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Case Report

Dyskinesia as an Inaugural Symptom of Diabetes Mellitus

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Summary

The prevalence of diabetes mellitus has been increasing, along with its multiple complications. Diabetic striatopathy is a rare but potentially life-threatening complication of diabetes mellitus.

We present the case of an 87-year-old woman who experienced a 2-day course of right hemiballismus associated with facial dyskinesias. Analytically, the patient had a glycated hemoglobin level of 14%, and a brain CT scan showed hyperdensity of the left basal nuclei, with no other relevant positive findings. Thus, the diagnosis is compatible with diabetic striatopathy. After glycemic control and the introduction of haloperidol, there was a clear symptomatic improvement.

This case emphasizes the need to consider diabetic striatopathy as a differential diagnosis in diabetic patients with dyskinesias, as timely treatment can improve outcomes. Although it is a rare entity, the increasing prevalence of diabetes may make it more frequent. Therefore, this case also aims to raise awareness of this pathology..

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Background

It is well known that the prevalence of diabetes mellitus (DM) is increasing, along with its complications. Diabetic striatopathy (DS) is an uncommon and potentially life-threatening manifestation of diabetes mellitus (1). It classically presents with a limited period of choreiform or ballistic movements associated with an episode of nonketotic hyperglycemia or diabetic ketoacidosis, along with characteristic imaging findings (2). DS can also be the first presentation of DM (3).

There are numerous possible etiologies of hemichorea-hemiballism, including cerebrovascular, metabolic, toxic, infectious, inflammatory, autoimmune, neurodegenerative, hereditary, and malignant causes (1). This clinical case highlights the importance of considering diabetic striatopathy as a potential cause of hemiballismus, despite its rarity, since it is a correctable condition. Early diagnosis and treatment can lead to improved patient prognosis and a shortened length of hospital stay (1).

Case Presentation

We report the case of an 87-year-old woman with mild cognitive impairment, dyslipidemia, arterial hypertension, chronic kidney disease (KDIGO G3b/A1), autoimmune thyroiditis, and depression. Her regular medications included lisinopril 5 mg, levothyroxine 75 mcg, sertraline 50 mg, atorvastatin 80 mg, and sulpiride 50 mg, none of which had undergone recent dosage adjustments.

The patient presented to the emergency department (ED) with a 2-day history of involuntary movements in her face, right arm, and leg, with a clear worsening on the day of admission. Upon clinical evaluation, vital signs were normal. Notably, there was the presence of oral dyskinesias associated with irregular, coarse, continuous, nonsuppressible, and involuntary movements involving the right limbs, suggestive of right hemiballismus.

Except for this movement disorder, the neurological examination was unremarkable, with a Glasgow Coma Scale score of 15.

The brain computed tomography (CT) showed no evidence of acute hemorrhagic lesions or recent ischemic lesions with cortical involvement. However, there was hyperdensity of the left basal nuclei (Image 1). The angiographic study revealed signs of atherosclerotic disease along the supra-aortic trunks, without hemodynamic repercussions.

The patient's complete blood count, electrolyte levels, liver function tests, and thyroid function tests were normal. Renal function was at baseline (creatinine 1.4 mg/dL). Postprandial blood glucose was elevated (glucose 311 mg/dL), as was glycated hemoglobin (Hb A1c 14.6%). There was no evidence of ketoacidosis. Serological testing for HIV, HBV, HCV, and syphilis was negative. Both serum antinuclear antibodies and antiphospholipid antibodies were also negative.

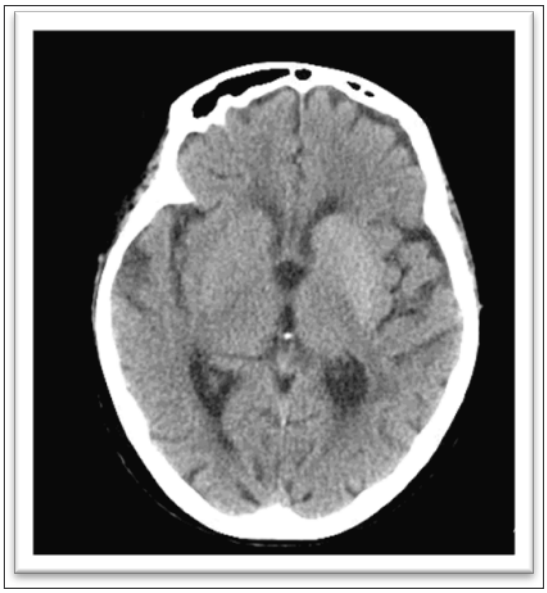

Image 1. *Brain computed tomography (CT) revealing an hyperdensity of the left basal nuclei. No evidence of acute hemorrhagic lesions nor recent ischemic lesions with cortical involvement.*

Differencial Diagnosis and Treatment

Unlike chronic chorea, where heredodegenerative causes predominate, three main categories play significant roles in acute chorea: vascular, metabolic, and inflammatory (4). A vascular cause was ruled out due to the absence of focal neurological signs and recent ischemic lesions on brain CT. Among metabolic causes, diabetic striatopathy was the most likely diagnosis. This was based on blood analysis and neuroimaging findings. Laboratory tests showed elevated blood sugar and HbA1c levels, indicating poorly controlled diabetes. The brain CT further provided evidence of diabetic striatopathy with the typical asymmetric hyperdense contralateral basal ganglia lesion.

Among other metabolic causes, it was important to consider drug-related chorea. Several centrally acting drugs, such as neuroleptics, have been identified as potential causes of medication-induced chorea (4). The patient was taking sulpiride, a selective antagonist of dopamine D2 receptors (5). These drugs are known to cause extrapyramidal syndrome (EPS), which includes dyskinesia that can manifest as either orofacial dyskinesia or limb and trunk movements (6). However, this would not explain the asymmetry observed or the basal ganglia lesions.

We did not find any evidence of other causes of metabolic chorea, such as hypo/hypernatremia, hypocalcemia, hyperthyroidism, hypoparathyroidism, or hepatic/renal failure. Additionally, we found no evidence of inflammatory chorea, including antiphospholipid antibody syndrome, postinfectious or postvaccinal encephalitis, paraneoplastic chorea, neurosyphilis, or viral encephalitis.

Although chorea can be successfully treated with glucose control, this occurs in only one-fourth of patients. The vast majority require additional anti-chorea medications for symptom control (3).

During hospitalization, the patient began glycemic correction using basal insulin (glargine), with mealtime insulin added as needed. As better glycemic control was achieved, there was significant and progressive improvement in choreic movements. She also started haloperidol, with the dose adjusted up to 8 mg/day, which provided symptomatic benefits. At the time of discharge (approximately 48 hours after admission), there were no involuntary movements, and the remaining neurological examination was normal. Both the patient and her family were engaged in learning how to administer insulin. The outpatient insulin regimen chosen was a combination of insulin lispro protamine and insulin lispro (Humalog Mix 75/25).

The patient was reassessed in an outpatient consultation at 5 months. On her own initiative, she stopped taking haloperidol. There was persistence of orofacial dyskinesias, but without functional impact. No other neurological changes were noted. There was a clear improvement in glycemic control, with a significant reduction in HbA1c to 5.5%.

Discussion

Diabetic striatopathy (DS) is defined as a hyperglycemic condition associated with either or both of the following: chorea/ballism and striatal hyperdensity on CT or hyperintensity on T1-weighted MRI (3). It tends to occur in the elderly, particularly in females and individuals of Asian descent (1), and most importantly, in patients with long-standing poor control of diabetes mellitus (DM) (3). Typically, a diabetic patient develops severe non-ketotic hyperglycemia, and as the hyperglycemia progresses, hemichorea-hemiballism begins (1). Although the syndrome primarily occurs in non-ketotic hyperglycemia, some patients also experience episodes of ketotic hyperglycemia (7).

The susceptibility of DS to non-ketotic hyperglycemia may arise from the underlying pathophysiology of chorea. In a non-ketotic hyperglycemic state, brain metabolism shifts to an alternative anaerobic pathway in the Krebs cycle, leading to rapid depletion of gamma-aminobutyric acid (GABA). This results in disinhibition of the subthalamus and basal ganglia, causing hyperkinetic movements. Conversely, in ketosis, GABA can be resynthesized using acetoacetate produced in the liver, which helps prevent its depletion, thus explaining the less common occurrence of DS in diabetic ketoacidosis (3).

To date, the precise pathophysiology of diabetic striatopathy is not well understood (2). Several hypotheses have been proposed to explain the pathogenesis of this condition beyond GABA depletion, including petechial hemorrhage, mineral deposition, myelin destruction, and infarction with astrocytosis (2)(3)(8). The reason for striatal vulnerability to DS remains unclear. The most common pattern of striatal involvement in DS is isolated putamen involvement, followed by combined caudate nucleus-putamen involvement. Concomitant anomalies in all three striatal components are more rarely observed (3).

Presentation varies among patients. Generally, there is unilateral limb involvement, with only 9.7% of cases reporting bilateral involvement. Involuntary movements can start abruptly or insidiously, ranging from low to high amplitude, and may manifest intermittently or continuously. In terms of affected body regions, the highest frequency of extremity involvement occurs in the order of arm-leg, arm-leg-face, and isolated arm. There are also reported cases of isolated facial hemichorea presenting with oral dyskinesia and grimacing. Most patients with chorea experience worsening of symptoms during periods of nervousness, with symptoms resolving after sleep (3).

Evidence suggests that the presence of diabetic retinopathy is an indicator of worse prognosis in patients with hyperkinetic disorders related to diabetes (8).

To our knowledge, there are currently no established guidelines for diagnosing or treating this entity. Regarding treatment, previous studies indicate that correction of hyperglycemia typically leads to complete or partial resolution of chorea as well as striatal abnormalities on neuroimaging studies. However, this approach is effective in only about one-fourth of patients. The majority appear unresponsive to strict glucose control and require additional anti-chorea medications for symptom management. The main categories of anti-chorea medications include antipsychotics, GABA-receptor agonists, selective serotonin reuptake inhibitors, and dopamine-depleting agents (3). The most commonly used monotherapeutic agent for DS-associated chorea is haloperidol, which was effective in this particular case.

If hyperglycemia remains uncorrected, it may lead to permanent abnormal movements and structural brain changes (2). The overall recurrence rate is estimated to be as high as 20% even after the resolution of striatal anomalies, highlighting the need for regular follow-ups regardless of neuroimaging findings (3).

This case underscores the importance for clinicians to recognize diabetic striatopathy as a potential cause of hemichorea-hemiballism, given that it is a correctable condition. Early diagnosis and treatment can significantly improve patient prognosis and shorten the length of hospital stay (1).

References

1. Lin CJ, Huang P. Delayed onset diabetic striatopathy: Hemichorea-hemiballism one month after a hyperglycemic episode. Am J Emerg Med [Internet]. 2017;35(7):1036.e3-1036.e4. Available from: http://dx.doi.org/10.1016/j.ajem.2017.02.018
2. Lucassen EB, Delfyett WT, Stahl MC. Persistent Hemichorea and Caudate Atrophy in Untreated Diabetic Striatopathy: A Case Report. Case Rep Neurol. 2017;9(3):299–303.
3. Chua CB, Sun CK, Hsu CW, Tai YC, Liang CY, Tsai IT. "Diabetic striatopathy": clinical presentations, controversy, pathogenesis, treatments, and outcomes. Sci Rep. 2020;10(1):1–11.
4. Silva GD, Parmera JB, Haddad MS. Acute chorea: Case series from the emergency room of a Brazilian tertiary-level center. Arq Neuropsiquiatr. 2021;79(3):233–7.
5. Lin Y, Tsai MDK, Chou FH, Ph D. Amisulpride implicated in the onset of tardive dyskinesia in a man who previously took sulpiride. 2009;15(2):135–40.
6. Choudhury S, Chatterjee K, Singh R, Shubham S, Trivedi S, Chatterjee S, et al. Levosulpiride - induced Movement Disorders. 2018;2–3.

7. Abe Y, Yamamoto T, Soeda T, Kumagai T, Tanno Y, Kubo J, et al. Diabetic striatal disease: Clinical presentation, neuroimaging, and pathology. Intern Med. 2009;48(13):1135–41.

8. Homaida M, Kanodia AK, Young N, Yu WM. Diabetic striatopathy: A rare condition and diagnostic dilemma. BMJ Case Rep. 2021;14(1):1–3.