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CASE STUDY Open Journal of Psychiatry & Allied Sciences

Catatonia in Japanese encephalitis: an unusual presentation and treatment challenge

Abstract

Background: Catatonia is a syndrome of multiple psychomotor disturbances, occurring in the background of numerous disorders, among which neurological disorders are quite frequent. However, long-standing catatonic symptoms, under such conditions, can pose diagnostic and management dilemma. **Case description:** Here, we describe a young female patient who developed fever, headache, and altered sensorium, who was diagnosed with Japanese encephalitis (JE). During the illness course, she developed catatonic symptoms, which persisted even after other symptoms had settled. The catatonic symptoms responded minimally to benzodiazepines, which are the first line agents in catatonia. She responded to low-dose of atypical antipsychotics, which are not the usual choice of treatment in organic catatonia. **Conclusion:** There is dearth of data pertaining to catatonia and more so in Indian context. This report highlights the unusual presentation of JE in the form of catatonia. Secondly, clinicians can explore the possibility of using second-generation antipsychotics for treating such condition, which does require more robust data and further research.

Keywords: Organic catatonia, benzodiazepines, antipsychotics

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INTRODUCTION

Catatonia is a complex clinical syndrome characterised by a myriad of psychomotor disturbances. It can occur in context of several disorders, including medical conditions, neurodevelopmental disorders, and psychiatric disorders.[1] Although epidemiological data about catatonia is not robust, mean prevalence is around 9.2% among a variety of psychiatric or medical conditions.[2] Another review suggests that 20% of catatonia has a general medical cause, of which central nervous system (CNS) inflammation (comprising both infective and immune causes) accounts for 29%.[3] Such neurological conditions presenting as catatonia, often are accompanied by mental state changes, thereby creating a diagnostic and management dilemma for clinicians. We present a case where the patient developed catatonia during the course of Japanese encephalitis (JE) and posed management issues.

CASE STUDY

An 18 years old female presented in emergency room with complaints of fever 15 days back which lasted for four to five days, was high grade, continuous, and associated with headache and vomiting. Headache was holocranial, moderate to severe in intensity, intermittent, and barely responding to analgesics. After initial five days, fever was of low grade and intermittent. Meanwhile, the patient's speech output reduced, became nearly mute, slow in all her activities, had staring looks, and required assistance in activities of daily living. Two days prior to consultation, the patient appeared confused and agitated. She was not identifying family members and was not aware of the surroundings, disoriented to time and place; at times, she was running around in the house and trying to run out as well. Confusion and agitation were fluctuating and her behaviour and sensorium would appear normal at times. On enquiry, patient did not have any past history suggestive of neurological or psychiatric illness.

On examination, she was afebrile. Her pulse, blood pressure, and oxygen saturation were within normal limits. On CNS examination, pupils were dilated bilaterally but reactive, deep tendon reflexes were two-plus in both upper and lower limbs, plantar reflex was flexor on both sides, neck rigidity was present, and Kernig's sign was positive. She was admitted in medicine ward and investigated. Blood investigations revealed elevated lymphocytes (70%) and reduced neutrophils (26%) with normal total blood count (9000/cu mm); serum sodium 131 mEq/l and serum potassium 2.9 mmol/l; raised aspartate aminotransferase (73 U/l), alanine aminotransferase (88 U/l), and alkaline phosphatase (282 IU/l); normal kidney function test, blood sugar levels, and serum proteins; non-reactive for human immunodeficiency virus, hepatitis B surface antigen, and hepatitis C virus. Electrocardiogram, chest x-ray, and ultrasound abdomen revealed no abnormality. Mantoux was negative. However, immunoglobulin M antibody for JE in serum was present. Blood culture and sensitivity were sterile, negative for cryptococcus. Cerebrospinal fluid (CSF) culture sensitivity was also negative, CSF glucose 66 mg/100 ml (normal), CSF protein 255 mg/dl (raised). Diagnosis of JE was established. Compute tomography (CT)

of brain was normal. Patient was treated symptomatically. Within next ten days, her fever and agitation improved; appetite improved minimally. But she maintained posture for long duration (minutes to hours), had slowness in movements and speech, and responded occasionally to verbal stimulus. As these symptoms persisted, psychiatry referral was sought. The patient was diagnosed to have catatonia. Score on the Busch Francis Catatonia Rating Scale (BFCRS) was 14.[4] Lorazepam 4 mg in divided doses was started and subsequently, in view of minimal change was increased to 8 mg. However, she began to remain sedated most of the time, and thus lorazepam was tapered and stopped. Option of electroconvulsive therapy (ECT) was denied by the patient's family members. A trial of antipsychotic was then considered and quetiapine 50 mg was introduced in divided doses. Patient started showing improvement in three to four days, and relative got her discharged at request and followed-up in psychiatry outpatient services after seven days. At this time, patient had shown improvement in slowness, was walking normally, had better conversation, speech was comprehensible and relevant, and she was oriented and attentive. According to the brother (who had been accompanying her till now), she was maintaining well at home since last two days, had normal sleep and appetite, and was taking self-care and doing minimal household chores on being asked. Repeat BFCRS score was four. The patient was asked to continue the same dose for seven days and review. In next two follow-ups, there was gradual improvement, and quetiapine was tapered and stopped within next three weeks. She was asked to review after 15 days or in case of resurgence of the symptoms. She maintained well at the end of 15 days and did not follow-up thereafter.

DISCUSSION

Aetiology of catatonia can be structural or functional disturbance in CNS as well as systemic diseases causing neurological symptoms. Earlier research has shown that neurological disorders are commonly found as underlying pathology in these cases.[5-7]

Neuropsychiatric manifestations of encephalitis are although well-known, but still baffling. Infections cause catatonic symptoms probably due to direct neurotoxic effect, psychological reaction to infection, immune-mediated, or through an acute-phase response.[3] This case firstly highlights a rare presentation of JE, i.e., catatonia. Literature search reveals only one earlier report of catatonia with JE.[8]

Another important aspect here is related to patient management. Supportive treatment is the mainstay of JE management.[9] In case of index patient, in spite of improvement in other symptoms, e.g., fever, appetite, sensorium, headache, there was persistence of catatonic symptoms. Recent reviews mention benzodiazepines as first choice agents in catatonia and role of ECT where response to benzodiazepines is poor or rapid response is required due to life-threatening conditions. Neuroleptics are generally considered ineffective and even have possible risk of neuroleptic malignant syndrome; however, role of second generation antipsychotics (SGA) is ambiguous, and is mostly restricted to catatonic symptoms occurring in the background of schizophrenia.[10,11] In our patient, after the failed trial of lorazepam and inability to give ECT, the patient started showing good response to quetiapine (a SGA). It has been postulated that SGAs have weak gamma-aminobutyric acid (GABA)-agonist activity and 5-hydroxy tryptamine (5HT)-2 antagonism that could stimulate dopamine release in the prefrontal cortex and thus alleviate catatonic symptoms,[10] and might serve as an alternative management option. However, inherent to a case study, our propositions also are subjected to further research involving follow-up studies.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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