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CASE REPORTS

A Case of Recurrent Hypersomnia With Autonomic Dysfunction

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We report the case of a 50-year-old man with disabling recurrent hypersomnia with autonomic instability due to catatonia in the setting of atypical bipolar disorder. Treatment with valproic acid for bipolar disorder resulted in complete resolution of symptoms.

Keywords: autonomic dysfunction, catatonia, recurrent hypersomnia

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INTRODUCTION

Recurrent hypersomnia is a rare condition often seen in patients with Kleine-Levin syndrome, autoimmune encephalitis, menstrual-related hypersomnia, and seasonal affective disorder, as well as those with anxiety and depression.[1]

Catatonia is a neuropsychiatric syndrome. Per the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), at least three of the following criteria have to be present to diagnose catatonia: catalepsy, waxy flexibility, stupor, agitation that is not induced by external stimuli, mutism, negativism, posturing, mannerisms, stereotypies, grimacing, echolalia, or echopraxia.[2]

We report a case of a man with recurrent hypersomnia with autonomic instability due to catatonia in whom atypical bipolar disorder was later diagnosed. Catatonia can occur in patients with underlying psychiatric and general medical disorders. Common psychiatric conditions associated with catatonia include bipolar disorder, unipolar major depression, psychotic disorder, and autism spectrum disorder. Recognition of catatonia is important, as it may be caused or exacerbated by treatment of the underlying disorder.[1,3,4]

REPORT OF CASE

We evaluated a 50-year-old man with a medical history of hypertension and syncope for recurrent episodes of extreme hypersomnia. The episodes started 6 years prior, occurring every 3 to 5 days, lasting 2 to 3 days, with 16 to 18 hours of sleep each day. They would occur mostly over the weekend, and begin with extreme sleepiness when the patient returned home from work. He would go to sleep early, and would awaken hypersomnolent after 16 to 18 hours of sleep. His sleep between these episodes was reported as normal. His wife noted fainting spells at the onset of his symptoms, as well as slurred speech, slow response latency, and reduced recollection of actions during the episodes. The end of these episodes would be significant for tremulousness, sweating, and tachycardia. There were no additional neurological abnormalities during the episodes. The patient's symptoms waxed and waned during the episodes. He denied changes in his appetite or sexual disinhibition during the episodes. His general and neurological examination was unremarkable, as was his birth and developmental history. He was recently initiated on sertaline 50 mg daily for anxiety. He denied intake of alcohol or illicit substances. The patient is married, has two children, and works as a consultant for an accounting firm. His family history is significant for bipolar disorder, depression, and episodes of cyclical hypersomnia in his mother. Further questioning about his clinical presentation revealed stupor (sleep-like state), mutism (little verbal response), negativism (not following instructions), agitation and anxiety during episodes beyond sleepiness, and bizarre behavior. He denied echopraxia, echolalia, grimacing, stereotypies, mannerism, posturing, or catalepsy. However, these were not tested during the episodes. The patient denied suicidal ideation, homicidal ideation, auditory or visual hallucinations, or delusional thinking.

Results of brain magnetic resonance imaging wake and sleep electroencephalogram, electrocardiogram, Holter monitoring, transthoracic echocardiogram, and metabolic work-up were normal. Overnight polysomnography revealed severe obstructive sleep apnea with an apnea-hypopnea index of 61 events/h with sleep fragmentation and oxygen desaturations to the mid-80s. Normal sleep elements and architecture were seen during video electroencephalogram monitoring during hypersomnia events, as well as during continuous positive airway pressure titration performed in between events.

Multiple Sleep Latency Testing was not performed. He was initiated on continuous positive airway pressure and modafinil 250 mg daily, which improved sleep fragmentation, but the patient remained hypersomnolent. Formal autonomic testing was not performed, but laboratory studies were notable for low serotonin, high catecholamine, positive antinuclear antibodies, and

negative antiglutamic acid decarboxylase antibodies, antivoltage-gated potassium channel antibodies, anti-double stranded DNA, erythrocyte sedimentation rate, and C-reactive protein.

The patient's clinical presentation, family history, and laboratory results raised concern for possible catatonia secondary to atypical bipolar disorder. Bipolar disorder was diagnosed based on the patient's family history, clinical presentation, and DSM-5 criteria. A treatment trial with 1 g of valproic acid at bedtime for bipolar disorder resulted in remission of hypersomnolence episodes as well as mood stabilization for 1 year until the patient's last follow- up. His valproic acid level was 60 µg/mL. The patient was not taking any other medications. His Epworth Sleepiness Scale score during events was 18 and improved to 8 on treatment. The patient's last episode was a few days before treatment initiation, and no further episodes occurred afterward. One year later, a trial of medication cessation led to recurrence of the episodes within 2 weeks. During that time, the patient had a motor vehicle accident while driving.

DISCUSSION

We report a case of recurrent hypersomnia due to atypical bipolar disorder, successfully treated with valproic acid. Catatonia is an underdiagnosed neuropsychiatric syndrome, with a reported prevalence ranging from 7% to 17% in acute adult psychiatric inpatients.[5] A wide spectrum of signs and symptoms have been described in catatonia.

In our case, the initial concern for catatonia was raised because of the patient's fluctuating clinical course combined with autonomic instability and a family history of bipolar disorder. Further directed questioning revealed stupor, mutism, negativism, agitation, and anxiety during hypersomnia episodes. An imbalance between the monoaminergic (dopaminergic and serotoninergic) and cholinergic systems has been described in catatonia,[6] which was present in our case as well and supports the diagnosis.

Catatonia should be considered in the differential diagnosis of cases of unexplained recurrent hypersomnia. Its pathophysiology remains unknown, but treatment with anticonvulsants and electroconvulsive therapy has shown success.[1,3]

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