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Curious Cases—The Curious Case of a Catatonic Patient

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Introduction

Catatonia is a syndrome characterized by the coexistence of psychiatric and motor symptoms.[1] It is associated with a wide range of psychiatric, medical, neurological, and drug-induced disorders.[2] The concept of catatonia was first described by the German psychiatrist Kahlbaum in 1874.[3] It is more frequently found among patients diagnosed with mania, depression, and neurotoxic syndromes than among those with schizophrenia. Yet, it is mainly classified as a form of schizophrenia.[4] The exact cause of catatonia has not been elucidated.

The syndrome of catatonia is defined by the objective presence of motor signs, over 40 of which have been described. These catatonic signs are listed in table 1. There is no agreed threshold for the number or duration of symptoms that should be present to justify a diagnosis of catatonia. Research has suffered from this, and studies can rarely be compared with confidence.[7]

There are consistent clinical reports that benzodiazepines are effective in acute catatonia syndromes, particularly stuporous conditions, but no placebo-controlled randomized studies have been published.[8,9] However, benzodiazepines are the drugs of choice for catatonia.[10] In most cases, lorazepam is administered parenterally or orally beginning with 3 mg/d and increasing rapidly to effective resolution. Dosages of 20–30 mg/d are occasionally necessary.[5] Patients who are unresponsive or insufficiently responsive to benzodiazepines need electroconvulsive therapy (ECT).[5,10]

Case Report

Patient A is a 28-year-old male of Mediterranean origin diagnosed with paranoid schizophrenia at the age of 23. He was hospitalized several times due to psychotic episodes characterized by religious delusions and auditory and visual hallucinations. He is living in an assisted living facility, where the medication is offered to the residents, but where they have to take it by themselves. He uses cannabis daily and does not use any other substances. Drug history mentions the use of risperidone and flupentixol decanoate, the latter since 2008 up to the present. At the end of 2008, he developed a progressive condition in which he showed less mimicry, staring, negativism, mutism, and immobility. There were no signs of autonomic dysregulation, such as increased body temperature or unstable blood pressure.

Because catatonia was assumed in April 2009, he was orally treated with lorazepam, starting at 2 mg a day. The lorazepam dose was increased based on the clinical state until 40 mg a day without any subjective or objective effects. He was admitted to the psychiatric ward to receive parenterally administered lorazepam up to 60 mg daily. After 2 days, there still was no measurable effect nor was there any effect on his consciousness. We resumed oral treatment with lorazepam 40 mgs daily and patient agreed to undergo ECT. During the lorazepam and ECT treatment, the patient continued to receive 30 mg of flupentixol decanoate every 2 weeks. After 3 ECT sessions (Mecta 5000, bilateral, 1 ms, 40 hz 2 s, 128 mC, 800 mA, [a relatively low, common, dosage]), the catatonic signs receded rapidly and patient refused to take the lorazepam, because "he was cured." He soon afterward developed an acute catatonic state, in which he was found completely immobile next to his bed. He received lorazepam immediately and ECT the following days. After 2 more ECT sessions, the catatonic signs receded again. During the weeks afterward, patient received 40 mgs lorazepam daily, which was reduced and finally stopped on his demand.

A few months afterward, patient presented to the acute psychiatric service with signs of acute dystonia (cervical dystonia and dysphagia). He was treated with biperiden 2 mg and the dystonia almost immediately disappeared. Flupentixol decanoate dosage was lowered to 20 mgs every 2 weeks. Patient denied the use of any drugs except

Table 1. Principal Features of Catatonia[5,6]

Clinical Feature	Description
Stupor	Altered arousal during which the patient fails to respond directly to queries (similar in presentation to the effects of dissociative anesthesia); when severe, the patient is mute and immobile and does not withdraw from painful stimuli.
Posturing (catalepsy)	Maintaining postures for long periods. Includes facial postures, such as grimacing or Schnauzkrampf (lips in an exaggerated pucker). Body postures, such as psychological pillow (patient lying in bed with his or her head elevated as if on a pillow), lying in a jackknifed position, sitting with upper and lower portions of the body twisted at right angles, holding arms above the head or raised in prayer-like manner, and holding fingers and hands in odd positions; prolonged mundane positions are common examples.
Flexibilitas cerea	The patient's initial resistance to an induced movement before gradually allowing himself or herself to be postured, similar to bending a candle.
Mutism	Verbal unresponsiveness, not always complete nor always associated with immobility.
Staring	Fixed gaze.
Negativism	The refusal of orders without any specific motive.
Autonomic instability	Abnormalities in body temperature, pulse, blood pressure, respiration rate, and sweating.
Echophenomena	Includes echolalia, in which the patient repeats the examiner's utterances, and echopraxia, in which the patient spontaneously copies the examiner's movements or is unable to refrain from copying the examiner's test movements, despite instruction to the contrary.
Stereotypy	Non–goal-directed, repetitive motor behavior. The repetition of phrases and sentences in an automatic fashion, similar to a scratched record, termed "verbigeration," is a verbal stereotypy. The neurological term for similar speech is "palilalia," during which the patient repeats the sentence just uttered, usually with increasing speed.
Mannerisms	Odd, purposeful movements, such as holding hands as if they were handguns, saluting passersby, or exaggerations or stilted caricatures of mundane movements; odd speech cadences and feigned accents are other examples.
Automatic obedience	Despite instructions to the contrary, the patient permits the examiner's light pressure to move his or her limbs into a new position (posture), which may then be maintained by the patient despite instructions to the contrary.
Motoric opposition (Gegenhalten)	Resistance to the examiner's manipulations, whether light or vigorous, with strength equal to that applied, as if bound to the stimulus of the examiner's actions.
Motoric cooperation (Mitmachen)	Exaggerated cooperation in the examiner's manipulations, even when asked not to do so. Needs to be repeatable.
Ambitendency	The patient appears "stuck" in an indecisive, hesitant movement, resulting from the examiner verbally contradicting his or her own strong nonverbal signal, such as offering his or her hand as if to shake hands while stating, "Don't shake my hand. I don't want you to shake it."

cannabis and urine examination confirmed this. After this episode, patient experienced several other episodes of dystonia, each time successfully treated with biperiden 2 mgs.

Considerations

This case has many remarkable features. To begin with, the simple fact of a slowly progressive, during multiple months, catatonic state emerging elicited our curiosity. We could not relate it to a mood disorder nor to excessive cannabis use. Then again, the administration of doses of lorazepam up to 60 mg per day without any effects whatsoever seems remarkable, especially in the case of a young man not habituated to benzodiazepines. Of interest to those practicing ECT is the remarkable fact that the quality of the ECT did not suffer under the administration of high doses of benzodiazepines. We used the dosage titration method to determine the energy level needed for the ECT. We chose to temporarily halt the action of the lorazepam with the administration of 0.5 mg of flumazenil i.v. immediately prior to the ECT and achieved a therapeutically sufficient convulsion at a relatively low energy level. After 2 ECT sessions in this manner, we chose to try a treatment session without the use of flumazenil. This had no influence on the energy necessary for the ECT; on the contrary, we obtained a convulsion of the same length and electroencephalogram waveform as we did using the flumazenil, at precisely the same energy level. After the fifth treatment session, the patient did not return for further treatment sessions, in spite of his incomplete remission and in spite of his having been warned of the possibility of relapse. He was observed to be in worse condition in his home, but he himself seemed to be less distressed by his condition than his caregivers did, in spite of the many observations that catatonia is usually accompanied by anxiety. Because of the outpatient situation, there were limitations according to physical and blood examinations and the medication intake during the (acute) catatonic state of our patient. We have had our doubts of his acceptance of the benzodiazepines, but during his stay on the ward, the administration has been closely supervised

by trained psychiatric nursing staff. Unfortunately, we did not determine a plasma level of benzodiazepines. Other laboratory results were unremarkable.

Questions

Should we consider other diagnoses than catatonia, and, if so, which? Perhaps a syndrome caused by cannabis consumption?

Does such a diagnosis explain the progression, over months, of the ''catatonic-like'' state?

How is the absence of an effect on the necessary ECT energy level by benzodiazepines to be explained?

How should we interpret the absence of distress?

Have we used the correct treatments or should we have had other considerations?

Submissions should be sent to the email address as listed in the author information. Any outcome will subsequently be published in this journal.

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