Original Article

Catatonia in Children Following Systemic Illness

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ABSTRACT

Background: The term catatonia was first introduced in 1874 and several etiologies, both organic and psychiatric have been attributed to the clinical phenotype of catatonia. The interesting feature is their response to lorazepam irrespective of their etiology. **Patients and Methods:** Four patients admitted with verbal and motor unresponsiveness following febrile illness were evaluated for infective and metabolic causes. Those who qualified for catatonia as per Diagnostic and Statistical Manual of Mental Disorders-Fifth Edition criteria and Bush-Francis Catatonia Screening Instrument screening tool and rating scale were evaluated in detail and reported. Observations: Catatonia occurs in clusters, females are more affected than males. Electroencephalogram can be abnormal based on the precipitating symptom. Minor changes in serum total iron and transferrin saturation and nonspecific elevation of viral antibody titers are seen in some patients. Lorazepam challenge always gives the diagnosis. **Result:** All patients where females and had preceeding systemic or CNS infection. Three out of the Four patients where independent at the end of one month. **Conclusion:** Catatonia should be considered as a differential diagnosis in all children with verbal and motor unresponsiveness, which have no other explanation. Early initiation of treatment is very rewarding at least during short term follow-up.

Key words: *Lorazepam challenge, organic background, pediatric catatonia*

INTRODUCTION

Catatonia as a syndrome was described first in the year 1874 by the German Psychiatrist Karl Karrl Kahlbaum as "tension psychiatry" of varied etiology in 26 cases.[1] Prevalence is estimated to be between 6% and 38%.[2] Cardinal features noticed are not communicating or repeating what others speak or gesturing what others gesture, extreme negativism or automatic obedience without interpretation of the command, which is supposed to be due to inability to disobey commands. They may demonstrate waxy moldability of their body parts by others. They may maintain peculiar postures for prolonged periods of time as well as have features of purposeless stereotypies and mannerisms. Catatonia can complicate mental illness such as schizophrenia, bipolar disorders, depression, and posttraumatic syndromes. Medical causes known to be associated are autoimmune disorders, paraneoplastic syndromes, seizures, encephalitis, metabolic disorders, use of steroids, neuroleptics, phencyclidine, inhaled abuse substances and abrupt withdrawal of benzodiazepines.[3] Diagnostic and Statistical Manual of Mental Disorders-Fifth Edition (DSM-V) separates catatonia from Schizophrenia resulting in diagnostic and therapeutic changes in the approach.[4] Catatonia is categorized as follows:

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1. Catatonia associated with general medical problems (293.89)
2. Catatonia as a subtype of schizophrenia (295.20),
3. Catatonia as an episode specifier for depressive and bipolar disorders (without any code),
4. Neuroleptic malignant syndrome considered as malignant catatonia.[5]

Irrespective of the context, it is a very debilitating disease in view of the difficulty in communicating verbally, motor dysregulation clubbed with pronounced distress. When confounded in the background of an organic neurologic illness, it poses great problem in getting an insight into the diagnosis. Most often it is mistaken as sequelae of the primary insult resulting in under diagnosis more so with reference to children. Targeted diagnostic approach is indicated when there is the slightest suspicion as treatment options are different and outcome is good if treated early at least in a small percentage of the cases.

Criteria for catatonia in DSM-V includes the presence of three or more of the following:

1. Catalepsy (i.e., passive induction of a posture held against gravity),
2. Waxy flexibility (i.e., slight and even resistance to positioning by examiner),
3. Stupor (no psychomotor activity; not actively relating to environment),
4. Agitation, not influenced by external stimuli,
5. Mutism (i.e., no, or very little, verbal response [Note: Not applicable if there is an established aphasia]),
6. Negativism (i.e., opposing or not responding to instructions or external stimuli),
7. Posturing (i.e., spontaneous and active maintenance of a posture against gravity),
8. Mannerisms (i.e., odd caricature of normal actions)
9. Stereotypies (i.e., repetitive, abnormally frequent, nongoal directed movements),
10. Grimacing,
11. Echolalia (i.e., mimicking another's speech),
12. Echopraxia (i.e., mimicking another's movements).[6]

Catatonia is a treatable disorder which carries 60 fold increased risk of premature death compared to general population if not treated properly.[7] There is very scanty literature in catatonia in children and it is said that children present with motor and verbal slowness, difficulty initiating and completing action, increased dependence on cues, passivity, parkinsonian features, reversal of Circadian rhythm, agitation and ritualistic behavior more than adults.[8] In this study we report four cases of pediatric catatonia seen by us in the last 1-year which posed great diagnostic challenge before the correct diagnosis was made.

PATIENTS AND METHODS

Children admitted to the pediatric neurology ward under our team in the last 1-year with features of psychomotor dysfunction following systemic illness and referred to neurology for diagnosis and later turned out to be catatonia are discussed. Inclusion criteria-patients satisfying DSM-V criteria for catatonia were included in the study. They were screened and severity was graded using Bush-Francis Catatonia Screening Instrument (BFCSI) [Table 1]. Case detection was done by the presence of the following signs: Excitement, immobility and stupor, mutism, staring, posturing, grimacing, echopraxia, stereotypy, mannerisms, verbigeration, rigidity, negativism, waxy flexibility and withdrawal. Their severity was rated for items 1-23 using a 0-3 point scale for each parameter. Once the suspicion was raised all the patients were given slow intravenous (IV) lorazepam 2 mg over 2 min and relief of the catatonic symptoms were looked for. Patients were investigated for central nervous system infections, acute and chronic, hepatic, renal and hematological assessment, electroencephalography (EEG) and magnetic resonance imaging. They all underwent ultrasound abdomen and chest X-ray for any detectable mass lesions. Paraneoplastic antibody screening, autoantibody screening, iron and ferritin estimation could not be done in all cases due to financial constraints. Rheumatoid factor, antinuclear antibodies, thyroid functions, HIV/venereal disease research laboratory (VDRL) screening was done in all the cases.

Case 1

A 14-year-old girl who had features of viral meningoencephalitis in the form of fever and convulsions which was diagnosed as herpes encephalitis and received the full course of antiviral agents and anticonvulsants. During convalescence after a brief period of recovery from the illness, which lasted for about a week, patient became noncommunicative, irritable in between, confused, agitated and stiff. Child refused oral feeds and therefore referred to our institution. She was investigated for persistent infections and metabolic

Table 1: Screening and severity grading scores using BFCSI

Patients	Screening	Severity score
Case 1	11/14 (no excitement, echopraxia or verbigeration)	33/69
Case 2	10/14 (no excitement, echopraxia, waxy flexibility or verbigeration)	30/69
Case 3	13/14 (no waxy flexibility)	39/69
Case 4	14/14 (all features present)	63/69 (no mitgegen/gegenhalten and combativness)

BFCSI – Bush-Francis Catatonia Screening Instrument

problems. All investigations turned out to be negative. In view of the clinical suspicion of postinfective catatonia, patient was given lorazepam 2 mg IV slowly over 2 min. After about 30 min patient became responsive with few words and moved her limbs. This response to lorazepam and absence of evidence for any other illness, possibility of postencephalitic catatonia was considered. She was maintained on 1 mg of lorazepam IV for 3 days, followed by oral lorazepam 2 mg 3 times daily. Slowly, patient improved and at the time of discharge (15 days after admission) patient was taking oral feeds, ambulant with support and at 1-month follow-up child was independent for activities of daily living.

Case 2

A 6-year-old child who was admitted with fevers with rashes of 3 days duration and claimed to have been punished for poor obedience at school following which child went into withdrawal state with occasional eye blinking, not communicating, paucity of movement in all four limbs, occasional involuntary tongue protrusion.

All her investigations were negative except for low total iron and transferrin saturation values [Table 2].

She showed very good response to IV lorazepam, which was later followed-up with nutritional care and lorazepam 2 mg 4 times a day IV and later shifted over to oral. She was discharged after 2 weeks of admission at request. At the time of discharge child was walking without support, was communicating and independently feeding.

Case 3

A 16-year-old female was admitted with history of complex seizures with fever and altered sensorium, which persisted for more than 3 weeks and therefore referred to our center. At the time of examination, patient was not opening eyes and was resisting attempt to open. She had intermittent grimacing and abnormal posturing of the limbs. She was not responsive to verbal questions and was mute. She had psychomotor withdrawal clubbed with infrequent periods of restlessness. She had involuntary passing of urine. She was occasionally asking mother whether her experiences are real or false and there were occasional features of lack of inhibition. Therefore, possibility of limbic encephalitis, infective versus noninfective was considered and her investigations were noncontributory. Therefore, a trial with IV lorazepam was given to which she showed significant response. She was continued with IV lorazepam for 3 days followed by oral lorazepam.

Table 2: Heralding illness

Patients	Heralding illness	Initial diagnosis
Case 1	Fever and seizures	Viral encephalitis
Case 2	Fever and drowsiness	Autoimmune encephalitis
Case 3	Fever with seizures	Delirium
Case 4	Fever with drowsiness	Systemic infection

Case 4

A 12-year-old girl born to nonconsanguineous parents had been symptomatic for the last 6 months. 6 months ago she had a febrile illness, which was treated as viral meningitis and following recovery from the same to almost independent activities of daily living, child started showing signs of deterioration. She was not communicating and not moving. Parents could understand her toilet needs through some gesturing. She was drooling and had a constant silly smile on her face. Occasionally during sleep she was moving her limbs. When made to sit, she was bent on her trunk. She had episodes of protrusion of her tongue and imitating gestures of others.

Her investigations for infections and autoimmune problems were noncontributory. She was given a trial with 2 mg IV lorazepam following which she became extremely hyper dynamic, was tossing in the bed violently, occasionally trying to get up, and making frightening gestures with protrusion of the tongue, which stopped after some time. She also had autonomic features in the form of shiny skin, which was severely blanching on mild pressure in addition to coldness of the limbs. Patient showed fluctuating response to treatment and at the time of discharge she was occasionally vocalizing, taking oral feeds, showing bizarre postures, moving limbs in sleep, gesturing to consultant in the form of waving but she was going back to episodes of motor and verbal inactivity. Patient was discharged with oral lorazepam 2 mg 4 times daily, oral iron and Syndopa [Figures 1 and 2, Video 1].

Observations

All the four children in this group were females. Their age varied from 6 to 16 years. Except one child all the three children belonged to higher socioeconomic status and all four of them had fever preceding the illness with an average gap of 2 weeks to 2 months from fever to onset of catatonic symptoms. Two patients had seizures. All of them satisfied DSM-V criteria for the diagnosis of catatonia and their scores as per BFCSI was 10-14 in screening and the rating score varied from 33 to 63. The highest score was seen in the patient with longest period of symptoms which was 7 months. Herpes simplex virus titers in cerebrospinal fluid (CSF) were 1:125 in two cases which is not significant. All other CSF parameters were normal. EEG showed abnormalities at the onset in three of our patients. Total iron and transferrin

Table 3: Demographic details

Patients	Age	Gender	Socioeconomic status	Religion	Place	Time of admission	Onset of illness
Case 1	14	Female	APL	Muslim	Mysore	March, 2014	March, 2014
Case 2	6	Female	APL	Hindu	Karnataka	January, 2014	January, 2014
Case 3	16	Female	BPL	Hindu	Karnataka	March, 2014	March, 2014
Case 4	12	Female	APL	Hindu	Orissa	January, 2015	June, 2014

BPL – Below poverty line; APL – Above poverty line

Table 4: CSF findings

Patients	CSF protein (mg/dl)	CSF sugar (mg/dl)	Chloride	Cells	Anti-HSV	Anti-JE
Case 1	36	77	126	Nil	1:125 (not significant)	—
Case 2	20	54	123	Nil	Negative	Negative
Case 3	21	72	140	Nil	1:125 (not significant)	—
Case 4	22	40	120	6	Negative	—

CSF – Cerebrospinal fluid; HSV – Herpes simplex virus; JE – Japanese encephalitis

Figure 1: Prelorazepam

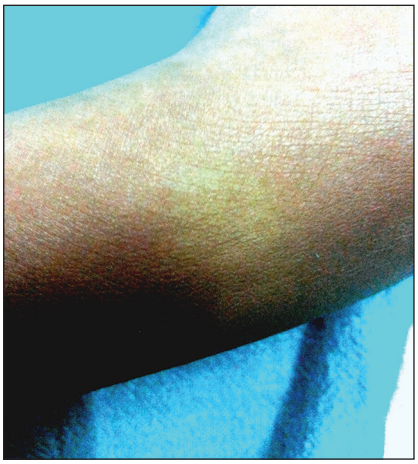

Figure 2: Autonomic changes over the skin

saturation was low in one patient. HIV, VDRL and tuberculosis-polymerase chain reaction negative in all patients.

Magnetic resonance imaging was normal in all the cases. Neuropsychological assessment could not be done in any of the patients at the time of admission and three of the patients were active and responding at the time of discharge. One patient was not co-operative for assessment. detailed neuropsychological workup was not done in any of the patients as they were children [Tables 1-8].

DISCUSSION

Catatonia in children is a relatively less known condition. It may be due to relatively less insight into the same. Most often, they have an organic cause. Females are more affected than males. They often occur in the background of minor and major systemic illness and therefore get masked under the title of some organic disorder. They often start acutely and go through a prolonged phase of unresponsiveness. Whether organic catatonia and purely psychogenic catatonia, catatonia in adults and catatonia in children differ pathophysiologically and in long term prognosis is not well known. However, benzodiazepines, specially lorazepam and antiparkinsonian drugs are very useful in the management of these patients irrespective of the underlying pathology. Electroconvulsive therapy (ECT) is also useful in resistant cases.[9] Clinical features of psychological verbal and motor withdrawal unsupported by laboratory data and lack of response to conventional methods of treatment in those circumstances should raise the possibility of catatonia. Shorter the duration of illness, better is the outcome as evidenced in our own observations.

CONCLUSION

Catatonias are not uncommon in children. Usually it occurs in the setting of systemic disease. Nonspecific viral antibodies may be found in the CSF. Alteration in iron binding capacity and ferritin levels can be seen. EEG might show abnormality based on the heralding organic disease. Females are more affected than males and probably occur in clusters suggesting a common underlying etiology in the environment like some unknown neurotropic viruses. Prognosis is good if diagnosed early. Irrespective of the cause, patient show good response to lorazepam challenge as well as follow-

Table 5: General laboratory data

Cases	Total iron (37-170 mcg/dl)	Total iron binding capacity (265-497 mcg/dl)	Unbound iron binding capacity (193-350 mcg/dl)	Serum transferrin (175-320 mg/dl))	Transferrin saturation (20-55%)	Soluble transferrin receptor (0.76-1.76 mg/L)	Serum ferritin (13-150 mcg/L)	Serum TPO-antibody (<34)	Serum VGKC antibody	Serum NMDA antibody	Serum ammonia	Serum lactate
Case 1	—	—	—	—	—	—	29.4	7.7	—	—	—	—
Case 2	21	335	314	234.5	6.3	1.1	44.80	15.7	—	—	—	—
Case 3	—	—	—	—	—	—	—	6.7			14	15.7
Case 4	—	—	—	—	—	—	—	—	—	—	55	28.6

TPO – Thyroid peroxidase; VGKC – Voltage-gated potassium channel; NMDA – N-methyl-D-aspartate receptor

Table 6: EEG findings

Cases	EEG findings
Case 1	Diffuse slowing in the delta range in left and normal on the right side, repeat EEG normalized
Case 2	Background slowing and FIRDA
Case 3	Diffuse slowing in the theta range with FIRDA
Case 4	Normal

EEG – Electroencephalogram; FIRDA – Frontal intermittent rhythmic delta activity

Table 7: MRI findings

Cases	MRI findings
Case 1	Normal
Case 2	Not done
Case 3	Normal
Case 4	Normal

MRI – Magnetic resonance imaging

Table 8: Neuropsychology assessment

Cases	Status at discharge
Case 1	Active and responding
Case 2	Active and responding
Case 3	Active and responding
Case 4	Not amenable for testing

up treatment with lorazepam. None of our patients needed ECT and 75% of our patients became completely normal at 1-month of follow-up. The only patient who had a partial response had duration of illness more than 6 months. Hence it is important to be conscious about the possibility of catatonia in unresponsive children following minor or major systemic disease longer follow-up and larger number of patients needs to be studied to know the long-term outcome of these patients.

Declaration of Patient Consent

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

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