A Case of Pediatric Catatonia

Susan Ciccolari Micaldi, M.D., M.A., Christopher W. Dao, D.O., Dannette N. Napier, D.O., Joycy Samson, M.D. University of Kansas School of Medicine-Wichita, Wichita, KS Department of Psychiatry and Behavioral Sciences

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INTRODUCTION

Karl Ludwig Kahlbaum was the first to describe catatonia in 1868.¹ Although it has been described as an adult condition, Kahlbaum observed that the majority of the adults had their first symptoms of catatonia in childhood, and described catatonia as a syndrome of abnormal motor function.² Catatonic symptoms can be divided into motor (e.g., posturing, catalepsy, waxy flexibility), behavioral (e.g., negativism, mutism), affective (e.g., uncontrollable emotional reactions, withdrawal), and regressive (e.g., enuresis).

Pediatric catatonia usually presents acutely, but its onset can be insidious.² Duration can be brief or chronic for weeks or months. Though schizophrenia was believed, for a long time, to be the major cause of catatonia, the syndrome is now known to occur in a wide range of psychiatric and medical conditions.³ Catatonia in children and adolescents occur most commonly in the context of the schizophrenia spectrum followed by affective disorders. Trauma also plays an important role in the onset of catatonic presentations in youths.²

In addition to psychiatric diagnoses, in more than 20% of cases of pediatric catatonia, an underlying medical condition could be identified.² Systemic autoimmune disorders, such as systemic lupus erythematosus and autoimmune encephalitis, are the two common classes of autoimmune disorders associated with catatonia.

The most common autoimmune encephalitis underlying catatonia are anti-NMDA-receptor (anti-NMDAR) encephalitis and pediatric autoimmune neuropsychiatric disorders associated with streptococcal infection (PANDAS). Various drugs or toxic compounds can cause catatonia in youths, including steroids, lithium, phencyclidine, and cannabis abuse. Furthermore, catatonia can be associated with metabolic and genetic conditions such as Wilson's disease and porphyria.

Regarding treatment, benzodiazepines and, in particular, lorazepam, are the first line treatment for pediatric catatonia. At times, dosing in the range of $15\,\mathrm{mg}$ daily is necessary for resolution of symptoms. Lorazepam treatment needs to be maintained until the underlying cause of catatonia is found and appropriately treated.

Antipsychotic medications, specifically first-generation, worsen catatonia especially in the acute phase.² However, neuroleptics can be used with caution to treat the underlying psychiatric disorders when catatonia symptoms are stabilized. Electroconvulsive therapy should be considered when catatonia does not respond to benzodiazepine treatment.²

CASE REPORT

This case concerned a young patient who, while admitted to our psychiatric adolescent unit, developed symptoms of catatonia in the context of a difficult-to-reach diagnosis due to the patient's lack of verbal communication and detailed prior history. The patient was a 16-year-old African American female in state custody with a complicated and somewhat uncertain psychiatric, psychosocial, medical,

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and family history. She was transferred from a neighboring hospital's pediatric intensive care unit (PICU) to the adolescent inpatient psychiatric unit, after medical stabilization, following a witnessed intentional overdose at her foster mother's home on 82 tablets of her prescribed clonidine 0.1 mg.

The note from the emergency department reported the patient as having a history of schizophrenia and bipolar disorder with multiple past inpatient psychiatric hospitalizations. The patient's home medications were clonidine 0.1 mg three times a day (TID) and risperidone 2 mg two times a day (BID). During her three-day PICU course, she was reported to be uncooperative, primarily nonverbal, with psychomotor retardation, and with an episode of enuresis on the hospital floor. Once deemed medically stable, she was transferred to the psychiatric unit on day four of her medical admission.

According to the case manager, the patient's history was not clear. Her mother had died of cancer six years prior, while she was living with her. The patient also had reported that when she was seven years old and in out-of-state foster care, she was abused physically and sexually by the foster family and would attempt suicide if she were to return to that home. After her mother's untimely death, the patient went to live with her father and two half-sisters, one of which reportedly suffered from schizophrenia. School records indicated that she was a straight-A student until halfway through ninth grade, when she was first psychiatrically hospitalized.

Six months before coming to our unit, the patient's father sent her to live with his friends since she required a high level of care. While staying at the friends' house, she had an episode of acute agitation and assaulted responding law enforcement officers. She was re-hospitalized and taken into state custody.

According to the patient's foster mother, she had lived with her on two occasions. The first time she resided with the foster mother for a few weeks right after entering the foster system (about six months prior to admission to our unit). The medications she was prescribed at that time were unknown. According to the foster mother, during the patient's first stay with her, she was talkative, and she would laugh and joke with the foster mother and other girls in the home. However, the mother had installed nanny cameras throughout the house, and the patient was observed debating with her father over the phone when alone. The patient was removed from the home after assaulting another female foster child residing with the same foster mother.

The second time the patient resided with the foster mother was after discharge from a later psychiatric hospitalization, just five days prior to her admission to our hospital. In contrast to her previous stay, upon return to the foster mother's place, the patient was primarily nonverbal, but was taking care of basic needs. However, this situation dramatically worsened after she resumed contact with her father. Afterwards, she began refusing her medications (risperidone 2 mg BID and clonidine 0.1 mg TID).

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continued.

The week prior to admission to our unit, the patient was described by the foster mother as isolated, pacing, anxious, depressed, and was seen talking to herself. On the day of admission to the PICU, she grabbed the bottle of clonidine from the foster mother's hands and quickly ingested all the pills in the bottle (82 tablets).

Past Psychiatric History. Based on the case manager's report and review of documents from prior hospitalizations, about two years before the patient was admitted to our unit, she began demonstrating aggressive behavior at home at the same time her half-sister with schizophrenia moved in with the family. Six months before her first psychiatric hospitalization (18 months before she was admitted to our unit), her primary care provider (PCP) found a moderate degree of depression and anxiety on Patient Health Questionnaire-9 and General Anxiety Disorder-7 evaluations (sleep and psychomotor symptoms were the most severe). She was diagnosed with Adjustment Disorder with mixed anxiety and depression.

One week prior to her first hospitalization, she reported to her PCP severe anxiety and depression with additional symptoms of fatigue and social withdrawal. She also related that dizziness had begun eight months prior, and that bilateral lower extremities joint pain and numbness had begun three months prior. She also requested contraception and was given a medroxyprogesterone acetate injection. She had no history of psychotic symptoms up to this point.

One week later, following a physical altercation with her father, the patient was admitted for her first psychiatric hospitalization with suicidal/homicidal ideation and psychosis, including auditory and visual hallucinations. Police reports indicated that the patient accused her father of sexually molesting her. She was acting "strange and nonsensical", and was agitated, self-harming, and assaultive with police. The patient made and retracted allegations of sexual assault by her father multiple times throughout that stay. A urine drug screen was positive for methamphetamine.

Throughout that admission, the patient was assaultive towards staff, sexually inappropriate, had enuresis on the floor and chairs, and required intra-muscular benzodiazepines and antipsychotics as needed for agitation and medication refusal. Multiple medication changes were made, and she was cross titrated from chlorpromazine 50 mg four times a day (QID) to discharge medication of olanzapine 15 mg at bedtime (qHS) and benztropine 1 mg daily (QD) with a discharge diagnosis of schizophrenia.

After spending one month in the hospital, the patient was admitted again, shortly after discharge, for another three-week psychiatric hospitalization due to depressive symptoms, including anhedonia, sleep disturbances, low mood, constant suicidal ideation, and critical and demeaning auditory hallucinations. According to those hospital records, her father described her as having symptoms consistent with a "catatonic state" and blamed the medications. The patient again made repeated allegations, with subsequent retractions, that her father had sexually abused her. She was diagnosed with complex post-traumatic

stress disorder (PTSD) and was discharged home to her father on fluoxetine 10 mg QD.

Six months after her initial hospitalization, the patient's younger sister reported to the PCP that she had to assist her with showering and dressing. This was one year before this team saw her. The patient had two more inpatient psychiatric hospitalizations in the following few months with records documenting that she demonstrated varying degrees of avolition, psychomotor retardation, self-care failure, mutism, unresponsiveness, depression, and suicidality.

About one year before being admitted to our hospital, at the funeral of a family member, the patient reportedly exhibited psychotic symptoms, involving a demonic persecutory delusion along with dysregulated behavior, which required inpatient psychiatric admission. Her case manager was unable to obtain those records. Aside from her second hospitalization, when she was prescribed only fluoxetine, the antipsychotic medications ordered at discharge consistently were stopped by her father, with complaints that his daughter was in a "zombified state" and not able to talk.

Records documented additional hospitalizations with suicide attempts via hanging once, intentional overdose twice, as well as self-harm via cutting. Also, a report was documented wherein the patient was found by the police task force to be involved in sex trafficking in exchange for methamphetamine.

Family Medical History. The patient's family medical history included that her father had been diagnosed with depression and rheumatoid arthritis, her half-sister with depression, her other half-sister with schizophrenia, and her maternal uncle with completed suicide. Her mother died of cancer of unknown type.

Treatment Course. Assessment of the patient was hindered by her inability to engage in a diagnostic interview (she remained primarily mute except for occasional one-word replies or rare short sentences) and to provide information about the onset, characterization, and progression of her psychiatric symptoms. On observation, she demonstrated notable latency, psychomotor retardation, significant social withdrawal, blank staring, and flat affect. Given the patient's history of trauma at an early age (e.g., witness to custodial mother dying of cancer when she was seven years old, possible physical and sexual abuse, including sex trafficking), her history of methamphetamine use (with extent unknown), her family medical history, and incomplete past hospitalizations and PCP records, all in the context of her initial clinical presentation, this team formulated the following list of initial diagnoses:

- Unspecified Schizophrenia Spectrum Disorder and Other Psychotic Disorder
- Complex PTSD
- Rule out Major Depressive Disorder, recurrent episode severe, with psychotic features
- Rule out Catatonia associated with another mental disorder
- Rule out Adjustment Disorder with mixed disturbance of emotions and conduct
- Rule out Substance/Medication-Induced Psychotic Disorder (including cognitive and behavioral changes due to past methamphetamine use)

Given the patient's lack of meaningful participation in any interview, it was difficult to discern whether she was exhibiting symptoms of a

Schizophrenia Spectrum Disorder or psychotic symptoms in the context of a severe episode of Major Depressive Disorder. When she first came to the unit, she was eating and, throughout the day, ambulating to and from the day room, though primarily isolating to her room. On her fourth day on our service, a weekend call team felt strongly that she was experiencing psychotic symptoms and responding to internal stimuli. Specifically, she was darting her eyes around as if trying to locate the voices talking to her or the people out to get her. On the same weekend, she required intramuscular haloperidol for an episode of acute agitation. The weekend team restarted the patient's home risperidone, but at the much lower dose of 0.5 mg BID instead of 2 mg BID she was prescribed prior to admission to our unit.

In the couple of days that followed, the patient was started on sertraline 25 mg to address her apparent depressive symptoms, anxiety, and PTSD. During that time, she became very intrusive, resisting staff's attempts to redirect her physically when invading peers' rooms, entering closely into staff and peers' personal space, and staring at them intensely. Staff reported her as "fixated" on certain staff members, following them around and becoming frightening to certain staff and peers. During that time, she also started displaying stereotypies, as she began frequently and gently poking peers, staff, and objects in slow motion and without apparent purpose. She began blocking unit entrances, required "constant redirection", and was placed on one-on-one sitter precaution.

Over the next week, her risperidone was titrated up to 1 mg in the morning (qAM) and 0.5 mg qHS to target psychotic symptoms. On day 10, she began lying on the floor and resisting encouragement to move to her bed. Over the following five days, she progressed to lying on the floor facing her bed, refusing to look or respond to staff, and became completely mute (negativism). Additional behaviors indicative of catatonia developed, including limited oral intake, mannerisms, (e.g., sticking sanitary pads to walls and chewing on them, while refusing to use feminine hygiene products properly during menstruation), and an episode of posturing, Also, on examination, she demonstrated some waxy flexibility.

The neurology service was consulted, but the patient refused multiple attempts by staff to obtain a brain Magnetic Resonance Imaging (MRI) and an electroencephalogram (EEG). The pediatrics service was consulted to monitor the patient's general medical status, including nutrition and hydration, eating nonfood items (Pica), and potential development of decubitus ulcers, and rhabdomyolysis.

Once catatonia became apparent, our team discontinued risperidone and initiated treatment of catatonia, while continuing sertraline 25 mg qAM. Lorazepam was started at 1 mg BID with target doses up to 8 - 12 mg total daily dose, if needed. Challenges were encountered with the treatment of the patient's catatonia. Firstly, our hospital did not have a PICU and the nearby hospital with a PICU did not have an inpatient psychiatry unit or service, outside of consult liaison. Because the patient would not allow for intravenous (IV) administration and due to the potential risk for respiratory depression, lorazepam had to be titrated slowly and administered orally, with intramuscular dosing as needed for refusal. Secondly, the patient experienced symptoms consistent with orthostatic hypotension, though without falls, but refused to allow our team to obtain vital signs. Consequently, intake and output

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continued.

were tracked, and individual psychotherapy was attempted.

The patient's symptoms improved mildly within a day of lorazepam initiation. She tearfully verbalized having nightmares and flashbacks to the nursing team (but otherwise remained primarily mute). She demonstrated some gradual improvement in oral intake, getting off the floor, and ambulating for brief periods of time throughout the day as lorazepam was titrated up to 3 mg PO TID over the next five days.

Dosing was changed to smaller QID doses to avoid sedation and, over the five following days, lorazepam was titrated to 2.5 mg PO QID. The patient showed improvement in eating, moving around, and participating in interviews though primarily with nodding and shaking of her head. Since the nursing staff reported the patient appearing off balance, overly sedated, and refusing vitals, dosing was decreased to 2 mg QID for safety. The dose was increased again to 2.5 mg PO QID five days later, with some additional mild improvement in symptoms, including the patient speaking in sentences at times, improvement in self-care/hygiene, increased ambulation, improvement in cooperation with taking medications, and allowing a brain MRI, but with symptoms fluctuating in severity. The patient continued to require a high level of care with much encouragement to get off the floor and complete activities of daily living.

On day 13, the patient attempted suicide by drowning via sticking her head in the toilet. Meanwhile, the result of the brain MRI without contrast was within normal limits. Laboratory tests were ordered by our team to rule out autoimmune encephalitis as a possible underlying etiology of the patient's catatonic symptoms and sent to the Mayo Clinic laboratory.

Later, lorazepam was titrated up to 11 mg total daily to address the patient's catatonia. During this time, she continued to make suicide attempts, swallowing labels and plastics, while hiding plastic condiment containers in her blankets. When she began showing improvement in her catatonic symptoms, lorazepam was decreased gradually to 2 mg QID over the course of one week. During that time, the patient's laboratory results returned positive for anti-GAD65, N and P/Q type paraneoplastic antibodies, and levels of anti-streptolysin O were elevated. Therefore, the patient was transferred to a pediatric hospital in a neighboring major city to receive workup for autoimmune encephalitis and multidisciplinary care.

While at the pediatric hospital, the patient continued to attempt to swallow plastic items. During that hospitalization, she underwent a battery of tests, including EEG, which showed mild diffuse cerebral dysfunction and encephalopathy. Two attempts were made to complete a lumbar puncture, but failed due to inability to retrieve any cerebrospinal fluid.

Based on the workup completed, the negative MRI result, and a multidisciplinary discussion including the psychiatry, neurology, and oncology services, the patient's symptomatology was deemed not to be due to an autoimmune or oncological etiology, but rather a psychiatric one. Hence, after about one week, she was transferred back to our inpatient psychiatric unit.

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continued.

On arrival, the patient's mutism seemed improved. The treatment team attempted to decrease lorazepam to 6 mg daily in divided doses (QID). However, she gradually became mute again. At this time, lorazepam was titrated back up to 8 mg total daily and a second opinion was obtained with another child psychiatrist to clarify the patient's psychiatric diagnoses. She was diagnosed with Schizoaffective Disorder with Catatonia as the primary diagnosis, but Major Depressive Disorder, recurrent episode severe with psychotic features, remained in the differential diagnosis. Given the patient's multiple trials of previous antipsychotic medications, she was started on clozapine and titrated up to 100 mg PO BID.

A psychologist also was consulted to start the patient on a behavioral plan with a reward system whenever she tended to her activities of daily living and left her room to participate in milieu therapy.

Over the next month, the patient showed gradual improvement in her symptoms and was transferred to a psychiatric residential treatment facility (PRTF) for further stabilization. While at the facility, clozapine was titrated to 150 mg PO BID with good response. Lorazepam was tapered slowly to 0.5 mg PO TID without recurrence of catatonic symptoms. Sertraline 25 mg daily was continued.

The patient's case manager reported that the patient was conversing much better with others. The case manager also related that the patient indicated that her thoughts were clearer, and she wanted to continue taking her medications. She remained at the PRTF while plans were being made for her next living situation.

DISCUSSION

Pediatric catatonia is a rare occurrence with high risk for morbidity and mortality.³ It can occur in children and adolescents in the context of psychiatric and medical conditions.

Our case of pediatric catatonia was in the context of a psychotic disorder whose assessment was complicated by the inability of the patient to participate in a meaningful diagnostic interview (she was mostly nonverbal) and by the lack of collateral information from guardians (the patient entered the foster system a few months prior to admission to our hospital).

The case confirmed the need for psychiatrists to first rule out medical causes of pediatric catatonia, then address catatonic symptoms with, at times, high doses of benzodiazepines carefully titrated and divided throughout the day to reduce side effects.² The need for physicians to be patient regarding emergence of positive response to benzodiazepine treatment and to taper the regimen slowly, once catatonic symptoms have stabilized, was underscored.

Even if the literature indicates that antipsychotics worsen catatonia symptoms, 2 this case seemed to confirm that neuroleptics have a place in the treatment of catatonia: when the patient's catatonic symptoms appeared to have stabilized and a psychotic disorder became the clear underlying etiology, an antipsychotic (clozapine, which has low binding affinity for the dopamine D2 receptor like quetiapine) was added to the treatment regimen with positive results.

Finally, the difficulty of this case needs to be framed in the context of the current state of inpatient hospital care where brief hospitalizations are the mainstay and longer ones are discouraged. The aforementioned case fueled plenty of disagreements and conflicts among the many agencies involved in her care.

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