Case Report

"Dancing belly" in an old diabetic lady

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Abstract

Movement disorder, although rare, is increasingly being recognized as the initial presenting sign of the hyperglycemic state. Although chorea-ballism has frequently been reported among diabetics, monoballism is a very rare phenomenon. While myoclonus is common, diaphragmatic myoclonus is extremely rare. Moreover, diaphragmatic myoclonus as the initial presenting manifestation has never been reported before. Herein, we report an index case of a 62-year-old previously undiagnosed diabetic lady presented with acute onset constellation of multiple abnormal movements viz. monoballism, focal myoclonus, action myoclonus, and diaphragmatic myoclonus. All of them disappeared with achieving normoglycemia. This case underscores the importance of rapid capillary blood glucose testing in any patient presenting with acute onset abnormal movements. This approach can especially be rewarding as it helps in the rapid diagnosis of a reversible catastrophe and avoiding unnecessary costly investigations.

Keywords: Diabetes mellitus, diaphragmatic myoclonus, hyperglycemia, monoballism, movement disorders, myoclonus

Introduction

Diabetes mellitus is associated with a wide spectrum of movement disorders ranging from epilepsia partialis continua, chorea-ballismus, myoclonus, rubral tremor, dyskinesia, restless leg syndrome, hemifacial spasm to parkinsonism.[1,2] These abnormal movements can stem from a direct complication of both hyperglycemia and hypoglycemia or may be associated with diabetic vasculopathy, neuropathy, or nephropathy.[1] These might herald the onset of acute ketotic or non-ketotic complications among previously undiagnosed diabetics.[3,4] Thus, identification of these abnormal movements can lead to earlier diagnosis and prevention of mortality.

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Although hemiballismus-hemichorea (HB-HC) has fairly been reported as a direct complication of hyperglycemia, monoballism is rare.[1] Myoclonus is common in the context of uremia following end-stage diabetic nephropathy.[5] While diaphragmatic myoclonus is extremely rare, this particular movement disorder secondary to hyperglycemia has never been reported before. Herein, we are reporting a case of a previously undiagnosed diabetic old lady who presented to us with a constellation of different types of abnormal movements viz. monoballism, focal myoclonus, action myoclonus, and diaphragmatic myoclonus which was abolished with achieving normoglycemia.

Case Report

A 62-year-old lady presented with some acute onset abnormal movements simultaneously involving bilateral upper limbs and abdomen for the last 12 h. She described the movement of the right upper limb as "involuntary jerks" whereas her description

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regarding the left upper limb was that she could not control the concerned limb. She also jokingly stated her abdominal movement as an "automatic dancing belly." There was no history of loss of consciousness, focal weakness, headache, fever, and vomiting. She was a healthy lady without any medical or surgical issues so far. She had no family history of any movement disorder. Drug history was non-contributory.

Examination revealed GCS 15/15, normal cognition, no cranial nerve deficit, meningeal, sensory, or autonomic involvement. In right upper limb involuntary, rapid, flinging, high amplitude, and arrhythmic movement suggestive of monoballism was observed [Video 1]. In the left upper limb, there were frequent, involuntary, rapid, brief, arrhythmic jerks suggestive of focal myoclonus at rest [Video 1]. There were abnormal, involuntary, arrhythmic, inward, and outward movements of the abdominal wall suggestive of diaphragmatic myoclonus [Video 2]. No abnormal movements were noted in the lower limbs. There was no muscle wasting anywhere. The tone and power of bilateral upper limbs could not be assessed due to abnormal involuntary movements. The tone and power of bilateral lower limbs were normal. Deep tendon reflexes were 2+ in bilateral upper and lower limbs, plantar reflexes were bilateral flexor. Cerebellar function testing (finger-nose, finger-nose-finger) revealed action myoclonus which was brief, involuntary, arrhythmic jerks persisting during full range of motion [Video 3]. No abnormal orolingual movement was observed.

She was investigated instantly with possibilities of metabolic and acute onset structural lesion (vascular) affecting extrapyramidal circuit keeping in mind. Bedside capillary blood glucose (CBG) was done immediately and was found to be 616 mg/dL. Arterial blood gas (ABG) analysis was done next and was noncontributory. ABG was marked for the striking absence of acidosis and normal osmolarity. Urine dipstick test revealed glucosuria, but no ketone bodies. Magnetic resonance imaging (MRI) brain and electroencephalogram (EEG) were normal. All other tests of the metabolic panel (complete hemogram, serum electrolytes, renal, liver, and thyroid function tests) and autoimmune profile were within normal limits.

Immediate treatment with injection regular insulin at the rate of 7 IU/h by infusion pump was started along with adequate hydration followed by regular monitoring of CBG and ABG. Interestingly, once blood sugar comes down to less than 200 mg/dL, approximately 6 h after therapy, all movements disappeared simultaneously. Her HbA1c was 11.37%, fasting and post-prandial blood glucose were 117 mg/dL and 201 mg/dL, respectively. The rest of her hospital stay was uneventful and did not have a recurrence of involuntary movements.

Discussion

"Ballism" is characterized by violent, flinging, large-amplitude, arrhythmic, and non-suppressible movement of the body due to the predominant involvement of proximal muscles. Monoballism refers to the involvement of a single limb and is a much rarer phenomenology than hemiballism. The most common etiology of monoballism is stroke, involving the subthalamic nucleus.[6] Hyperglycemia as a reversible cause of monoballism is rare. Till date, only three cases have been reported where monoballism was presenting features in the hyperglycemic state.[6-8] HB-HC, in the setting of inadequate glycemic control, is more common among Asian's, old people, and females.[9-11] To explain HB-HC in hyperglycemia there are several postulations such as striatal petechial hemorrhage, decreased cerebral perfusion due to hyperviscosity, acanthocytosis, associated dyselectrolytemia, metabolic acidosis, and decreased γ-amino butyric acid (GABA)- an inhibitory transmitter in the thalamocortical pathway.[1] Brain imaging findings are variable ranging from normal to intensity change in the putamen, with minimal extension to surrounding structures, but no perilesioal edema or diffusion restriction.[1,9,10] Most radiological lesions disappear with correction of hyperglycemia, while few may take months to regress.[9,10] There is a report on the development of diabetic striatopathy even 1 month after a hyperglycemic episode.[12] The rate of improvement varies ranging from a few days with only supportive therapy directed at achieving euglycemia to refractory HB-HC necessitating other therapies.[13,14] In our case, neuroimaging was normal, which might indicate acute onset and rapid reversibility of hyperglycemia before neuroradiological changes could take place.

Myoclonus is sudden, brief, non-rhythmic, shock-like involuntary movements, associated with either burst of muscular contraction (positive myoclonus), or muscular inactivity (negative myoclonus).[5] It may be present at rest, during voluntary movement (action-induced), or secondary to provocation by sensory stimuli. Anatomically it can be cortical, subcortical, segmental or peripheral, and propriospinal.[5] While myoclonus can be physiological or a part of various epilepsy syndromes, a wide array of metabolic derangements cause transient myoclonus, particularly among the elderly.[5] Similar to other hyperkinetic disorders, hyperglycemia may also cause myoclonus by thalamocortical GABA depletion.[1,5]

The most striking part of this case was the presence of three different types of myoclonus (focal, action, and diaphragmatic) and the simultaneous disappearance of the same with achieving euglycemia. Diaphragmatic myoclonus or flutter, also known as van Leeuwenhoek's disease or moving umbilicus syndrome or belly dance syndrome, is a high-frequency, involuntary, non-suppressible contraction of diaphragmatic muscles.[15] It has been described in association with atrioventricular dysycnhrony, encephalitis, stroke, drugs, demyelination, cervical spine lesion, phrenic nerve irritation by cardiac electrode dislocation, and metabolic abnormalities.[16] Reversible diaphragmatic myoclonus in the setting of non-ketotic hyperglycemia has not been described previously. Its diagnosis needs a high index of suspicion due to its rarity and varied presentations such as a hiccup, epigastric pulsation, palpitation, hyperventilation, mostly to non-neurologists.[16] Although diaphragmatic myoclonus

remains a clinical diagnosis, video-fluoroscopy, electromyography, and ultrasonography can aid in the precise diagnosis.[17] Treatment includes phenytoin, carbamazepine, haloperidol, phrenic nerve crushing, etc.[16] In our case only correcting the hyperglycemia simultaneously abolished all the myoclonic jerks.

Conclusion

Movement disorders as the chief presenting sign of hyperglycemia are being increasingly appreciated. Thus, it is crucial that the emergency doctors, being primary care providers, become well-oriented with various movement disorders that might signal the presence of diabetes. Thus, we recommend that simple bedside CBG test should be done in any acute onset movement disorder even among previously undiagnosed diabetics, before jumping into costly investigations.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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