CASE REPORT

Catatonia in Young Male with Cerebral Palsy and Intellectual Disability: A Case Report

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Abstract

Aim: To consider the possibility of an organic cause vs mental health-related cause in a young patient of cerebral palsy presenting with catatonic features.

Background: The catatonic symptoms in a young patient with cerebral palsy and intellectual disability can either be a sequelae of the preexisting organicity or has an independent organic or psychological cause. One of the organic causes is the white matter loss in the brain parenchyma-like periventricular leukomalacia (PVL) due to perinatal hypoxic insult that leads to cerebral palsy and consequent intellectual disability.

Case description: We present the case report of a 20-year-old male who presented with acute onset of catatonic symptoms in the absence of any preexisting psychiatric disorder. His general physical examination manifested spastic diplegia, mutism, negativism, posturing, and staring gaze without any other focal neurological deficits. The routine blood investigations were within normal limits, whereas the MRI brain was suggestive of PVL with white matter loss and thinning of corpus callosum. The neurology consults suggested conservative management for the same, and the patient was managed with Lorazepam and Olanzapine, following which his catatonic symptoms started improving. During his hospital stay, intellectual quotient was assessed that revealed moderate level of intellectual disability.

Conclusion: It is difficult to absolutely ascertain whether the organicity had direct influence on the patients presenting psychiatric symptoms or not. However, due to the neuropsychological changes, one cannot exclude the possibility that the sequelae of PVL or intellectual disability might have played a role in this case.

Clinical significance: This raises considerable problems when it comes to choosing a therapeutic strategy for such a patient like whether the medical intervention alone or along with assistive therapies for the damaged white matter and intellectual disability would change the course, prognosis, and the outcome of the psychiatric symptoms or whether the psychopharmacological intervention would be sufficient for a better outcome and quality of life of the patient.

Keywords: Catatonia, Cerebral palsy, Intellectual disability, Periventricular leukomalacia, Quality of life.

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Background

Catatonia in young people is infrequent and most severe. It is a neuropsychiatric syndrome characterized by psychomotor disturbances (retarded or excited) and behavioral abnormalities. It can have multiple underlying etiologies, varying from medical conditions to mental disorders. The organic conditions include infectious diseases, neurological conditions, toxic-induced states, and genetic conditions like inborn errors of metabolism, especially in cases with early age of onset of catatonic symptoms. The onset in such a case is predominantly acute, and the clinical presentation is most frequently stuporous. Mental disorders associated with catatonia are most frequently affective disorders, psychotic disorders, drug-induced states, or neurodevelopmental disorders like autism and intellectual disability.[1]

Retarded catatonia shows paucity of movements, immobility, staring, mutism, rigidity, withdrawal, refusal to eat, posturing, grimacing, negativism, waxy flexibility, echolalia or echopraxia, stereotypy, verbigeration, and automatic obedience. Excited catatonia is characterized by severe psychomotor agitation leading to serious complications like hyperthermia, altered consciousness, and autonomic dysfunction.[2,3] The relative prevalence of catatonic signs differs among studies, but there is a general agreement that catatonia occurs in 9–17% of patients with acute psychiatric illnesses, and the retarded type of catatonia is the more frequently observed subtype.[4]

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Mental health problems occur more commonly in individuals with cerebral palsy than in the general population. The most common comorbidity being anxiety and depression.[5–7]

The available literature on the association of catatonia in patients with cerebral palsy or intellectual disability is limited and in the form of case reports. Moreover, not much evidence is present for therapeutic approaches in such cases.

The cause of catatonia can be ascertained on detailed lab workup including blood, urine, and cerebrospinal fluid (CSF) analysis along with neuroimaging like CT/MRI brain. Only if organic causes or other underlying medical conditions are ruled out, then the

possibility of a psychological cause (psychotic/affective disorder) could be considered, which is much more common in such a patient presentation.

The finding of PVL in our patients with cerebral palsy and intellectual disability raises diagnostic and therapeutic problems that are extremely significant from a clinical point of view to work out the course, prognosis, and outcome of the condition and its impact on the quality of life of the patient.[8–10]

When considering the cause of catatonia in our index case, we considered it to be the sequelae of the hypoxic injury of the brain at birth that caused cerebral palsy or that it could have been another organic cause (infectious cause/space occupying lesions) or that it could have a psychological basis (psychotic/affective).[11–14]

The aim and objective of our case report is to highlight the cross-sectional diagnostic and therapeutic challenge to establish the causal relationship (if any) of the presence of organicity in the patient of cerebral palsy and intellectual disability presenting with catatonia. It also poses significant concerns regarding the course and outcome of the psychiatric disorder and raises the relevance of therapeutic intervention (medical/surgical/psychopharmacological) when both the conditions are co-existent in a patient.

Case Description

We present the case of a 20-year-old male who was brought to us in the state of catatonic stupor. The ethics committee permission has been sought for the same.

The patient was brought by the worried mother with the complaint of altered behavior of her son. According to her, since a week, her son had been observed to develop abrupt onset of symptoms that had a continuous course. The symptoms were of mutism, vacant staring, social withdrawal, hypoactivity, negativism, refusal to eat or drink, and intermittent labile affect with crying spells or sudden bursts of irritability and increased energy. He had disturbed sleep and decreased self-hygiene.

The general physical examination in the emergency room (ER) revealed an average weighted male body mass index (BMI 22.3 kg/m^2) with spastic diplegic gait. He was conscious and awake but not amenable for detailed neurological workup initially. During examination, he was observed to be in a state of catatonic stupor with mutism, negativism, posturing, withdrawal, and refusal to eat. His Bush Francis[15] screening scale was positive, and the severity score was 25 at the time of admission. He was evaluated, and the Lorazepam challenge test was given, following which the patient showed improvement in signs and symptoms of catatonia. The blood investigations and urinalysis were normal on all parameters of laboratory workup. A 2D-MRI brain done during admission revealed the presence of white matter loss, i.e., PVL and thinning of corpus callosum. The neurology liaison was obtained, which suggested conservative management for PVL. The patient was started on mouth-dissolving tablets of Lorazepam (1 mg thrice daily) and antipsychotic therapy (Olanzapine 2.5 mg twice daily), keeping in view his catatonic symptoms. No other clinical signs of any abnormality were found. The neurological examination was also insignificant in context of focal neurological deficits other than spastic diplegia. As his catatonic symptoms started improving, his mood lability was unmasked with episodes of dysphoria. His Bush Francis severity score improved to 12 on day 7 of admission.

His serial mental status examination (MSE) revealed constricted effect, psychomotor retardation, delayed reaction time, poverty of speech, and thought content. The doses of Lorazepam and Olanzapine were titrated up till adequate response was reached. The catatonic symptoms started improving progressively during the hospital stay, with amelioration of the intermittent psychomotor agitation. His oral intake also improved. Gradually, his interactions and response time improved. As he improved further, the neuropsychological testing of the index case was done. Intelligence quotient (IQ) was assessed based on developmental screening test (DST) and Vineland Social Maturity Scale (VSMS), which came out to be 47 (moderate intellectual disability).

His birth history revealed that he was born out of a spontaneous conception, and his prenatal period was uneventful. He was born by normal vaginal delivery after prolonged labor of 14–16 hours. He had low birth weight and delayed birth cry by 5–7 minutes as recalled by his mother. He had global delay in his milestones during the early developmental period. His vaccination schedule was completed on time. He started walking with a limp around 2 years of age. Faith healing was sought but turned out to be futile. No formal treatment record was available. He was sent to school around 5 years of age, where he could not get along well with his peers, but he was made to pass the classes, and resultantly, he was dropped out of school in class 10th when he failed twice. He is, however, able to take care of his hygiene and do minor chores when instructed under family supervision.

He had no history suggestive of substance abuse, and his urine drug screen (done on the day of admission) was also negative. He had no significant past psychiatric history. Family history for psychiatric problems was also negative.

The patient is on regular follow-up with us and is maintaining well. Lorazepam was gradually tapered off, and Olanzapine is continued keeping in view his mood charting that depicts fluctuations in his affective symptoms on depressive side. The family members have been psychoeducated about the need of regular scans annually to rule out any acute brain parenchymal changes that might affect the course, prognosis, and quality of life of the patient.

Discussion

This patients clinical picture is characterized by the abrupt onset of catatonic symptoms without any evidence of significant precipitating factor (organic/psychological stressor). He had preexistent cerebral palsy (spastic diplegia type) due to perinatal hypoxic brain injury. Consequently, his delayed developmental milestones also led to intellectual disability that made him more vulnerable to perception of distress.

The patient, however, did not present with the typical characteristics that distinguish whether the catatonia has a nonorganic (psychosis or mood disorder) or an organic cause like autonomic instability in the form of abnormal vital signs, recent memory deficits, altered sensorium, or focal neurological deficits, although his MRI brain showed PVL but no acute changes were depicted.[16,17] His urine toxicology panel was negative, which ruled out any drug or substance-induced psychopathology.

Hence, it is difficult to ascertain whether we are dealing with organic catatonia due to a medical condition/mental disorder or with a simple coincidence in which the organicity (PVL) is just an incidental finding without any temporal correlation with the development of the catatonic symptoms in the patient. Although the white matter loss in the form of PVL might be due to birth hypoxic injury in our case, it might have caused catatonia in the index patient as a sequela to it. As we do not have any evidence of the same due to the absence of serial MRI scans of

the patient, we could not establish any temporal correlation in this regard. Moreover, this age group is the most common for the presentation of psychotic/affective disorder otherwise and even more importantly in patients with preexistent brain dysfunction. The association of psychiatric symptoms without any neurological manifestations, young age of onset, absence of family history for psychiatric disorders, evidence of white matter loss in brain (PVL), and thinning of corpus callosum along with changes in the neuropsychological tests are the factors that might suggest a causal relationship either as a sequelae of it or due to undiagnosed underlying mental or neurodevelopment disorder.

The available literature on catatonia in young people relates most frequently to an organic cause. The prevalence of catatonia in inpatient youths varies from 0.6 to 17%.[18–21] Organic ailments may be associated and need to be investigated though no specific recommendations and guidelines are available as of now. The role of deprivation, abuse, or trauma in the development of pediatric catatonia cannot be overlooked. The history reveals that Kahlbaum gave trauma a central role in catatonia in many young adult cases, while Kanner described children with psychogenic catalepsy. Anaclitic depression, a condition found by Spitz in deprived institutionalized children, strongly resembles stuporous catatonia, and Leonhard considered lack of communication with the mother or substitute mother as an important etiological factor for childhood catatonia. Children who experience emotional and physical trauma also develop catatonia.[22] Therefore, it becomes more important to look for the psychological cause of catatonia in young adults when the possibility of any organic pretext is ruled out. Our patient showed affective symptoms toward depression when he was followed up after the resolution of catatonic symptoms indicating the importance of follow-up of such a case for better therapeutic intervention and quality of life of the patient.

Clinical Significance and Conclusion

It is difficult to ascertain whether the organic lesion had a role to play regarding this patient's overall psychiatric picture. The white matter changes in the brain and neuropsychological testing in our case (Bush Francis and IQ assessment) might conclude the possibility of the lesion in the etiopathogenesis of the catatonic symptoms in the absence of any evidence of an underlying apparent medical/mental disorder.[23] As we do not have any prior evidence for the presence of the lesion, so the temporal relationship could not be established. It is important to consider and evaluate for psychological cause of catatonia in such young adults, especially when the organicity is predominantly ruled out.

As per the current clinical practice, there are two aspects to the therapeutic approach in catatonic disorders, i.e., controlling the symptoms and correcting the etiology. In this case of PVL, where there are no focal neurological signs and symptoms, the decision is generally conservative as was taken in our case too by the neurology team.

As the number of cases described in the literature increases, the catatonic symptoms in young patients with this type of lesion cannot be unquestionably seen as just a coincidence. A more in-depth study of these types of cases is thus required in order to make it possible to optimize the therapeutic approach in cases involving the coexistence of cerebral palsy, intellectual disability, and catatonia without evidence of preexisting psychiatric disorder. The future prospects of the case aim to provide insight to the treating psychiatric team to look for and rule out organicity in all cases of catatonia in young people and to establish the temporal relationship between the same, so that the therapeutic interventions can be modified according to the individual case.[24] If the temporal relationship/organicity could not be established, evaluation of the psychological cause becomes more important so that it can be addressed in the long-term management and outcome of such a patient.

We would like to emphasize on the fact that along with regular follow-up for psychiatric symptoms, patients should also be followed up for the change in MRI findings of the brain for sequelae of PVL or other acute changes. The clinical symptoms should be addressed timely for any exacerbations or resurgence of either psychiatric symptoms or the neurological symptoms during follow-up of such a case. Hence, we propose a consensual and multidisciplinary diagnostic and therapeutic strategy to deal with catatonia in young people with preexisting cerebral palsy and intellectual disabilities. Recent advances in catatonia in youth research have offered major improvements in understanding catatonia and in new therapeutic opportunities. The syndrome is although rare, but these advances need to be acknowledged to guide patients to centers that have developed a specific expertise in this.

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