Periodic catatonia

Challenging diagnosis for psychiatrists

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ABSTRACT

يعد الجمود العضلي الدوري من الأنواع النادرة لمتلازمة الجمود العضلي حيث عادة ما تتكرر فترات الجمود العضلي بانتظام. وتختفي أعراض المرض نهائياً بين الفترات الدورية مما يجعل تشخيص المرض صعباً. نستعرض في هذا المقال حالة مصابة بالجمود العضلي والتي لم تظهر عليها أي من الأعراض باستثناء القلق والتململ في التحويل الأول للمستشفى. وبالرغم من ظهور أعراض المرض مثل الهيستيريا وعدم الحركة إلا أن المريض لم يُنوم في المستشفى. وبعد التحويل الثاني إلى المستشفى تم تنويم المريض بعد مرور 24 ساعة على إثر إصابته بأعراض الجمود العضلي. لقد تكررت فترات الجمود العضلي بانتظام قبل العلاج بالتخليج الكهربي وعلى ذلك فقد كان التشخيص الأولي للحالة هو الجمود العضلي الدوري. وبعد ذلك تم استنتاج إصابة الحالة بالاضطراب الوجداني ثنائي القطبين وذلك من خلال المقابلة أثناء العلاج بالتخليج الكهربي. نستنتج من هذا المقال بأن هذا الاضطراب قد يُقيم بطريقة خاطئة بسبب اختفاء الأعراض بين الفترات الدورية للجمود العضلي، كما أن العلاج قد يُؤخر.

Periodic catatonia (PC) is a rare type of catatonia syndrome, in which the catatonic phases often repeat regularly. Between the catatonic periods, the symptoms totally disappear and make the diagnosis difficult. We report a case with PC who did not show any catatonic symptoms except anxiety and restlessness in his first referral to the hospital. Although it was reported that he showed the symptoms of the disorder such as mutism, and immobility, he was not hospitalized. In his next referral (24 hours later), he was referred with catatonic symptoms and was hospitalized. The periods of catatonia were repeated regularly before electroconvulsive therapy, so the primary diagnosis was PC. During treatment with ECT, based on the semi-structured interview, a diagnosis of bipolar mood disorder was made. Due to the disappearance of symptoms between catatonic periods, this disorder may be underestimated and treatment postponed.

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The main cause of catatonia is always an underlying medical, neurological, or psychiatric illness. The catatonia syndrome has most often been divided into excited and withdrawn types. Patients with the classic withdrawn-type catatonia appear watchful, but with minimal spontaneous speech and movement, in comparison, the excited type of catatonia has excessive purposeless motor activity associated with disorganized speech, disorientation, aggression, and violence.[1] Periodic catatonia (PC) is characterized by qualitative hyperkinetic and akinetic psychomotor disturbance through psychotic episodes, and debilitating symptoms in the long term with psychomotor weakness, grimacing facial movements, and apathy.[2] Periodic catatonia usually has an acute onset, shows a bipolar and polymorphous symptomatology, and runs toward an intermittent course. After one or more acute attacks, a dynamic residual state of varying degrees of severity develops.[3] Periodic catatonia is most often described as a rare heritable subtype of catatonic schizophrenia with a deteriorating course,[4] and has been characterized by specific genetic mutations and autosomal dominant inheritance.[5] Gjessing[6] systematically studied metabolic disturbance in these patients and suggested that the behavioral fluctuation of PC was related to a cyclic

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nitrogen imbalance and thyroid dysfunction. The diagnosis of a patient who has PC is difficult, despite the taking of a complete history by the clinician. In recent decades, the number of patients with PC, without any underlying diagnosis, has decreased. Periodic catatonia is generally rare, and consequently most physicians and psychiatrists are not confronted by this disorder. Our objective in presenting this particular case of PC is to highlight its important and challenging points in the diagnosis and treatment.

Case Report. A 20-year-old man whose problems, based on the family's description, had started a week after going to military service with staring at a wall, crying, no speaking, no eating, and immobility. On his referral to a clinic, after serum infusion, he became well, he could eat, speak, and recognize his relatives. The next day his previous symptoms relapsed, and he was taken to the psychiatric hospital. However, he was not hospitalized as the on call psychiatrist did not observe any symptoms, except restlessness and agitation, and as the patient was in the military service, the psychiatrist diagnosed malingering. In his second referral to the hospital, after 24 hours, he had the aforementioned symptoms; and while there he could not move, like a statue. On mental state examination the following was observed: posturing (**Figure 1**), catalepsy and waxy flexibility (**Figure 2**), negativism, and mutism.

On physical and neurological examination, he had rigidity in the distal limbs, Babinski reflex was flexor, the deep tendon reflex was normal, and his curling maneuver was negative. He did not cooperate for ophtalmoscopy. He and his family had no history of any psychiatric disorder or drug abuse. After hospitalization, an infusion of diazepam drip 10 mg in 1000 cc normal saline was prescribed. After diazepam infusion, his catatonic symptoms subsided and he was regularly visited and monitored. The following symptoms were observed: he was perplexed, agitated, and sometimes cried. He spoke, but incoherently. His behavior was like an obsessive-compulsive disorder patient, for example repeatedly touching the faucet. An evaluation of perception and delusion was impossible and, therefore, electroconvulsive therapy (ECT) (6 sessions) was prescribed to treat all symptoms. Laboratory tests showed that complete blood count, and thyroid function tests were normal. To rule out organic causes, a CT scan and EEG were performed and the results were normal. After 3 sessions of ECT some typical symptoms of mood disorder, such as euphoria and irritability were observed. With a semi-structured interview based on DSM-IV-TR; the underlying disorder was diagnosed as bipolar mood disorder (BMD). As a result, antipsychotics (risperidone 4 mg per day) and a mood stabilizer (sodium valproate 200 mg, 3 times a day) were prescribed. After 15 days, the catatonia and all the other symptoms were in remission, and he was discharged on the above drug regimen.

Discussion. Catatonia has attracted increasing attention in relation to basic theoretical problems of psychiatry.[2] The Bush-Francis Catatonia Rating Scale (BFCRS) is used in research studies and case reports. Signs in this scale including : mutism, immobility/stupor, excitement, staring, posturing/catalepsy, grimacing, stereotypy, mannerisms, verbigeration, rigidity, negativism, waxy flexibility, echolalia, echopraxia, and withdrawal should raise a high suspicion for the presence of catatonia. Impulsivity, ambitendency, perseveration,

Figure 1 - Picture showing posturing of the patient, especially in the hands.

Figure 2 - The clinician is checking waxy flexibility and catalepsy in the patient. The patient's limbs remained in position for long periods.

combativeness, and automatic abnormalities can be found in catatonia, and are also included in the scale.[1] In this patient, the symptoms of catatonia were mutism, posturing, rigidity, negativism, and waxy flexibility.

Periodic catatonia is characterized by catatonic episodes that are occurring in a cyclic pattern with clinical features of combined stupor and excitement, and remissions to an interval state.[5] This disorder is presently viewed as a subtype of schizophrenia with an autosomal dominant pattern of transmission. Stöber et al[7] reported considerable evidence for linkage on chromosome 15q15 in PC, which is a sub-phenotype of schizophrenia. Although, this case had all the PC symptoms, there was no history of psychiatric disorders either in the patient or his family. Gjessing et al suggested that the behavioral fluctuation of periodic catatonia was related to thyroid dysfunction. In the present case, thyroid function tests were normal. Bajaj et al[8] presented symptoms of PC preceded by an increase in the amount of cannabis intake and resolution of the catatonia following abstinence from the substance. In this case, the patient did not have any history of substance abuse, even cannabis.

Current treatments for PC are similar to those for other catatonic syndromes, and include benzodiazepines and ECT.[5] Lorazepam is the most commonly used benzodiazepine in the treatment of catatonia, but other benzodiazepines such as diazepam, oxazepam, and clonazepam have also been reported to treat catatonia.[1] Presented studies and case reports suggest a high success rate in treating all types of catatonia with ECT. It has been found to be effective in catatonia refractory or partially refractory to lorazepam.[9] Other recommended treatments for PC consist of lithium as an anticycling agent, to aid in slowing or arresting the periodicity of the illness. Neuroleptics aggravate catatonic syndromes.[5] In this case, after primary supportive care with adequate fluid and nutrition supplementation, the catatonic symptoms subsided with an infusion of diazepam (10 mg stat) and ECT. After conduction of a semi-structured interview based on DSM-IV-TR; the underlying disorder was diagnosed as BMD. As a result, antipsychotic and mood stabilizers were prescribed for treatment of the BMD. The differential diagnoses could be schizophrenia. Clinical experience has shown that most catatonic patients referred to this center suffered from mood disorders. In the 6 month follow up of the presented patient, persistent psychotic symptoms were not observed, and the probable diagnosis of schizophrenia was ruled out.

In conclusion, catatonia is a disorder requiring urgent hospitalization and treatment. In PC, because of the disappearance of symptoms between periods, the underlying disorder might be underestimated and the treatment postponed. Preparation of a short video clip from the crisis time of the disorder by the family of the patient, and emphasis on taking a thorough and complete psychiatric history was helpful in reaching an exact diagnosis.

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