Dear Editor,

A 64-year-old Chinese woman presented at our hospital with obvious cognitive decline and generalized tonic–clonic seizures for the previous four months. Two months before coming to our hospital, she had begun to experience frequent episodes of faciobrachial dystonic seizure (FBDS), and her family noted further cognitive deterioration. She had previously been prescribed triple antiepileptic drugs, but they did not control her symptoms effectively.

Upon admission to our hospital, the patient could remember only her name and therefore could not complete the Mini-Mental State Examination (MMSE). Brain magnetic resonance imaging (MRI) based on T2-weighted-fluid-attenuated inversion recovery showed hippocampal hyperintensity [Figure 1a and b]. Positron emission tomography with 2-deoxy-2-[18F]fluoro-D-glucose (18F-FDG PET) integrated with computed tomography showed increased metabolism in bilateral basal ganglia [Figure 1c–e]. Laboratory analyses of blood routine, blood biochemistry, and coagulation function were unremarkable. Serum and cerebrospinal fluid were positive for antibodies against leucine-rich glioma-inactivated 1 (LGI1). Therefore, the patient was diagnosed with anti-LGI1 encephalitis, and she was started on immunosuppressive therapy of intravenous immunoglobulin and methylprednisolone.[1,2] She was discharged after two weeks with an MMSE score of 14 and without recurrent seizures or FBDS.

Anti-LGI1 autoimmune encephalitis is a rare neuroinflammatory brain condition that usually manifests as limbic encephalitis. Anti-LGI1 encephalitis is characterized by acute or subacute cognitive impairment, epilepsy, FBDS, psychiatric disturbance, and hyponatremia.[1,2] Initial diagnosis of anti-LGI1 encephalitis is based mainly on clinical manifestations, antibody testing, and neuroimaging. Cranial MRI showing abnormal signals in the temporal lobe medial or hippocampus is an important basis for diagnosing anti-LGI1 encephalitis.[1,3] According to prior studies, the FBDS usually precedes the anti-LGI1 encephalitis and is a specific clinical marker for this form of encephalitis.[4,5]

Figure 1: (a and b) Brain magnetic resonance imaging (MRI) of the patient: (a) the T2-weighted image demonstrated bilateral hippocampal hyperintensity, (b) the MRI fluid-attenuated inversion recovery image showed bilateral hippocampal hyperintensity (arrows). (c–k) Positron emission tomography based on 2-deoxy-2-[18F]fluoro-D-glucose showed a diffuse decrease in glucose metabolism (green color) in the cerebral cortex and cerebellar hemisphere, but increased metabolism (yellower/redder color) in the bilateral caudate putamen, lenticular nucleus, and dorsal thalamus. Increased glucose metabolism was also observed in the right hippocampus.
Our patient presented with typical clinical manifestations including FBDS and characteristic MRI correlates of limbic encephalitis. Furthermore, the present patient also showed glucose hypermetabolism in the hippocampal and basal ganglia, accompanied by glucose hypometabolism in cortical and cerebellar areas [Figure 1c–k]. Patients with anti-LGI1 encephalitis can show different metabolic patterns in the hippocampus and neocortex, and such patterns may correlate with neurologic disability.[6]

Most patients with anti-LGI1 encephalitis respond well to immunotherapy, which quickly relieves FBDS and mitigates most symptoms.[1] Cognitive function, however, returns slowly, and some patients experience irreversible neurological sequelae.[1] Thus, early recognition and treatment of anti-LGI1 encephalitis are critical. Since FBDS often accompanies the onset of amnesia and limbic involvement in anti-LGI1 encephalitis, detecting FBDS early may permit prompt immunotherapy to prevent progression to limbic encephalitis and subsequent cognitive impairment. However, MRI findings are typically unremarkable at the stage of FBDS. 18F-FDG PET can facilitate early diagnosis of limbic encephalitis and monitoring of treatment response, and the technique is more sensitive than conventional imaging.[6] Since relying on autoantibody testing for diagnosis may significantly delay treatment initiation, 18F-FDG PET may provide an early indication of limbic encephalitis.

The origin of FBDS has been extensively debated, with no definitive conclusions so far. Some studies hypothesize that FBDS has an epileptic origin, but other work suggests that it is more likely a form of ictal dystonia.[7] Based on PET results indicating the involvement of basal ganglia, our result seems more support that FBDS arises from network dysfunction between cortical and subcortical areas. Because FBDS is frequently misdiagnosed as psychogenic or indeterminate, PET findings of abnormal glucose metabolism in cortical and subcortical areas should lead clinicians to suspect anti-LGI1 encephalitis. Timely treatment with immunotherapies may then prevent the development of limbic encephalitis.

This case report confirms that early diagnosis of anti-LGI1 encephalitis is uneasy and critical; clinicians should grasp the characteristics of the disease to improve recognition. PET examination may help with early diagnosis and further study of the mechanisms of anti-LGI1 encephalitis and FBDS.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patients understand that her name and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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