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Case Report

Hemiballism Revealing Hyperglycemia without Ketosis, Complicated by Cortical Laminar Necrosis

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Abstract: Hyperglycemia without ketosis is a rare cause of hemiballism. The secondary complication of this entity of Cortical Laminar Necrosis is rare. Brain MRI is of major help to detect basal ganglia involvement and gyriform hyper signals that are the signature of laminar cortical necrosis. We report a case of hemiballism associated with hyperglycemia without ketosis, which was secondarily complicated by cortical laminar necrosis with a fatal outcome.

Keywords: Hyperglycemia, hemiballism, laminar cortical necrosis, MRI.

INTRODUCTION

Hyperglycemia without ketosis is a rare cause of hemiballism [1]. The secondary complication of this entity of cortical laminar necrosis is rare. We report a case of hemiballism associated with hyperglycemia without ketosis that was complicated secondarily by cortical laminar necrosis of fatal progression.

OBSERVATION

Mr. KS, aged 60, a teacher by profession, right-handed laterality, and a known type 2 diabetic on insulin. Admitted for abnormal uncontrollable movements of the right upper limb. These abnormal movements were involuntary, abrupt, violent, and of great amplitudes that tended to increase by contrariety. The clinical laboratory tests revealed hyperglycemia at 10 g/dl without ketosis, with glycated hemoglobin (HbA1c) at 11%. The hydroelectrolyte, renal, and hepatic test results were normal. The styphinic and HIV serologies were negative. There were no other signs indicative of infection. A brain MRI exhibited a hypersignal in FLAIR and diffusion (Fig A) of the head of the left caudate nucleus and the left insular region with restriction on the ADC mapping. The gradient echo sequence (T2*) did not reveal signs of bleeding or calcification. The cardiac assessment and imaging of the supra-aortic trunks were without anomaly. The hypersignal of the head of the caudate nucleus in a context of diabetes and major hyperglycemia led us to believe that the disorders had a metabolic origin. The change in the condition was marked by gradual regression of the involuntary movements after 48 to 72 hours of haloperidol administration, with correction of the hyperglycemia. Two weeks later, the patient exhibited behavioral disorders with agitation, dreamlike confusion, and visual hallucinations "he saw animals and felt like he was free-falling." The follow-up brain MRI performed in light of the clinical exacerbation revealed a spontaneous gyriform hypersignal of the left cortical ribbon on FLAIR and diffusion (Fig B) sequences, with restriction on the ADC mapping without signs of bleeding on the gradient echo sequence. The EEG (Fig C) performed in light of acute delirium syndrome revealed an overall slowdown in brain activity without epileptic abnormality. The clinical progression was fatal, with death of the patient 48 hours later.

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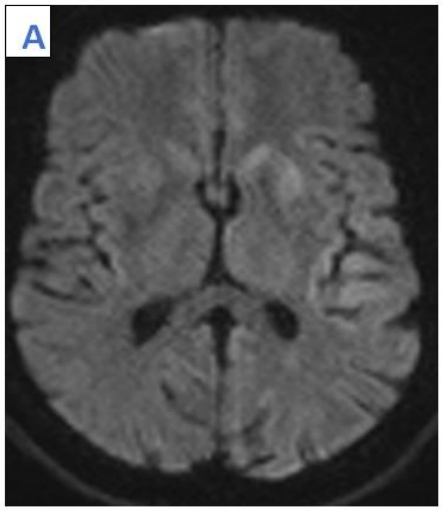

Fig A: The brain MRI performed initially revealed hyper signal in diffusion of the head of the left caudate nucleus and the left insular region

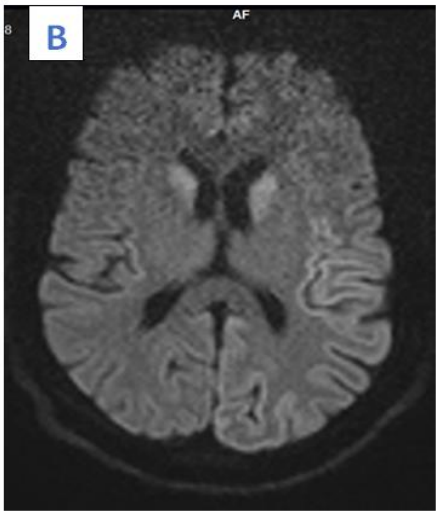

Fig B: The brain MRI follow-up performed in light of the clinical exacerbation revealed a spontaneous gyriform hyper signal of the left cortical ribbon on the diffusion sequences

Fig C: The EEG performed in light of acute delirium syndrome revealed an overall slowdown in brain activity, without epileptic abnormality

DISCUSSION

Hemiballism is a rare manifestation of hyperglycemia without ketosis [2], although in some sources, hyperglycemia without ketosis appeared to be the second cause of hemichorea-hemiballism in elderly diabetic subjects, after a hemorrhagic or ischemic vascular etiology [2, 3]. It may, however, be indicative of diabetes [2, 4]. The pathophysiological mechanism of this rare clinico-radiological entity is poorly understood and remains a controversial subject [5, 6]. During abnormal movements, the putamen is always affected, the head of the caudate nucleus is affected in the majority of cases, while pallidum involvement remains rare [3, 5]. The lesions are often unilateral but can be bilateral even without clinical symptoms [5]. Spectroscopic studies have shown a decrease in the peak of N-acetyl-aspartate (NAA) and creatine (Cr) as well as the NAA/Cr ratio in relation to neuronal loss or dysfunction [7]. The two most supported theories explaining these basal ganglia abnormalities during hyperglycemia without ketosis are ischemia and metabolic disorders [2, 3, 6]. It has been reported that hyperglycemia, whether chronic or acute, alters brain metabolism, with pronounced cellular acidosis related to a local increase in lactates, which contributes to cellular dysfunction by tissue ischemia [2], and that GABAergic striatal neurons are particularly vulnerable to ischemia, hence the occurrence of abnormal movements [2]. According to Battisti *et al.*, during hyperglycemia, brain metabolism switches to anaerobic mode. To compensate for the energy deficit, the brain transforms GABA into succinic acid. This results in GABA and acetate deficiencies as well as a reduction in

acetylcholine synthesis. The simultaneous deficiency of acetylcholine and GABA in the gray matter nuclei of the basal ganglia associated with an energy deficit and metabolic acidosis may be the cause of the dysfunction of the basal ganglia and the appearance of abnormal movements such as hemiballism [4]. This hypothetical pathophysiological mechanism does not explain the persistence of abnormal movements despite the correction of hyperglycemia in some cases [5] as well as the unilateral nature of the symptomatology in most cases [3, 4]. On MRI, there is a hypersignal of the striatum on the T1 sequence, with a variable signal on the T2 sequence that can be hyper-, iso-, or even hypo-intense. The diffusion sequence as well as ADC mapping may highlight restriction of diffusion [2, 5, 6]. The restriction of diffusion noted in our observation as well as in some published cases [2, 8] could be explained by limitation of the mobility of water molecules secondary to hyperviscosity induced by hyperglycemia, hypoperfusion, and cerebral ischemia. In our case, the diagnosis was made on the basis of clinical and radiological evidence, including hemiballism in a context of hyperglycemia without ketone with hypersignal of the head of the caudate nucleus contralateral to the affected hemibody on a brain MRI. Another feature of our case was the secondary occurrence of cortical laminar necrosis (CLN). CLN is a rare clinical-radiological entity. It corresponds to neuronal ischemia associated with a glial reaction and laminar deposition of lipid-rich macrophages [9]. Indeed, since the brain is a glucodependent organ, this cerebral glucodependency means that hyperglycemia and hypoglycemia are both likely to lead to cortical laminar necrosis. CLN can also be observed as a result of prolonged low blood pressure, epileptic disease, or cardio-respiratory arrest. Our patient was not experiencing hypoglycemia, and there was no epileptic abnormality by EEG. Radiologically, only MRI can detect and monitor the progression of CLN. It corresponds to a spontaneous linear or gyriform hypersignal visible on T1, T2, and FLAIR sequences, with the absence of any hemorrhage on gradient echo sequences. In the absence of complications, hemiballism during hyperglycemia without ketosis usually has a good prognosis. The treatment of choice remains neuroleptics and correction of hyperglycemia [6]. Our patient was on haloperidol with hyperglycemia correction. The clinical course was initially marked by the gradual regression of the abnormal movements. Two weeks later, the patient's condition worsened due to the onset of a confusional state with hallucinations, followed by death 48 hours later.

CONCLUSION

Hemiballism is rarely mentioned as a complication of hyperglycemia without ketosis. Brain MRI is a major contributor to detection of damage to the central gray nuclei and the gyriform hyperignals that are a signature of cortical laminar necrosis.

Conflict of Interest: None

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