Dysfunction	Yes	Often
Tachypnea	Yes	Often
Tachycardia	Yes	Often
Abnormal BP	Yes	Yes
Diaphoresis	Yes	Yes
LABORATORY RESULTS	**CATATONIA**	**NMS**
CPK elevated	Yes	Often
Serum iron reduced	Yes	Probable
Leucocytosis	Yes	Often

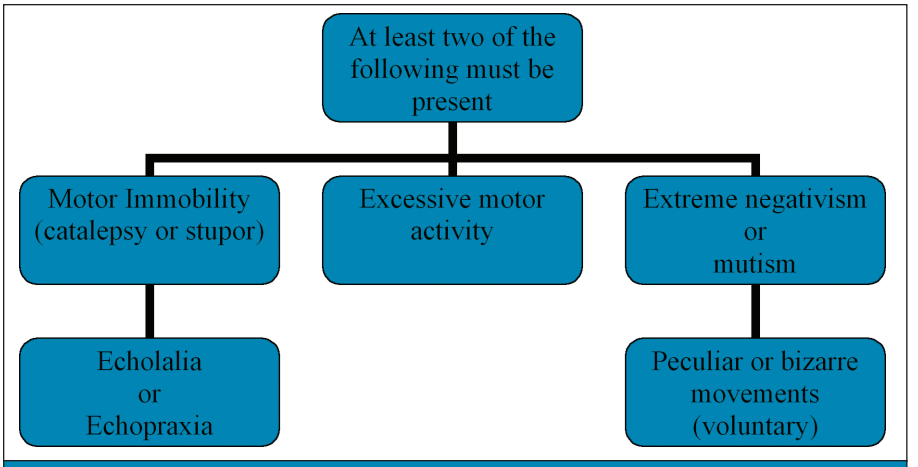


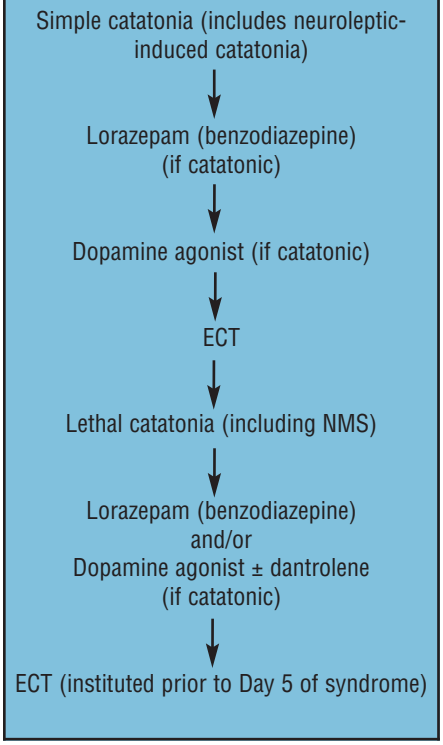
FIGURE 1. DSM-IV-TR criteria for mood disorder with catatonic features

Multiaxial diagnoses. The patient was diagnosed with the following: *Axis I:* Bipolar disorder, type I, most recent episode manic, severe with psychotic and catatonic features; ADHD by history; *Axis II:* Deferred; *Axis III:* None; *Axis IV:*

TABLE 2. Principles of management of catatonia[8]

1. Early recognition—patient must be closely observed and vital signs must be taken frequently.
2. Supportive care—hydration, nutrition, mobilization, anticoagulation, precautions against aspiration
3. Discontinue antipsychotics and other drugs that worsen catatonia.
4. Restart dopamine agonists, especially in Parkinsonian patients.
5. Supportive measures
6. High index of suspicion for medical complications

TABLE 3. Treatment of catatonia[9]



Suffering from serious mental illness; *Axis V:* GAF—30-21 (Unable to function in almost all areas, e.g., stays at home, in ward, or in bed all day without taking part in social activities or severe impairment in communication [e.g., sometimes incoherent or inappropriate]).

PERTINENT ISSUES/QUESTIONS FOR DISCUSSION

Descriptions of catatonia can be traced back to 1849 when Bell reported 40 cases of patients who presented with concurrent mania, psychosis, delirium, overactivity, and sleeplessness, then termed *Bell's Mania*.[5] Very little is known about phenomenology, etiology, and treatment of Bell's Mania.[5] More recently, this syndrome has been called *delirious mania* or *excited catatonia*.[5] Under current parameters, these symptoms would be classified as excited catatonia; however, Fink and Taylor believed there was no distinction between the two.[7,8]

Catatonia is diagnosed by the presence of at least two of the following symptoms: motoric immobility, excessive purposeless motor activity, extreme mutism or negativism, peculiarities of voluntary movement, echolalia, or echopraxia.[9] *The Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision* (DMS-IV-TR), acknowledges catatonia to be a syndrome associated with schizophrenia, general medical conditions, and affective disorders.[9] A combination of two or more of these (psychosis, mania, delirium, catatonic states, combative behavior, insomnia, and extreme psychomotor agitation) might lead to extreme exhaustion and death.[7,8] In the 40 cases described by Bell, severe insomnia and psychomotor agitation did lead to exhaustion and death in some of the cases. The etiologies of excited catatonia or delirious mania may parallel malignant catatonia, NMS, and serotonin syndrome (Table 1).[6]

Fink and Taylor hypothesize that excessive motor activity is the key characteristic in diagnosing excited catatonia and possibly malignant catatonia.[8] Our patient was unlikely to be suffering from NMS, as his serial creatine kinase levels were within normal limits. However, we remained watchful before introducing neuroleptics because he had suffered from EPS when exposed to these medications in the past. Previous studies of catatonia in adolescents have predominately shown posturing, stupor, staring, mutism, and dysfunctional neuromotor behavior.[5] Our patient fulfilled the criteria of DSM-IV-TR for catatonic disorder (Figure 1).[9]

Pathogenesis of catatonia is poorly understood. Catatonia can develop in several psychiatric disorders, metabolic disturbances, meningoenchephalitis, and toxicities.[1] As catatonia can be fatal without effective treatment, a swift diagnosis is necessary. Confusing symptoms in children and adolescents make this difficult.

It is hypothesized that GABA-ergic transmission in the orbitofrontal premotor and motor cortices is involved and explains the benefit of GABA-A potentiators like lorazepam.[10] Adult cases have shown electroconvulsive therapy (ECT), benzodiazepines, and, in one case, antipsychotics to be effective in relieving catatonia.[7,8] Catatonia may also arise from glutamate (NMDA receptor) hyperactivity. As such, in the event lorazepam or ECT are not affective, glutamate antagonist therapy with amantadine is a plausible treatment option.[11] See Tables 2 and 3[12,13] for principles of management and treatment of catatonia.

Our patient improved on lorazepam adequately without needing other modalities of treatment, particularly when he received higher doses of IV lorazepam. Eventually he was switched to oral tablets, with continued success. However, because additional medications, such as lithium, aripiprazole, and benztropine, were added, the true efficacy of lorazepam remains elusive. Lorazepam was shown to be an effective treatment in the only other reported incident of excited catatonia in an adolescent.[5]

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