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A Case of the Syndrome of Acute Bilateral Basal Ganglia Lesions due to Hypoglycemia in Hemodialysis Patient

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Authors' contributions

This work was carried out in collaboration between all authors. Authors SG and HSS designed the study, performed the statistical analysis, wrote the protocol, and wrote the first draft of the manuscript. Authors YNK, YSJ and HR managed the analyses of the study and the literature searches. All authors read and approved the final manuscript.

Case Study

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ABSTRACT

Patients with chronic kidney disease may have neurological complications including uremic encephalopathy, stroke, neuropathy and myopathy. Rarely, acute movement disorder associated with bilateral basal ganglia lesion is seen in patients with end stage kidney disease. The hallmarks of this condition include reversible and uniform lesions of the basal ganglia on MRI which stand for decreased signal intensity on T1-weighted images and increased signal intensity on T2-weighted images, and the clinical presentation includes acute parkinsonism and/or involuntary movements. This syndrome has been reported mainly in Asian patients, typically in the setting of long-standing diabetes. We report a case of bilateral basal ganglia lesions in a patient with chronic renal failure, poorly controlled diabetes, and incidents of severe hypoglycemia.

In our case, there was no evidence of acute metabolic disorders. Most reported patients with acute basal ganglia lesions in uremia also had diabetes and/or abnormal blood glucose levels. Our case had previously experienced occasional hypoglycemia before the onset of involuntary choreic movements. MRI of our patient showed acute bilateral basal ganglia lesion, corresponding to cytotoxic edema. This pattern was also observed

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in patients with hypoglycemic encephalopathy.

Keywords: Acute bilateral basal ganglia syndrome; chorea; diabetes; hypoglycemia; uremia.

1. INTRODUCTION

Patients with chronic renal failure may have neurological complications including uremic encephalopathy, stroke, neuropathy and myopathy [2]. Uremic encephalopathy is an especially well-known complication in chronic renal failure. Rarely, acute movement disorder associated with bilateral basal ganglia lesions is seen in patients with end stage kidney disease. The hallmarks of this pathology are reversible, uniform lesions of the basal ganglia found on MRI, and the clinical presentation includes acute parkinsonism and/or involuntary movements. This syndrome has been reported mainly in Asian patients, typically in the setting of long-standing diabetes mellitus [3-7]. Recently, a case of acute basal ganglia lesions in uremia preceded by severe hypoglycemia was reported [1] and we experienced a similar case. So, we describe a case of bilateral basal ganglia lesions in a patient with chronic kidney disease, poorly controlled diabetes, and incidents of severe hypoglycemia.

1.1 Case Report

A 42-year-old male patient was admitted to the hospital with involuntary choreic movements of the extremities that had appeared 1 week earlier. The patient had a 2-year history of chronic renal failure due to diabetic nephropathy and had received regular hemodialysis for 1.5 years. Three years prior to hospitalization the patient had been diagnosed with type 2 diabetes and treatment with insulin (NPH) 12 units and glimepiride 2mg once daily in the morning had been started at that time. He had maternal diabetic family history, and when he was diagnosed with diabetes, he had fundoscopy by an expert ophthalmologist. He had proliferative diabetic retinopathy at both eyes, already. He occasionally experienced hypoglycemia due to irregular diet and medications. One day before the onset of choreic symptoms, the patient had experienced hypoglycemia with mental confusion and sweating, but his finger capillary glycemia had not been checked. On admission neurological examination revealed fair grade motor power of both upper and lower limbs and left-sided unilateral positive Barbinski's sign with no further abnormalities. Laboratory investigations indicated that the patient's potassium level was 3.8 mmol/L (3.5-5.3 mmol/L), calcium 8.9 mg/dL (8.0-10.0 mg/dL), phosphate 2.6 mmol/L (3.0-4.5 mg/dL), bicarbonate 25.8 mmol/L (20.0-29.0 mmol/L), urea 26 mg/dL (5.0-23.0 mg/dL) and creatinine 8.6 mg/dL (0.7-1.3mg/dL). The patient's serum fasting glucose level was 101 mg/dL (70-126 mg/dL), glycated hemoglobin (Hemoglobin A_{1c}) was 5.2 % and his pH was normal. Our patient had went into cardiac arrest due to vasospasm of coronary arteries three month ago before presentation and after five-minute cardiopulmonary resuscitation, he recovered. For estimate existence of hypoxic brain damages, he had undergone MRI which revealed no definite lesions of the basal ganglia (Fig. 1A). A brain MRI was performed on admission which revealed hypointensities in both basal ganglia on T1-weighted images (Fig. 1B). There were no signs of acute hemorrhage in gradient-recalled echo (GRE) images (Fig. 1C). MRI showed hyperintensities in T2-weighted and fluid attenuated inversion recovery (FLAIR) images (Fig. 1D) that were located in the lentiform nuclei bilaterally with perifocal edema. Diffusion-weighted imaging (DWI) showed high signals in the involved brain regions (Fig. 1E) and the apparent diffusion coefficient (ADC) values of lentiform nuclei were decreased in the central part, indicating cytotoxic edema. Oral 60mg prednisolone and 300mg valproic acid daily treatment was initiated to address the

bilateral basal ganglia lesions and choreic movement, and prednisolone was tappering off slowly. Insulin injections were stopped, and the patient's blood glucose levels were well controlled. Hemodialysis was continued 4 times a week for 4 weeks and after 30 days the patient's extrapyramidal symptoms resolved, but he continued to exhibit muscle weakness and ataxic gait disturbance due to long-time bed rest. Brain MRI performed 3 months after the onset of symptoms revealed regression of the previous bilateral basal ganglia lesions (Fig. 1F).

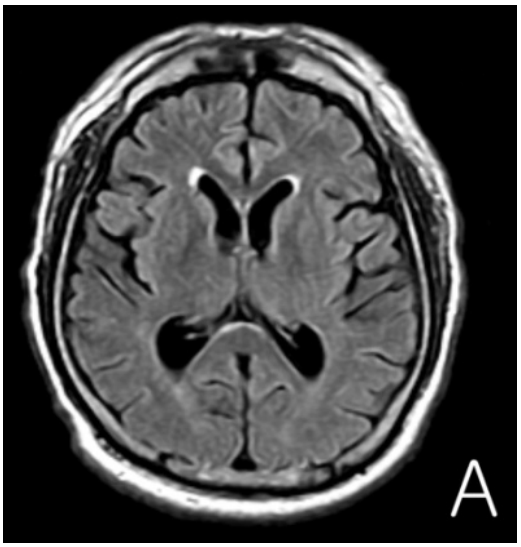

Fig. 1A

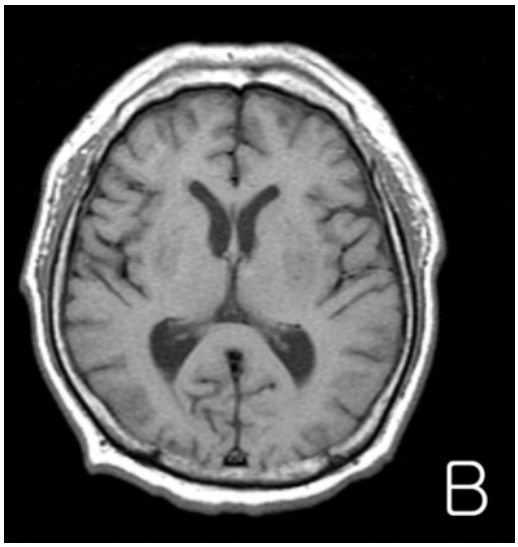

Fig. 1B

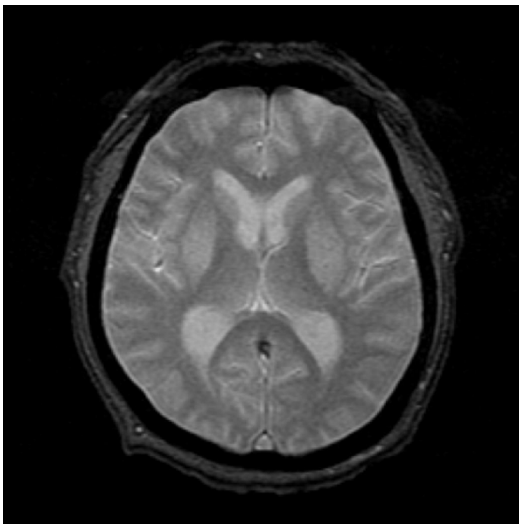

Fig. 1C

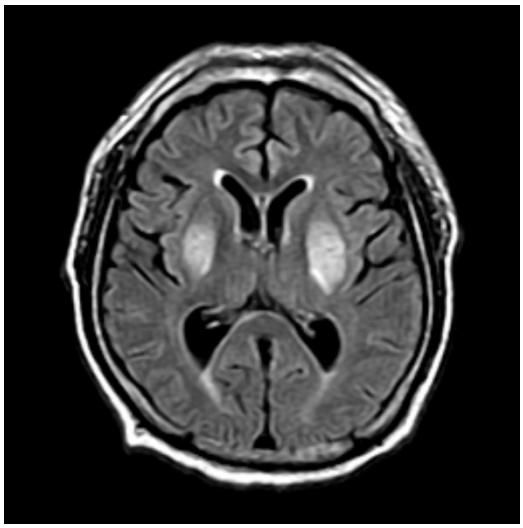

Fig. 1D

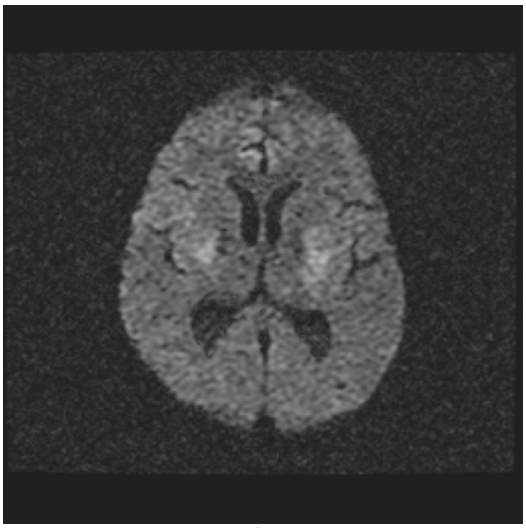

Fig. 1E

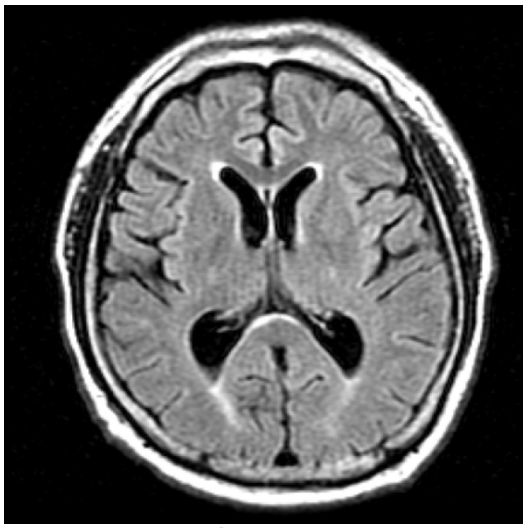

Fig. 1F

Fig. 1. A. Brain MRI before onset of symptoms. Previous MRIs showed no definite basal ganglia lesions. B-E. At the onset of symptoms. B. In the acute phase of disease, T1-weighted brain MRI revealed bilateral symmetrical hypointensities in the lentiform nuclei with surrounding edema. C. There was no evidence of brain hemorrhages in MPGR. D. FLAIR images showed hyperintensities in the same regions. E. The lesions had high signal on DWI, corresponding to cytotoxic edema. F. Three months after the onset of symptoms. Repeated MRI revealed marked regression of the bilateral basal ganglia lesions on FLAIR images

2. DISCUSSION

The basal ganglia require high energy input for motor control and may be selectively damaged by systemic processes that decrease cerebral metabolism. Causes of bilateral basal ganglia lesions include cerebral infarction, hemorrhage, vasculitis, intoxication, encephalitis, and acute metabolic disorders. Lesions induced by these causes are irreversible. The pathogenesis of bilateral basal ganglia lesion syndrome in uremia is unclear. Uremic toxins are prevalent in all uremic patients, whereas the syndrome of bilateral basal ganglia is a very rare complication of uremia. This renders it unlikely that uremic toxins per se are not the main trigger of this syndrome but it is possible that they may increase the vulnerability of basal ganglia to other deleterious factors, such as metabolic acidosis. In our case, there was no evidence of metabolic acidosis and previously experienced occasional hypoglycemia was found. Most reported patients with acute basal ganglia lesions in uremia also had diabetes and/or abnormal blood glucose levels. Recently, a case of acute basal ganglia lesions in uremia preceded by severe hypoglycemia was reported [1]. Our case had previously experienced occasional hypoglycemia (fasting glucose level 30-50 mg/dL) before the onset of involuntary choreic movements. In our case, DWI of MRI showed high signal intensity and low ADC values in the bilateral basal ganglia, corresponding to cytotoxic edema. This pattern was also observed in patients with hypoglycemic encephalopathy [8]. Similar patterns of DWI abnormalities have been described for syndrome of bilateral basal ganglia lesions [7]. Li et al. reported that there is no specific treatment for this condition, but this condition was accompanied by cytotoxic edema and choreic movements. Thus we prescribed prednisolone and valproic acid, and his symptoms and signs were improved [6].

In fact, our patient experienced typical hypoglycemic symptoms as confusion and sweating one day before the onset of choreic movement, and his glycated hemoglobin was low as 5.2 %, but we couldn't check his objective blood glucose level. This is limitation of our report to clarify this case of acute bilateral basal ganglia lesion is due to hypoglycemic event.

In summary, we treated a patient with hypoglycemia antecedent to syndrome of acute basal ganglia lesions in uremia. Clinicians should keep in mind that hypoglycemia is to be avoided in patients with end-stage kidney disease because 'tight' glycaemic control can lead to recurrent hypoglycaemia which is potential predisposition to acute reversible bilateral basal ganglia lesions.

CONSENT

In this paper, only published data from the literature were used for description. Thus, a statement of patient consent is not applicable for this paper.

ETHICAL APPROVAL

Since data from the literature, only, were used for description, ethical approval is not applicable to this paper. This study is not against the public interest. All authors hereby declare that all description have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki.

COMPETING INTERESTS

Authors have declared no competing interests.

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