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Case report

Adult mild encephalitis with reversible splenial lesion and catatonia: A case report

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ABSTRACT

The relationship between corpus callosum and schizophrenia is elusive. Neuropsychiatric symptoms in Mild encephalitis with reversible splenial lesion (MERS) such as delirium, and negativism, suggest a link between corpus callosum and psychiatric disturbances.

Here in, we report catatonia as an initial symptom of MERS in a schizophrenic patient.

The aim of this study is to discuss the likely causal relationship between catatonic syndrome and MERS.

To the best of our knowledge, the catatonia was not reported before as a prodromal symptom of MERS. We therefore report this case in order to enlarge the spectrum of MERS symptoms in psychiatric patients and discuss the relationship between catatonia and splenium lesions.

1. Introduction

Mild encephalitis with reversible splenial lesion (MERS) is a clinico-radiological syndrome associating a transient mild encephalopathy and MRI findings of a reversible splenial lesion of the corpus callosum [1]. Early symptoms such as fever, cough, vomiting, and/or diarrhea are a common manifestation of mild encephalopathy. Other symptoms related to the splenial lesion are still discussed.

Within the last decades, it became clear that catatonia had to be separated from schizophrenia, which was finally accomplished in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5). In fact, it showed that we may diagnose catatonia syndrome in several diseases such as general medical conditions, major mood disorders, psychotic disorders, and as catatonia not otherwise specified. This new definition of DSM-5 leads us to correlate catatonia as one of the neuropsychiatric symptoms of MERS. Here in, we report catatonic features as initial symptoms of MERS in a schizophrenic patient.

2. Case report

A 44-year-old man with a history of schizophrenia, was admitted to our psychiatric Department after an acute catatonic syndrome, with rigidity, negativism, mutism, catalepsy, waxy flexibility and stupor, rated 25/69 according to Bush-Francis Catatonia Rating Scale. The patient was under antipsychotic treatment (risperidone) with antiparkinsonian drug (Biperiden) without treatment withdrawal. The conscientiousness was disturbed, with a score of 7 on the Glasgow scale. His physical exam found a fever of 38–39, with tachycardia. Biological workup revealed increased creatine phosphokinase (CPK) and liver enzymes blood levels with leucocytosis. No other metabolic disorders were found. There was no hyponatremia or hypoglycemia. Infectious workup including urinary cytobacteriological examination and chest x-ray were unremarkable, as well as cerebral CT (scanner). Cerebral MRI showed a round lesion in the splenium of the corpus callosum which was hyperintense on FLAIR and T2-weighted images and hypointense on T1-weighted images with no enhancement after intravenous gadolinium administration. There was

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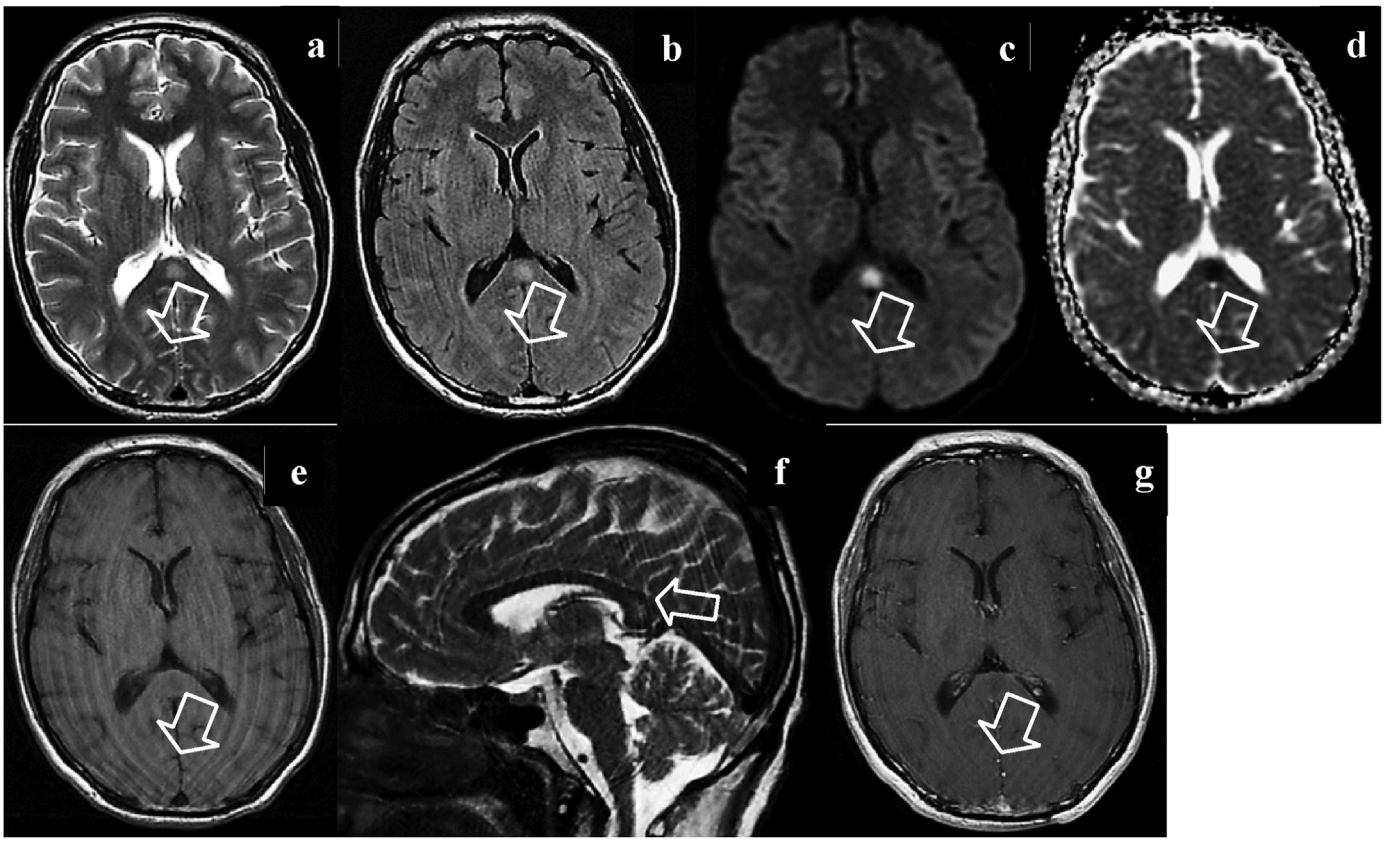

Figure 1. Brain MR images showing an oval lesion in the splenium of the corpus callosum (open arrow) appearing hyperintense on T2-weighted (a, f) and FLAIR images (b), with restricted diffusion (c, d). Axial T1 (e) displays the same lesion, with slight hypointense signal and without enhancement after intravenous gadolinium administration (g).

restricted diffusion within the lesion, suggesting the presence of a cytotoxic oedema (Figure 1). No other lesion was present on the MR exam. On day 3 of hospitalization, the patient presented consciousness impairment and thus he was transferred to the intensive care unit. He was conditioned, put on a non-invasive monitoring and was treated with parenteral hydration and nutrition, preventive proton pump inhibitor and low-molecular-weight heparin. For his psychiatric condition, he was put under diazepam 20mg/day.

The evolution under treatment was marked by the improvement of catatonic symptoms, the disappearance of fever and the recovery of awareness. The patient was discharged at day 40 with the same treatment. A follow-up MRI performed few weeks after the first one showed a complete resolution of the lesion in the splenium of the corpus callosum (Figure 2).

3. Discussion

Classical neurological and neuropsychiatric symptoms described in MERS are disturbance of consciousness, seizure, cognitive impairment, visual hallucination, ataxia, signs of interhemispheric disconnection (alien limb sign), and dysarthria [2]. Some neuropsychiatric signs were rarely reported such as negativism [3], and mania [4]. To the best of our knowledge, catatonia was not reported before as a prodromal symptom of MERS. The present case is the first case reporting this association.

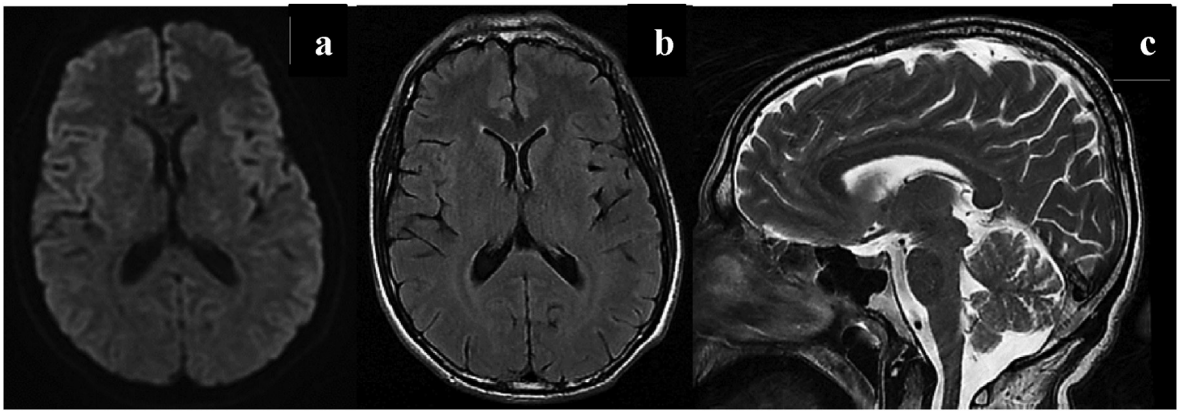

Figure 2. Complete regression of the abnormal signal in the splenium of the corpus callosum (a, b, c) on the follow-up MR exam.

This case is important in two aspects. First, MERS, a rare disease entity that still needs clarification regarding symptoms. Second, it adds to the significance in the etiopathogeny of MERS.

The corpus callosum (CC) is a brain structure in placental mammals that connects the left and right cerebral hemispheres. Containing numerous intra- and interhemispheric myelinated axonal projections, it is considered to be the largest white matter structure in the brain [5].

The study by Francis et al [6], showed that the portion of the corpus callosum that was most consistently associated with psychosis was the splenium: its volume was reduced in psychotic patients with schizophrenia, schizoaffective disorders, and bipolar disorders compared to controls, and was also reduced to an intermediate degree in nonpsychotic relatives. The localization of findings to this section of the corpus callosum is of particular interest because the splenium carries interhemispheric connections both to and from the hippocampus and adjacent medial temporal cortex. The hippocampus is perhaps the brain region most robustly implicated in structural and functional neuroimaging studies of both patients with psychotic disorders and people at high risk for psychosis [7]. Catatonia is very likely to be associated with dysconnectivity of large-scale neural networks. Dysconnectivity may be traced to abnormal white matter (WM) microstructure. In fact, Jakob et al [8] found significantly reduced fractional anisotropy in CC in catatonic compared to non-catatonic Schizophrenia spectrum disorder patients [8]. All these arguments lead us to believe that catatonia could be a symptom of splenial lesion.

MRI findings in MERS consist of transient white matter lesions that are hyperintense on Flair and T2-weighted images and isointense or slightly hypointense on T1-weghted images with restricted diffusion and no enhancement after intravenous gadolinium administration [9]. In MERS type I, there is an isolated lesion in the splenium of the corpus callosum which was the case in our patient. In MERS type II, similar lesions are present in cerebral white matter [9]. Diffusion weighted imaging allows an earlier detection of the lesions compared with Flair images and restricted diffusion within the lesions could indicate the presence of cytotoxic oedema [2]. A complete regression of lesion on follow-up MR exams is a hallmark of MERS.

The cause of MERS in our patient is unknown but many hypotheses are plausible. It could have been due to multiple vitamins deficiency [2] since the patient presented food refusal, or to a non-documented viral infection. In fact, MERS can be triggered by infections such as Influenza viruses A and B (19%): the most common pathogens, followed by the mumps virus (7%), adenovirus (6%), rotavirus (6%), Streptococcus (6%), and Escherichia coli (6%) [10,11]. It could have been caused by another aetiology such as adverse drug reaction in patients with malignant neuroleptic syndrome [1,12] since our patient presented fever with muscular and liver cytolysis. Nevertheless, in many reported cases the aetiology was not identified [13].

A broader entity is the Reversible splenial lesion syndrome (RESLES). It is a clinico-radiological syndrome with lesions that involve the CC on the MRI that have disappeared or improved significantly during follow-up [14].

In our case the diagnosis of MERS was established by the association of reversible radiological lesions with altered consciousness and fever. In the context of RESLES the aetiologies are very varied such as discontinuation of anti-epileptic drugs, collagen pathologies, metabolic disorders such as hyperglycaemia or hyponatremia and high-altitude cerebral oedema [15]. No metabolic disorder was found in our case. No antiepileptic treatment was taken. Another aspect of the etiological discussion is the use or discontinuation of antipsychotics. A possible correlation between neuroleptic malignant syndrome and RESLES has been described [16,17]. Dysnatremia has been suggested as a common cause of NMS and RESLES [18]. Excess intracranial glutamate concentration has also been suggested as a common basis for both disorders [19]. This mechanism is suggested by Sweni et al to be responsible for MRI changes and NMS symptoms such as hypernatremia, tremor and dystonia [19]. These hypotheses are interesting in understanding our case given the possible continuum between MERS and RESLES on the one hand and between NMS and catatonia on the other [20].

4. Conclusion

Despite the intimate relationship between psychiatric and somatic comorbidities, physical conditions are underestimated and go usually unrecognized in psychiatric patients. The slightest unusual symptom in the course must lead to a comprehensive biological work up and brain imaging. Catatonic syndrome could be a prodromal sign of splenial lesion but until date, the link between corpus callosum and catatonia is still mysterious. More reports and studies are required to establish this cause-effect link.

Declarations

Author contribution statement

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Declaration of interest's statement

The authors declare no conflict of interest.

Additional information

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