中文題目:以頑固性癲癇表現之胰島素瘤-病例報告

英文題目: Insulinoma presenting as medical refractory epilepsy: A case report

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**Background**

Insulinoma is a cause of recurrent hypoglycemia and is potentially curable; however, if the condition presents as neuropsychiatric symptoms often delayed the diagnosis. Here, we present a case of insulinoma that was misdiagnosed as medical refractory epilepsy.

**Case presentation**

A 49-year-old man had been well until about one and a half years before visiting our hospital on June 2021, when his family observed episodes of bizarre behaviors. He got lost while driving on his way home before lunch and recovered almost 1 h later without requiring medical care. Another episode occurred about 1 month after the first where his four limbs were stretched, and he showed consciousness disturbance for 1.5 h before lunch. Subsequently, he visited a regional public hospital, where his brain computed tomography (CT) and electroencephalogram showed no remarkable findings. A diagnosis of seizure has been diagnosed established by a neurologist, for which he was initiated on with medical (Levetiracetam - Keppra 500 mg twice a day) control about one year ago. However, the conditions are still recurrent with an increasing frequency, and he was considered to have medical refractory epilepsy. He experienced intermittent nonsensical speech, non-purposeful movements of the limbs, and various unusual behaviors. These symptoms and signs usually occurred before lunch and before dinner, persisted for 2–3 h, and resolved spontaneously. Furthermore, the patient could not remember the episodes. He experienced weakness and dizziness after these episodes. Throughout the course of the disease, he had no obvious palpitations, tremors, anxiety, sweating, or hunger sensations.

The patient visited the outpatient department of neurology of our hospital on 2021/06 based on the previous diagnosis of epilepsy. About 1 month before visiting our hospital, the patient experienced consciousness disturbance and displayed bizarre behavior again before lunch, which persisted for 3.5 h. He was sent to the emergency department of the medical center of southern Taiwan. Severe hypoglycemia (glucose level: 32 mg/dL) with

hyperinsulinemia was detected. His clinical symptoms and signs resolved after receiving a supplement of glucose-containing intravenous fluid. At our hospital, because of a history of hypoglycemia complicated with conscious change, he was referred to the division of endocrinology and metabolism. Hypoglycemia with a fasting blood glucose of 32 mg/dL was detected. Concurrently, the insulin level was still high (25.9 uIU/mL/C-peptide was 2.78 ng/mL), HbA1c was 5.0%, and he was admitted for further evaluation.

After admission, the patient had clear consciousness with a fair general appearance, the height/weight was 174 cm/77.3 kg and the body mass index was 25.5 kg/$m^2$. His vital signs were as follows: body temperature, 36.1°C; pulse rate, 71/min; respiratory rate, 16/min; blood pressure, 116/70 mmHg; and $SpO_2$, 99% under ambient air. Physical examination revealed clear breath sounds, no icteric conjunctiva, no jaundice, no palpable thyroid enlargement, no hepatomegaly, no rashes, and normal capillary refill. Neurological examination revealed intact cranial nerve, normal muscle power, normal gait, normal tactile sensation, negative Romberg test, and steady gait. The patient claimed he did not use any illicit drugs or Chinese herbs; his only ongoing medication was Keppra, and he had a family history of a cousin with an acute myocardial infarction. He had no history of trauma. Laboratory tests revealed the following: fasting blood glucose, 31 mg/dL; ACTH, 24 pg/mL; cortisol, 11.5 mcg/dL; IGF-1, 229 ng/mL; GH, 3.91 ng/mL; prolactin, 25.67 ng/mL; C-peptide, 2.83 ng/mL; insulin, 12.2 uIU/mL; and Insulin/C-peptide ratio, 0.09. Accordingly, endogenous hyperinsulinemic hypoglycemia was diagnosed. Finally, he was diagnosed with insulinoma based on laboratory findings, and a pancreatic tail mass that was revealed via CT. He underwent a distal pancreatectomy (pathology reported as a neuroendocrine tumor and positive immunostaining for insulin) with no further recurrence of his symptoms.

## Conclusion

Untreated hypoglycemia-related conscious changes may progress to life-threatening conditions. Insulinoma is a potentially curable endogenous hypoglycemia condition; however, its diagnosis is challenging because of its variable presentation, when neuropsychiatric symptoms with no plausible explanation, insulinoma should be considered.