Catatonia in a child with autistic disorder

Hasan Bozkurt, Nahit Motavalli Mukaddes

Department of Child and Adolescent Psychiatry, İstanbul University İstanbul Faculty of Medicine, İstanbul, Turkey

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Catatonia is a cluster of motor features that appears in many recognized psychiatric illnesses. It is being increasingly reported in individuals with autism, a disorder characterized by impaired reciprocal social interactions, aberrant language development and restricted behavioral repertoire. However, relatively little is known about the presentation and treatment of catatonia in children with autism. We describe herein an 11-year-old pediatric case with autism who developed catatonic symptoms and was treated effectively with lorazepam. The case reported here differs from previously reported cases in terms of age of onset and the display of all characteristics of catatonia as defined in the Diagnostic and Statistical Manual of Mental Disorders (4th ed) (DSM-IV). In addition, although it was stated that catatonia in autism is commonly associated with impaired language and social passivity, our case is an active verbal individual.

Key words: catatonia, children, autism, lorazepam.

Catatonia is a syndrome of motor dysregulation characterized by mutism, immobility, negativism, posturing, staring, rigidity, stereotypy, mannerisms, echophenomena, perseveration, repetitive speech, and imitative movements. The Diagnostic and Statistical Manual of Mental Disorders (4th ed) (DSM-IV) system classifies it as a subtype of schizophrenia or associated with mood disorders and general medical conditions¹. However, some experts in this field believe that it should be classified as individual abnormal behavior².

In recent decades, there has been an increasing interest in the coexistence of catatonia in individuals with autism spectrum disorders. Some experts discussed the common neurobiological basis for autism and catatonia. They stated that catatonia and autism may both qualify as neurobiological syndromes in their own right and that coexpression of autism and catatonia may be due to abnormalities in common neuronal circuitries. It was also pointed out that some children with autistic regression may suffer from early-onset catatonic regression, since late-onset autistic regression shares some features with catatonic regression^{3,4}. Moreover, several areas of overlap between autism and catatonia are emphasized in the literature by some authors, and there is emerging evidence that catatonia may be a common syndrome in autism⁵.

Despite the relative increase in the number of published reports on the comorbidity of autism and catatonia, there remains a lack of awareness on the comorbidity of these two important clinical entities. Here, an 11-year-old male with diagnosis of autistic disorder and coexistence of catatonia is presented.

Case Report

B.C., an 11-year-old boy, was referred to our outpatient unit by his mother due to mutism, slowness in his movements and posturing. He had toilet refusal due to his immobility and had also lost his appetite. His recent complaints had started two months before referral, and their severity had been increasing gradually. During his psychiatric evaluation, he was mute, had no interest in his surroundings, gave no response to questions, and showed posturing and waxy flexibility.

Information regarding his developmental and psychiatric history was taken from his mother.

According to his mother, he was diagnosed as autistic disorder at the age of three due to his lack of social responsiveness, language delay and repetitive behavior. There was no evidence of physical disorders, and his medical work-up was unremarkable. He was enrolled into a special educational program. He showed some improvements in terms of social responsiveness and language development. At the age of six, he was a well-adjusted child with diagnosis of autistic disorder. He was able to carry on a simple conversation spontaneously. His intelligence quotient in the Wechsler Intelligence Scale for Children-Revised test was 71. He attended a program for children with autism throughout his school years and received social skills training and language intervention.

He started mainstream school at the age of seven; however, he had some crying spells after starting school. The intensity and severity of these spells gradually worsened. He was given sertraline 25 mg/day and showed some benefit from this treatment. The medication had been ceased after nine months. However, he lost his father during that period due to a heart attack, following which, his crying spells worsened and became uncontrollable. Sertraline was re-started by his physician. There was no significant improvement with sertraline 25 mg/day. In addition, after some months, he developed temper tantrums and aggressive behavior such as kicking the doors, breaking the windows, shouting. His psychiatrist had added olanzapine 5 mg/day to the treatment, but there was no response. In addition to his aggressive behaviors, after one year, his mother noticed slowness in his movements and he also gradually lost his self-help skills in daily life. He developed food refusal, showed resistance to self-feeding and lost weight. He had a decline in his self-care and experienced both urinary and fecal incontinence. He had also refused to attend school or interact with his friends for a period of approximately one year before referral. A decrease in his interaction and communication with his peers and mother was reported, and in the last month he became progressively mute. At the time of the first psychiatric evaluation by the authors, he was totally mute and showed posturing. Waxy immobility was also observed during the examination.

The family history was positive for depression. His mother was diagnosed with depression one year before and was on citalopram 20 mg/day.

After clinical evaluation, the diagnosis of autistic disorder was confirmed and he also received the additional diagnosis of catatonia. The severity of catatonia was assessed using Bush-Francis Catatonia Rating Scale⁶, with a total score on this scale of 37.

His previous medication, olanzapine and sertraline, was ceased, and he was given lorazepam 1 mg/day to test the response. The symptoms of catatonia resolved dramatically within hours; his movements increased and he started talking to his mother and the other people around him. He began to go to the toilet and his sleeplessness during this period ameliorated. Lorazepam was therefore continued at a dose of 1 mg/day. In the second week of treatment, his total score on the Bush-Francis Catatonia Rating Scale dropped to 3. He returned to school just after lorazepam treatment.

Discussion

The case reported here differs from previously reported cases in terms of the age of onset and the display of all characteristics of catatonia as defined in DSM-IV. Previous systematic studies reported the full clinical characteristics of catatonia in individuals older than 15^{7,8}. However, the present case demonstrated all features of catatonia at the age of 11 years. Another difference between our case and the previously reported cases is that while comorbidity of catatonia and autism occurs commonly in the non-verbal and socially passive group⁷, the present case is an active verbal individual. Here, the authors will discuss the clinical presentation and course of catatonia in this case.

The first major point that needs to be clarified is the careful assessment of deterioration in adaptive functions and social-emotional and communication abilities of individual with autism. The most important factors that lead to regression in autistic individuals are additional psychiatric and medical disorders. Catatonia should be assessed in any patient with autism spectrum disorders when there is an obvious and marked deterioration in movement, pattern

of activities, self-care, and practical skills when compared with previous levels, through a comprehensive diagnostic evaluation of medical and psychiatric symptoms. Any underlying medical and neurological conditions should be treated, and culprit medications or other substances that may cause catatonia should be eliminated⁹. The present case was examined carefully, and there was no evidence suggesting the presence of any medical disorders that cause this clinical picture. Regarding the effect of the medications that he had been given, the patient was using olanzapine, an atypical antipsychotic drug. There is controversial data regarding the antipsychotics and catatonia. While some published case study reports emphasize the treatment of catatonia using antipsychotic drugs¹⁰⁻¹², some case reports define catatonia after exposure to antipsychotics 13,14 . In the present case, olanzapine may have contributed to the development of the catatonia. However, it is known that antipsychotic-induced catatonia is in its malignant form and other extrapyramidal side effects usually accompany that condition¹⁵. In our case, there was no dystonia, tremor, tardive dyskinesia, fever, or symptoms associated with other medical conditions. Therefore, it could be concluded that although we cannot totally exclude the probable role of olanzapine in the development of catatonia in this case, it was most probably not the main factor leading to the catatonia.

The catatonia in this case seems associated with depression. Although there was no detailed information about clinical symptoms of depression in this case, the presence of crying spells, irritability, aggressive behaviors, positive family history of depression, and positive response to sertraline could be suggestive of a depressive disorder. His first crying spells started after going to school and reemerged and increased after his father's death. Therefore, it could be concluded that the presence of life events such as a new environment and bereavement may have contributed to the development of depression. Interestingly, Wing and Shah⁷ highlighted the role of life events like bereavement and pressure at school as possible precipitating factors in connection with catatonia; however, they did not associate catatonia in their series with psychiatric disorders. Therefore, it could be concluded that in subjects with autistic disorder, the

presence of life events could be risk factors for either depression or catatonia¹⁶.

In the present case, catatonia can be seen as a "cry for help" since after his father's death, he was unable to express his feelings well or share his sadness verbally: thus, he developed aggressive behaviors and irritability. His aggressive behaviors had not been considered as part of his grief reaction and he had not received appropriate treatment for these behavioral problems. It seems that there is a lack of awareness among mental health professionals about life events such as bereavement having a deep negative influence on the quality of life in autistic individuals. Lack of sufficient support and appropriate treatment for treatable disorders such as grief reaction and depression may contribute to life-threatening conditions such as catatonia.

Regarding the course of catatonia and the effect of lorazepam, it was noticed that the patient had a dramatic response to this drug. Parallel with our finding, de Winter¹⁷ reported a successful treatment of catatonia using lorazepam. All these reports seem encouraging regarding the role of lorazepam in the treatment of catatonia.

In conclusion, this report aims to increase clinicians' awareness regarding the diagnosis and treatment of catatonia in individuals with autistic disorder. Due to some overlapping symptoms between the two disorders, there is a risk of diagnostic overshadowing. The possibility of catatonia should be borne in mind in individuals with autistic disorder plus depression who show a regression in their behaviors.

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