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CASE REPORT

Chronic Excited Catatonia: A Case Report

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ABSTRACT

The fifth edition of the DSM considers catatonia a separate disorder for the first time. The diagnosis requires the presence of at least three of its twelve characteristic features. Catatonia may occur in the context of numerous medical conditions and psychiatric disorders, including affective disorders and schizophrenia. It is listed as "unspecified" when the nature of the underlying condition has not been elucidated. Recognition of catatonia can be a complicated process, as there is a range of symptoms that can occur in myriad combinations. Catatonia is usually not optimally treated with antipsychotics, which may present a problem in patients who carry a diagnosis of schizophrenia and catatonia. This report describes a case of chronic hyperkinetic (excited) catatonia, a form of catatonia that is not as well-known as other forms. The prominent negativistic features interfered with treatment, (including the refusal of oral medications), and the hyperkinetic activity threatened the patient's health and the milieu of the inpatient unit. Ultimately, high-dose intramuscular lorazepam injections achieved only a partial remission. This report may stimulate increased rates of recognition of hyperkinetic catatonia.

Introduction

Catatonia is described as a psychomotor disturbance that can include an array of symptoms. The exact pathophysiology is unknown and may be heterogeneous. According to the DSM-5, catatonia may be associated with another mental disorder. The diagnosis is present in cases of psychiatric, neurodevelopmental and other medical conditions. The overall prevalence of catatonia is unknown, but over 15% of psychiatric inpatients may meet criteria for the condition [1].

DSM-5 lists 12 features, of which at least three are required for diagnosis: 1) Stupor (absence of psychomotor activity); 2) catalepsy (passive induction of a posture that is then held against gravity); 3) waxy flexibility (slight, even resistance to positioning by the examiner); 4) mutism (which can be selective); 5) negativism (no response or resistance to external stimuli); 6) posturing (spontaneous and active maintenance of a posture against gravity); 7) mannerisms (caricatures of normal actions); 8) stereotypy (repetitive non-goal-directed movements); 9) agitation (not influenced by external stimuli); 10)

grimacing; 11) echolalia; 12) echopraxia. The most common signs of catatonia are immobility, mutism, and refusal to eat [2]. There is no specific feature of catatonia that is required for its diagnosis. Accompanying features include abnormal motor functioning, inability to suppress complex motor activities (such as stereotypy), and even life-threatening physical symptoms such as autonomic instability [3].

Four main types of catatonia have been described, including classic withdrawn, periodic, malignant and hyperkinetic (excited). The classic withdrawn type may include stupor, mutism, negativism, and posturing with minimal movement despite being awake and watchful. Malignant catatonia includes fever and autonomic instability and is a risk factor for neuroleptic malignant syndrome. Periodic catatonia, considered to be a genetic form of catatonia associated with schizophrenia [4], is typified by recurrent brief episodes (days) of hyperkinetic catatonia, separated by asymptomatic intervals. Hyperkinetic (excited) catatonia does not encompass the quintessential stupor that is seen in the classic withdrawn type. Instead, it entails excessive motor activity with no end goal. Other variable features can include disorientation or disorganized speech, violent, aggressive acts and physical injuries without awareness [5]. A recent article highlighted that catatonia is under-diagnosed in general hospitals, and cites agitation and grimacing (among others) as criteria that are often overlooked in patients with catatonia [6], and therefore are less likely to be diagnosed.

Catatonia may be associated with malignant hyperthermia, Parkinson's disease, and metabolic-induced stupor. Additionally, the NMDA receptor antibody may be implicated as a precipitant of catatonia [7]. The differential diagnosis for catatonia can include about 100 different medical conditions, such as metabolic disorders, infections (HIV), autoimmune disorders (systemic lupus erythematosus), paraneoplastic encephalitis, head trauma and stroke. Notably, an overlapping set of features with catatonia is the "Stiff-Person Syndrome," a rare neurological disorder associated with autoimmune disorders which includes sensitivity to noise, touch, and emotional distress, and which can precipitate muscle spasms [8]. Laboratory studies including complete blood count, blood urea nitrogen, creatinine, serum glucose level, thyroid function tests, rapid plasma reagent, vitamins B12 & folate, liver function tests, creatine phosphokinase,

HIV test, serum iron, urine toxicology, urinalysis, electroencephalogram, anti-NMDA receptor antibody, and brain MRI may assist in ruling out organic causes.

Of importance, catatonia is best treated with benzodiazepines and with ECT for medication-refractory cases. Other treatments that are likely less effective and of which there is a small number of studies include the GABA-A agonist zolpidem, Repetitive Transcranial Magnetic Stimulation (rTMS), NMDA antagonists amantadine and memantine, and topiramate, which works on GABA receptors [9]. The response from memantine suggests that glutamate hyperactivity might be related to catatonic symptoms. NMDA hyperactivity may cause dysregulation of GABA-A function. NMDA antagonists indirect-ly restore GABA-A function in the frontal lobes. GABA-A agonists such as zolpidem may correct dysregulation of GABA ergic tone in the orbitofrontal cortex [10].

According to a recent review article, the treatment of catatonia may involve a stepwise treatment algorithm, starting with intravenous lorazepam, followed by ECT, amantadine, an anti-epileptic and ending with an atypical antipsychotic in combination with lorazepam [6]. Often, an unsuccessful lorazepam treatment leads to a treat-ment plan of ECT, especially in hyperkinetic catatonia; however, it is difficult to administer, especially in the context of refusal by the patient or proxy. The third step in the algorithm is amantadine, a glutamate antagonist. Carbamazepine or valproic acid is the next step, followed by aripiprazole (with its partial D2 agonism) or clozapine, atypical antipsychotics that may have a lower chance of exacerbating catatonia, versus typical high potency ones. These atypicals are especially recommended in patients with catatonia and a primary psychotic illness. If this is not sufficient, adding lorazepam to the antipsychotic may be therapeutic.

Notably, there is evidence that catatonia due to a primary psychotic illness such as schizophrenia may be

less responsive to just benzodiazepines [11], and therefore the addition of an atypical antipsychotic to a benzodiazepine may be warranted early on in treatment if benzodiazepines are not successful. As described above, ECT may be complicated to administer. Amantadine may increase the likelihood of psychosis and therefore should be used with caution in schizophrenic patients.

Case Presentation

Ms. X, in her 30s, a woman of Jewish descent, never married, who is a fraternal twin and whose twin is healthy. She was enrolled in a technical graduate school before the onset of schizophrenia and was living with her parents prior to hospitalization. Leading up to this admission, the patient evidenced marked isolation and lack of self-care. She was not eating for days at a time and manifested bizarre behavior, pacing in circles at the edge of a cliff. 911 were called by a bystander. Upon admission, she had disorganized behavior and required many stat medications for agitation. Over time, she was noted to be internally-preoccupied and psychotic. Her symptoms were refractory to risperidone, and she was transferred to a state facility for long-term treatment.

4.1 Past psychiatric history: The patient has had multiple past psychiatric emergency room visits and several hospitalizations, with no history of suicide attempts. According to her family, she began exhibiting bizarre behavior while in technical school and subsequently dropped out, moving back home with her family. She barricaded herself for several days in a storage room of her family's furniture store and barely eating and not showering, refusing to go home. Her family eventually called 911. In the emergency department, she denied suicidal ideation or intent. At the time, she denied perceptual disturbances or paranoia and denied substance use and was discharged home that very day.

Around one year later, she was admitted to the hospital when her family called 911 after she physically assaulted her family members in the context of worsening delusions of persecution, ideas of reference, thought broadcasting and limited food in-take. She was noted to be posturing at the hospital, a symptom of catatonia, and was described as being delusional, irritable, and with impoverished thought content. She was eventually discharged on risperidone after a two-week hospitalization. She was in and out of hospitals several times as a result of bizarre behavior, with hospitalizations leading to minimal lasting improvements.

4.2. Social history: The family described the patient as a shy child growing up, with healthy development and upbringing. There was no significant substance use history except for a remote self-reported history of alcohol use years prior to her psychiatric symptoms. She had no legal history, no family history of psychiatric illness and no exposure to trauma. She graduated high school with honors and enrolled in a technical graduate school.

4.3. Hospital course: During the early weeks of her hospitalization, she was minimally conversant with staff. She remained guarded isolated and had irregular eating habits. She had no in-sight into her illness. Her sparse speech revealed a thought process that was concrete and illogical. She was internally-preoccupied. She became selectively mute, yelling at her family on the phone but having virtually no contact with anyone on the unit. Notably, she was more verbal with a regular cleaning lady on the unit of Jewish descent. She refused treatment with the risperidone and all other medications, would not con-sent to the release of her other records or for laboratory testing, imaging or EEG.

The patient only showered with an administrative order and remained in soiled clothing for days at a time. On the inpatient unit, she increased her motor activity to the point where she exhibited constant motion, incessantly propelling herself at varying speeds around the circularly organized unit, narrowly missing the staff and other pa-tients. She was agitated, walking from morning until night, often demonstrating intermittent grimacing expressions. She would appear exhausted and develop a shuffling gait toward the evening, but would continue to pace the unit perimeter in circles. Her posture was notable for a tilt to the right, not explained by scoliosis or any other medical condition. She had brief periods of sitting still by the windowsill, looking only at her feet

with a blank stare and flat affect, as still as a statue. She ignored all staff requests and was negativistic, not taking medications orally but did not refuse nasogastric tube placement for medication administration (after it was court-ordered). She refused food for long periods and on several occasions underwent emergency hydration and tube feedings. A court order of retention was granted for one year to administer medications to her over her objection. Trials of fluphenazine decanoate, clozapine, and olanzapine were attempted without therapeutic response. Fluphenazine decanoate was discontinued when she developed a shuffling gait and a fixed gaze, became mute and was postur-ing. Benztropine only improved the extrapyramidal symptoms. She next received clozapine via nasogastric tube, as she continued to refuse oral medications, with titration up to 625 mg daily with a therapeutic blood level of 330 ng/mL. She began exhibiting escalating provocative and agitated behaviors, such as taking the house-keeping cart and attempting to spray cleaning supplies throughout the ward, hitting several staff members. On several occasions, she continuously poked a patient and could not be redirected. Intramuscular lorazepam 2 mg lessened her agitation and was observed to facilitate some appropriate verbalizations with the staff. Olanzapine was introduced at 10 mg intramuscularly, which she did not refuse. She became less aggressive thereafter; although her pacing around the unit continued unabated. She did not attend a single group throughout her lengthy hospitalization. She some-times provided one-word answers concerning food; Once, for example, she approached a staff member to ask for a second slice of pizza. Most of the time her eating habits were erratic. At times she did not eat for days, necessitating medical interventions.

4.4. Medical evaluations and interventions: The patient's vital sign assessments and weight measurements were obtained as part of the court order. Most measurements were within normal limits, aside from hypotension attributed to a decreased fluid intake. She refused all medical appointments but was brought to the hospital for intravenous fluid hydration on multiple occasions, averaging once every two months. Her only laboratory abnormality revealed mild anemia. A workup was conducted for Addison's disease, which was excluded by normal ACTH stimulation test results and morning cortisol levels. No EEG or imaging studies could be performed due to her refusal to cooperate. Her pacing eventually led to the development of varicose veins and the patient did not appear to be able to appreciate the connection between her varicose veins and longstanding pacing.

4.5. Diagnosis of catatonia and altered approach: A diagnosis of hyperkinetic (excited) catatonia was considered, and the treatment focus shifted to high doses of benzodiazepines. Her Bush-Francis Catatonia Rating Scale was 22 at the time. She had no signs of extrapyramidal symptoms. Signs such as waxy flexibility were not assessed as she refused physical examination. Eventually, a court order for medications over objection was granted for lorazepam. She was started on lorazepam intramuscularly (as she refused medication per os), which was titrated to a high dose of 4 mg three times per day. Her symptoms of catatonia partially improved, with attenuation of her selective mutism and negativism. For instance, before receiving lorazepam, she did not react to greetings by staff members or other patients and was devoid of any change in facial expression. Afterward, she provided verbal responses to some questions, although she still had not developed spontaneous speech. She spoke slowly in a soft, monotonous rhythm, with a raspy quality. She sometimes answered questions in full sentences, and her affect appeared more reactive. She began making some eye contact, a stark contrast from the negativism she exhibited before the initiation of lorazepam, and would eat on a more regular basis. Her Bush-Francis Catatonia Rating Scale had improved to 8 after the lorazepam trial, versus a high score of 22 prior to the high dose lorazepam regimen. Due to of bruising at the injection site, the lorazepam was tapered off.

After the lorazepam taper, her symptoms of catatonia worsened again, but she did not revert to her original catatonic state. She remained more talkative and less negativistic. When staff greeted her, she replied with 'hi' in a low volume, with a constricted smile, establishing

brief eye contact while continuing to pace, but this time not just in circles, but also up and down the halls.

Discussion

This case of catatonia differs from common presentations that include stupor as a central component. This patient displayed longstanding hyperkinetic catatonia, which is less common and was obscured by her schizophrenia diagnosis. However, she did meet criteria for DSM-5 catatonia, including features of selective mutism, posturing, negativism, agitation, grimacing and stereotypy with the latter evidenced through repetitive, continuous pacing that she exhibited for almost all of her waking periods over several years.

There aren't many studies on excited catatonia, particularly such cases of longstanding excited catatonia. The patient's symptoms only partially remitted upon administration of high doses of standing lorazepam, which did reduce her negativism, agitation, and grimacing. Notably, her verbal output increased, and she regularly produced non-spontaneous speech that included recognition of others and minimal replies to their questions.

This case is noteworthy in that the patient had longstanding catatonia that was hyperkinetic, not adequately responsive to benzodiazepines. Her history also points to catatonic behavior, including negativism, mutism, and stereotypy, and it is unclear how long she had some or all of these criteria. Given the patient's belief that she was not mentally ill and did not need treatment, court mandates for treatment over objection were required, or she may have starved herself to death. While it might be unsurprising that she experienced anger and resentment toward the treatment team, her lack of cooperation can also be explained by her negativism along with her non-linear, irrational thought processes. Negativism prevents the patient from engaging with the treatment team and therefore cooperating with care. Additionally, her lack of engagement with her external environment (negativism) contributed to her lack of insight into the potential medical complications from her condition, including worsening varicose veins. In excited catatonia, the patient may be a danger to self and others, and thereby re-quire hospitalization. Severe psychomotor agitation can potentially lead to hyperthermia, altered mental status, and autonomic dysfunction [12]. The patient did not exhibit any signs consistent with neuroleptic malignant syndrome during her hospitalization; however, she was at an increased risk of developing the syndrome based on her presentation. Notably, the patient had anemia, which increases the risk for neuroleptic malignant syndrome (NMS) [10], which fortunately did not manifest.

The refractoriness of treatment with benzodiazepines in this case of catatonia may speak to its unknown etiology or the chronicity of the condition before treatment. Another critical barrier to treatment is the non-compliance and noncooperative behavior evidenced by such cases, particularly those with prominent negativism, such that catatonia symptoms indirectly impeded their successful treatment. The noncompliance and negativism also hinder the medical evaluation. This case may be consistent with some literature that antipsychotics (particularly of high potency) can exacerbate catatonia, particularly in the absence of a catatonia-focused approach. Clozapine withdrawal has also been associated with the emergence or worsening of catatonia through a proposed mechanism of serotonergic and cholinergic hyperactivity [13], although the patient showed signs of catatonia prior to initiation of clozapine.

At present, the patient remains catatonic, with improved mutism after her treatment course of lorazepam, but the mutism persists significantly non-spontaneous. Early ECT treatment may be justified in cases of excited catatonia [12]. ECT is effective in greater than 80% of all types of catatonia, which includes cases of failure of pharmacotherapy to achieve remission [4]. Even though chronic catatonia is harder to treat, ECT may allow patients to achieve remission and may require more treatments than usual [14].

When a course of ECT may provide relief or improvement in symptoms in desperate circumstances, and a patient has no capacity to understand the risks and benefits of the treatment, a legal decision can be

sought for treatment over objection. ECT is a relatively safe procedure, but not without risks, as it involves general anesthesia, fasting overnight and could foreseeably involve the use of restraints. On the other hand, catatonia can entail life-threatening consequences. Acting on the principle of beneficence is thus paramount, as the patient's catatonia leaves her without the ability to act with autonomy as a participant in her medical care or to be discharged to a less restrictive setting. The court decides whether treatment over objection with ECT is the appropriate next step. For reasons outlined above, when ECT may be an ameliorative intervention, seeking consent from the court for treatment over objection is an appropriate clinical intervention when no other less restrictive option remains available.

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