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Letter to the Editor

Acute psychosis secondary to atypical Bickerstaff brainstem encephalitis

Dear Editor

Psychosis is a common and functionally disruptive symptom caused by not only psychiatric disorders but also medical conditions. Psychotic symptoms with behavioral disturbance may indicate neurological disorders rather than psychiatric disorders. We report a rare case of Bickerstaff brainstem encephalitis (BBE) presented with psychosis including behavioral disturbance before neurological features emerge.

1. Case presentation

A 38-year-old man was admitted to the hospital because of acute psychosis. He exhibited emotional incontinence and personality changes such as agitated and cried incomprehensively two weeks after upper respiratory tract infection. Routine blood analyses on admission were normal. The following day, the patient described auditory hallucinations involving hearing voices conversing with one another and screamed irritably like a child. On day 3, the patient displayed hypersomnolence and cerebellar-like ataxia in the absence of external ophthalmoplegia. Mann's test was positive and standing on one foot was impossible. Babinski's sign was positive bilaterally. Magnetic resonance imaging (MRI) showed a normal brain. Cerebrospinal fluid (CSF) examination showed no abnormalities. Serum autoantibodies assays against neuronal cell surface receptors and glycosphingolipids were performed. Among those antiganglioside antibodies assay revealed that anti-GQ1b IgG antibody titer was the highest. Thus, the patient seemed to be BBE even though the absence of external ophthalmoplegia. He was transferred to the intensive neurological care unit in order to receive intravenous immunoglobulin therapy. The patient began to recover consciousness with intravenous immunoglobulin and corticosteroid therapy and finally discharged on foot 6 months later. After one year, the patient exhibited no psychotic and neurological complications.

2. Discussion

In this patient, the diverse clinical features such as agitation, auditory hallucinations, hypersomnolence, and ataxia are challenging to diagnose and treat. A high level of suspicion is needed for the diagnosis. Acute psychosis is primary if it is symptomatic of a psychiatric disorder, or secondary if caused by a specific medical condition. There are no reliable pathognomonic signs to distinguish primary or secondary psychosis. Primary psychosis is a diagnosis of exclusion and psychiatrists must rule out secondary causes. Some patients with autoimmune encephalitis present psychotic symptoms. Psychosis due to the central nervous system disorders such as autoimmune encephalitis may appear before neurological features emerge (Chatterjee et al.,2017). However, there is no clear consensus which clinical signs in patients with psychosis should prompt further investigations including measurement of autoantibodies. When the patient may exhibit cognitive changes with abnormal behaviors, secondary psychosis should be considered (Griswold et al.,2015). Moreover, disturbances of consciousness and orientation, catatonia, speech dysfunction, focal neurological signs, epileptic seizures or autonomic dysfunction are warning signs indicative of secondary psychosis (Sharma et al.,2016). Ancillary testing with MRI and CSF examinations may be helpful to diagnose secondary psychosis and appropriate autoantibodies testing can confirm specific diagnoses (Lancaster,2016).

Anti-GQ1b antibodies have been found in patients with Fisher syndrome as well as BBE (Shahrizaila and Yuki, 2013). In the 1950s, Bickerstaff and Fisher independently described cases with a unique presentation of ophthalmoplegia and ataxia. The neurological features were typically preceded by an antecedent infection. In the cases with BBE, there was associated altered consciousness and hyperreflexia in support of a central pathology whereas in Fisher syndrome, patients were areflexic in keeping with a peripheral etiology. BBE is characterized by external ophthalmoplegia, oropharyngeal palsy, and impaired level of consciousness (Ito et al.,2008). But the patient did not show ophthalmoplegia. Such cases of incomplete BBE could be defined as atypical BBE, so called 'ataxic hypersomnolence without ophthalmoplegia' (Wakerley et al.,2013). Although precise pathophysiology is unclear, BBE is associated with the presence of anti-GQ1b antibodies, indicative of autoimmune encephalitis, that is one of the most common causes of noninfectious encephalitis. Among autoimmune encephalitis patients, there are a variety of clinical manifestations including behavioral and psychiatric symptoms, autonomic disturbances, movement disorders, and seizures (Hao et al.,2017). Some patients may present psychotic symptoms prior to neurological features, resulted in first visit to a psychiatric hospital. Among BBE patients, however, psychosis is a rare condition. Only, Wada et al. described two BBE patients with delirium who fulfilled the diagnostic criteria for BBE (Wada et al.,2008). The annual onset of BBE in Japan was estimated as only 100 cases by the nationwide survey of patients (Koga et al.,2012). Although it is a very rare condition, to diagnose this disease in psychiatric settings is very important because BBE patients are ameliorated by commonly available immunotherapies, although powerful immune suppression for weeks or months may be needed in difficult cases. It is important for psychiatrists to be aware of secondary psychosis as delay in recognition can have serious consequences.

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Ethical statement

The patient agreed to this submission.

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