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Case Report

Diabetic Striatopathy: An Uncommon Complication Of Diabetes Mellitus

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Abstract

Diabetic striatopathy is an uncommon neurological manifestation of hyperosmolar hyperglycemic syndrome. A case of a 74-year-old woman is presented here. She presented with abnormal movements of left upper and lower limbs and was found to have high index blood sugar. Non contrast CT brain showed hyperdense area in right striatal region. Based on clinical presentation, high blood sugar and radiological finding, she was diagnosed with diabetic striatopathy. The radiological abnormality and the symptoms improved rapidly and remarkably with strict glycaemic control.

Keywords

Diabetic striatopathy, hyperosmolar hyperglycaemic syndrome

Background

Diabetic striatopathy (DS) can be defined as a hyperglycemic condition associated with sudden onset of either or both of the following two conditions: chorea/ballism; striatal hyperdensity on CT or hyperintensity on T1-weighted MRI (1). Also termed as "chorea/hemichorea associated with non-ketotic hyperglycemia", "hyperglycemic non-ketotic hemichorea/hemiballism", or "chorea, hyperglycemia, basal ganglia syndrome", "diabetic hemiballism/hemichorea", it is an uncommon neurological manifestation of hyperosmolar hyperglycaemic syndrome (HHS).

Chorea is a hyperkinetic disorder characterized by involuntary, rhythmic, purposeless, jerky movements mainly involving the distal limbs. Proximal muscle involvement with larger amplitude flinging movements can be defined as ballism. Vascular, metabolic, structural, inflammatory, infectious, autoimmune, and iatrogenic aetiologies leading to basal ganglia and subthalamus dysfunction, cause chorea or ballism.

Case Presentation

A 74-year-old woman presented with abnormal involuntary movements of left upper limb and lower limb for a duration of three weeks, worsening over last two days. The movements first developed in the left leg and then progressed to left arm. She could not suppress the movements voluntarily, but they reduced during sleep. She has been a type-2 diabetic with a strong family history for the past 7 years and was poorly compliant to diet control and oral hypoglycaemic agents.

She did not have a history of stroke, there was no associated tongue bite, upward eye rolling, frothing from mouth or sphincter incontinence. There was no trauma history. She was not on any antipsychotic drug history. She also had hypertension, dyslipidemia and was on losartan, aspirin and atorvastatin.

Neurological examination was normal, with normal power, sensation, and reflexes with down-going plantar reflexes. There were no cerebellar signs and cranial nerves were intact.

On admission, her RBS was high index, her last HbA1c report value was 10.7%. pH was in the normal range with no ketones found in urine. Serum sodim, Pottasium, Thyroid fuction test, Lipid profile, Serum Ca, Mg, CRP, FBC, Creatinine values were all normal. ECG, Echo and CXR did not show any abnormalities. Urgent non-contrast CT – Brain showed a well demarcated hyper-

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density over right striatal region without peripheral oedema. MRI was not done due to the unavailability. A diagnosis of diabetic striatopathy was made.

In-ward management included adequate hydration of 3-3.5 L/day, strict blood sugar control with the use of soluble insulin, dose adjusted according to pre-meal RBS. Small doses of haloperidol and tetrabenazine were also started to alleviate the symptoms.

After seven days, the involuntary movements diminished in the leg, and were completely absent in the hand. In the follow up CT Brain, the hyperdense area had disappeared. RBS was within the normal range.

Patient was discharged with mixtard insulin, MRI date was taken and HbA1c was planned in the subsequent clinic visits.

Discussion

Diabetes mellitus striatopathy is a rare medical condition with a prevalence of 1 in 100,000 (2). Although usually reported in Asians, females and elderly with hyperosmolar hyperglycaemia, diabetes mellitus striatopathy has been reported in males and children (3, 4) also. In a systemic review, the average blood glucose and glycated haemoglobin values were found to be 414 mg/dL and 13.1% respectively (1).

There are characteristic radiological findings in CT and MRI brain: CT scan shows hyperdense region in the contralateral caudate nucleus and putamen (striatum) while MRI shows T1 hyperintensity of the affected striatum, MRI being more sensitive (1). Exact pathology for the radiological findings have not been understood clearly. The proposed hypotheses include petechial haemorrhages, mineral deposits, myelin destruction and infarction with astrocytosis.

The commonest presentation of diabetic striatopathy is in a diabetic patient with hemichorea/ hemiballism and radiological evidence (1). Sometimes, it may be the first presentation of diabetes mellitus (5). Recurrence of hemichorea/ hemiballism have also been reported (6). There is also a possibility of bilateral limb involvement (7). Few cases have been reported with involuntary movements but no radiological findings (8). One patient presented with altered consciousness and T1-hyperintense lesion in striatum but with no abnormal movements (9).

When diabetic retinopathy is present, evidence suggests that the prognosis is poor in patients with hyperkinetic disorders associated with DM (8). When the symptoms are refractory, the use of medications such as haloperidol, tetrabenazine and risperidone has also been reported as efficient to control the movements (10).

Conclusion

Diabetic striatopathy is an uncommon presentation in patients with poorly controlled diabetes mellitus. High degree of suspicion is required when a patient presents with involuntary movements with or even without hyperglycaemia. Although poorly understood, it is condition which is almost always completely reversible with early and aggressive glycaemic control.

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