A Case of Hyperglycemic Hemichorea and Review of the Literature

Keywords: Hemichorea; Hyperglycemia; Ballism

Abstract
Chorea is a kind of movement disorders result from many conditions. Hyperglycemic hemichorea is one of them which can be seen in patients with type 2 diabetes in long-term poor glycemic control. In this case, we reported a 58-year-old woman who presented with involuntary movements of left limbs. Her MRI can see hyper intensity in T1. After hypoglycemic therapy, her symptom of hemichorea got reverse to some extent. This case is relatively rare in clinic. We reviewed the clinical cases and discussed the possible mechanism.

Case presentation
The patient was a 58-year-old woman presented with persistent involuntary movements of the left upper and lower limbs. These movements can be disappeared during sleep (Video 1). She had a previous history of hypertension for 3 years and was known diabetic for 1 year. In August this year, she self-discontinued taking drugs. Recently, her blood sugar has risen significantly. Her head CT scan showed hyperintensity in bilateral caudate nucleus and globus pallidus, considering the possibility of metabolic lesions. She denied the history of fever, respiratory and gastrointestinal infections.

After admission into our hospital, on her physical examination, there was left-sided unilateral hemichorea. Patient remained conscious, part of the answer is correct, self-talk, lack of cooperation. Her pupils were equal and reactive to light and did not see no Kayser-Fleischer ring appreciated on naked eye examination. The muscle strength of the limbs is about grade 5, the muscle tone is still normal, the sensory and ataxia examination is uncooperative, bilateral Barthel sign (-), meningeal irritation sign (-).

Her laboratory examination showed that her blood glucose was 11.72 mmol/L (FPG), while HbA1c was 15.7%. Urine routine showed urine glucose 3+. Her blood routine, coagulation series, pre-transfusion examination, procalcitonin, immune complete set, vitamins did not find obvious abnormality. Her MRI scan showed a change that we can see high signal in bilateral basal ganglia in T1 (Figure 1, 2) and he EEG showed no obvious abnormality.

On admission, we injected insulin to control her plasma glucose. We used haloperidol to control her involuntary movements which start with a small dose (4mg/d). Since the movements were more stubborn, we gradually increase the dose later (6mg/d). Her symptoms were controlled to some extent, but her left lower limbs still showed involuntary movements. (Video 2) After continue treatment for a while, her hemichorea symptom got reverse.

Discussion
The onset of involuntary movement of one limb often occurs in diabetic patients with chronically poor glycemic control. The disease was first reported in 1960 by bedwell [1]. The published cases have shown that this disease is more common in Asian and is more frequently in elderly female [2]. Researchers think that may be related to changes in hormone levels after menopause [3]. Most cases occurred in patients with type 2 diabetes who had long-term poor glycemic control. Jeremy B Lin et al. presented a case that a 20-year-old lady presented with acute hemichorea with poorly
controlled type 1 diabetes [4]. In most cases, chorea symptoms can be significantly relieved with blood sugar correction therapy. However, some cases reported that after hyperglycemia correction, the patients showed hemichorea instead [4]. Almost all cases of hyperglycemic-hemichorea can see characteristic imaging of basal ganglia, especially in T1 can see high signal, in T2 most cases can see low signal in the basal ganglia [5]. After hypoglycemic therapy. The prognosis for dyskinesia is good, and the radiographic abnormalities are fully reversible [6].

The pathogenesis of hyperglycemic-hemichorea is remain unclear the following are several possible hypotheses. One hypothesis thought because of long-term poor blood sugar control the cerebral metabolism shifts to anaerobic pathway, this change made the brain metabolizes GABA in order to provide extra energy [3]. Due to the decrease in GABA level, thalamic disinhibition and hyperkinesis are enhanced, which makes the emergence of hemichorea [7]. Other hypothesis thought that because of hyperosmolar environment caused by hyperglycemia, edema of brain cells and demyelination of peripheral nerves cases the symptom of hemichorea [5]. Other than these, focal microhemorrhage, ischemic injury, hyper viscosity, and mineralization also play a role in the pathogenesis [8].

The managements of hyperglycemic-hemichorea include hypoglycemic therapy and controlling of chorea, haloperidol is the most commonly used and proven to be the most effective [9]. After these treatments, the majority of patients with hemi lateral chorea have a good prognosis. It has been reported that low-frequency repetitive transcranial magnetic stimulation, deep brain stimulation, and surgical techniques such as ablation are also effective, but they are rarely used in clinical practice [10,11].

Conclusion

In conclusion, hyperglycemic hemichorea is usually due to poor long-term blood glucose control in diabetic patients. At present, the mechanism of the disease still needs to be further explored. The most important imaging feature is the high signal expression in the bilateral basal ganglia of T1. Most patients can get relief after giving hypoglycemia and controlling motor symptoms. The overall prognosis was satisfactory.

Conflict of interest statement

We declare that we have no financial and personal relationships with other people or organizations that can inappropriately influence our work, there is no professional or other personal interest of any nature or kind in any product, service and/or company that could be construed as influencing the position presented in, or the review of, the manuscript entitled.

References