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'I'M SUGAR HIGH, I CAN'T STOP DANCING' A RARE CASE OF HYPEROSMOLAR HYPERGLYCEMIC STATE (HHS) PRESENTED WITH HEMIBALLISMUS HEMICHOREA

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Introduction

Hemichorea is a non-rhythmic, non-suppressible one-sided, involuntary jerky movement due to overactivity of dopamine in area that control movement in brain, hemiballismus is a severe form of chorea which characterized by involuntary and violent course of movement. The most common cause of hemiballismus or hemichorea is stroke (infarction or hemorrhage), however it can also cause by non ketotic hyperglycemia [1-2]. Non-ketotic hyperglycemia presented as hemiballismus hemichorea is a rare manifestation but it can easily be reversed with tight glucose control. Here we describe a case of an elderly lady which was diagnosed with hyperosmolar hyperglycemic state (HHS) and presented to us with hemiballismus hemichorea.

Case report

This is a case of 71-year-old lady who had underlying poorly controlled long standing Diabetes Mellitus (DM) (hba1c 14.8%), hypertension, dyslipidemia, ischemic heart disease, chronic kidney disease stage 3A and history of upper gastrointestinal bleeding presented to the Emergency Department (ED) with one day history of involuntary left sided upper limb and lower limb twitching and jerking movement. The movement has been continuously present for 5 hours and disturbed her daily activity. She was fully conscious during the twitching episodes, other than that she also complains of poor oral intake for the past 1 week and lethargy for the past 1 week. Otherwise, there is no tongue biting, no up rolling of eyeball, no fever, no neck stiffness and no blurring of vision, no limb weakness. She had no recent head trauma, no previous history of seizure.

On presentation, patient is a medium build elderly lady, vital sign included blood pressure is 84/55 mmHg, heart rate of 91 beats per minute, respiratory rate of 16 breath per minute, not appear in respiratory distress with GCS 15. Patient was jerking both upper limb and lower limb upon examination and orientated to time place and person. Otherwise her neurological examination shows normal cranial nerve function, normal power and reflex bilaterally, other physical examination was unremarkable. Her blood pressure response to fluid challenge of 100 ml NS and subsequently her blood pressure remains normotensive with maintenance intravenous fluid given. She was given 5 mg IV diazepam which stopped her twitching movement.

Laboratory evaluation shows high blood glucose (49.9 mmol/L) with pseudohyponatremia 131 mmol/L and increase effective plasma osmolality of 324 mOsm/kg with absence ketoacidosis. On top of that she also has acute kidney injury with creatinine of 213.16 µmol/L and urea 13.37 mmol/L. Other laboratory investigation is within normal limit. Non contrast CT brain was done to rule out intracranial bleeding or cerebrovascular accident which shows bilateral basal ganglia small hypodensities and calcification, there was no sign of intracranial bleeding noted, no cerebral edema and no midline shift. She was admitted for glucose control, in the ward her symptoms subsided with tight capillary glucose control. However, she had one episode of jerking of left upper limb and lower limb when her blood glucose increases again (16 mmol/l), similar to initial presentation which spontaneously resolved. Non contrasted CT brain was repeated at 72 hours of admission and showed no changes from initial presentation.

Subsequently there was no more jerking or twitching episode observed and her glucose level was adequately controlled, she was discharge well after 5 days of admission with diabetic education given prior to discharge. Subsequently during her clinic review 2 months later patient did not complain of any twitching movement and her glucose level is satisfactory upon review.

Discussion/ Conclusion

Hyperglycemic hemiballismus hemichorea can easily be overlooked due to unfamiliarity with this syndrome. It is usually mistreated as other more common disease such as cerebrovascular accident, meningitis or seizure. In this case patient was initially thought to have focal seizure or intracranial bleed however it did not explain that patient was fully conscious during the incident and repeated CT brain did not shows any bleeding. It was later, after discussion with specialist that the diagnosis of non ketotic hyperglycemia hemichorea hemiballismus was established.

Non ketotic hyperglycemia associated with chorea or ballism was first described by Bedwell in 1960 [3], it is a rare manifestation of hyperglycemia with a prevalence of less than 1 in 100 00 which majority affecting elderly Asian women [4] .Classically it presented with triad of unilateral or bilateral involuntary movements, hyperglycemia and contralateral or bilateral striatal abnormality on neuro imaging in patient with known case of diabetes or newly diagnosed diabetes [5].Its exact pathogenesis was still unknown but was thought related to hyper viscosity due to hyperglycemia causing ischemia, striatal microhemorrhage and cerebral mal perfusion[6]. The symptoms of involuntary movements can last up for days or even months and its bizarre presentation can be overwhelming for some clinician, nonetheless this symptom has good prognosis and can easily resolved with early recognition and treatment to reduce glucose level. Thus, it is imperative for clinician to be vigilant about this syndrome.

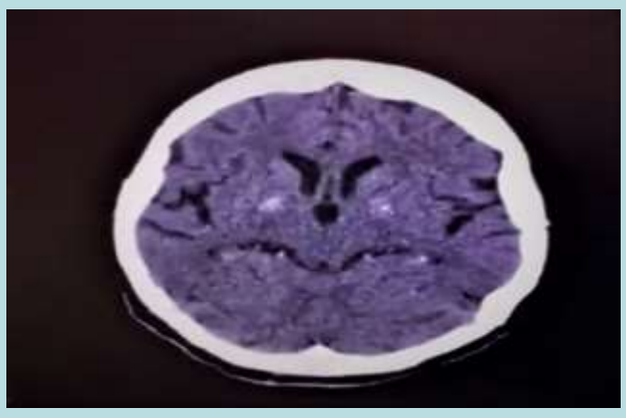

Figure 1: CT brain on initial presentation showing bilateral basal ganglia calcification (hyperdensities)

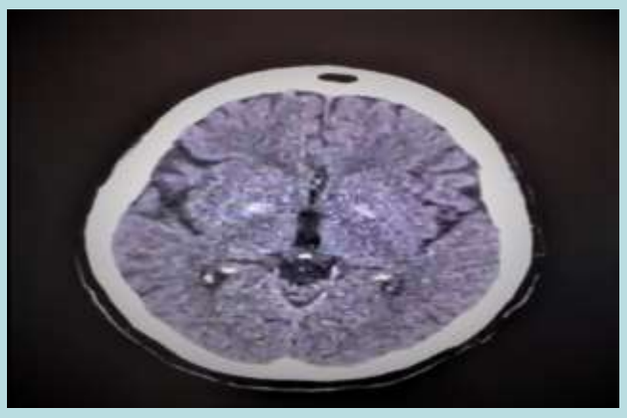

Figure 2: Repeated Ct brain after 72 hours showing similar changes as presentation

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Declaration of conflict for all authors

Informed consent was obtained from the patient to be included in this case report. All authors have no conflict of interest.

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