Case Report

Dancing Feet Syndrome in Diabetes : Para-ballism and Para-chorea in a Diabetic Patient with Diaphragmatic Myoclonus

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While movement disorders in Diabetes have long been recognized,the terminology of diabetic striatopathy is relatively more recent. Herein,we report a rare case of diabetic striatopathy in a 62-year-old woman with uncontrolled Type 2 Diabetes Mellitus who presented with Para-ballismus and Para-chorea along with diaphragmatic Myoclonus, a constellation of rarely reported before simultaneously in a hyperglycemic state.While these movement phenomena are extremely rare, the case also highlights they may persist even after acute control of hyperglycaemia, emphasizing on the need for achieving long term Glycemic control for its management. [*J Indian Med Assoc* 2023; **121(2):** 00-00]

Key words **: Striatopathy, Para-ballismus, Para-chorea, Myoclonus, Diabetes.**

Case Report

A 62-year-old known diabetic patient on oral anti diabetic drugs for 1 year presented with acute onset abnormal movements of her Left Lower Limb for 1 month. These movementswhich she described as involuntary jerky and dance like movements soon involved the Right Lower Limb and sequentially the upper limbs within the next 15 days. No history of any Focal Limb weakness, Sensory, Cranial Nerve or Sphincter abnormalities was present. Fever, headache, loss of consciousness, alteration of sensorium or specific drug use apart from oral anti diabetic drugs were not reported. Family history of movement disorders was not present.

On Examination she had a Glasgow coma Scale score of E4V5M6 with no significant cognitive impairment. Cranial nerve, sensory and autonomic examination were within normal limits. Motor examinationwas significant for abnormal, involuntary, hyperkinetic movements in the form of rapid, high amplitude and arrhythmic, flinging movements of both Lower Limbs suggestive of Para-Ballismus. Abnormal involuntary movements of distal muscles of Bilateral Lower Limb were also observed that were brief, random and without purpose, suggestive of Chorea. Additionally, there were also abnormal, involuntary, arrhythmic, undulating, inward and outward movements of the abdominal wall suggestive of diaphragmatic myoclonus. No abnormal movements were noted in the Upper Limbs. There was no muscle wasting anywhere. The tone and power of Bilateral Lower Limbs could not be assessed due to abnormal involuntary movements. The tone and power of Bilateral Upper 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 no abnormality. No abnormal orolingual movement was observed. Meningeal signs were absent.

Editor's Comment :

- Diabetes is a multisystem disorder affecting the whole body, Central Nervous System is also not an exception.
- Diabetes presenting with a movement disorder is very rare but is being noticed nowadays with increasing proportions.
- Any movement disorder presenting acutely it is a good habit to check the capillary blood glucose, which can be easily managed & patient will get immediate relief.

In view of the acute nature of the dyskinesias possibilities including metabolic and acute onset structural lesion (likely vascular) affecting the basal ganglia or its connections were considered. Bedside Capillary Blood Glucose (CBG) done immediately was found to be 170 mg/dl. However, she was found to have uncontrolled Blood Glucose levels with a Fasting Blood glucose level of 540 mg/dl and HBA1c 10.38. As euglycemia was achieved with Insulin and OADS, the dyskinesia decreased in intensity and persisted only present in Bilateral Lower Limbs. Arterial Blood Gas (ABG) analysis was done next which did not reveal acidosis and she had normal serum osmolarity. Urine dipstick test revealed glucosuria, but no ketone bodies. All other tests of the metabolic panel, Complete Hemogram, Serum Electrolytes, Renal, Liver and Thyroid Function Tests and Autoimmune profile were within normal limits. NCCT Brain showed bilateral caudate nucleus hyperdensity. MRI brain showed bilateral T1 and T2 hyperintensity of caudate and putamen. EEG was normal (Figs 1&2).

Discussion

With the ever expanding knowledge of the impact of diabetes on different organ functions, brain changes in diabetes is being increasingly recognized. Movement disorder is one amongst the myriad neurological presentations of Diabetes. While movement disorders have long been described, the terminology of “diabetics triatopathy” is relatively more recent[1]. Diabetic striatopathy

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Received on : 11/07/2021

Accepted on : 28/10/2021

is defined as a hyperglycemic condition associated with either both or one of the following conditions (1) chorea/ballism (2) striatal hyperdensity on CT or hyperintensity on T1 weighted MRI which can be reversible[1]. 4 main hypotheses to explain the pathogenesis resulting in striatal abnormalities on imaging include petechial haemorrhage,mineral deposition (Calcium or Magnesium), myelin destruction and infarction with astrocytosis (gemistocytopathy). In non-ketotic hyperglycaemia a shift in Brain metabolism to the alternative anaerobic pathway in Krebs Cycle leads to depletion of Gamma-aminobutyric Acid (GABA) a inhibitory neurotransmitter, consequently resulting in disinhibition of subthalamus and basal ganglia that translates into hyperkinetic movements. On the contrary, in Ketosis, GABA can be resynthesized using acetoacetate produced in the Liver to prevent its reduction, thus causing lesser incidence of movement disorder[1].

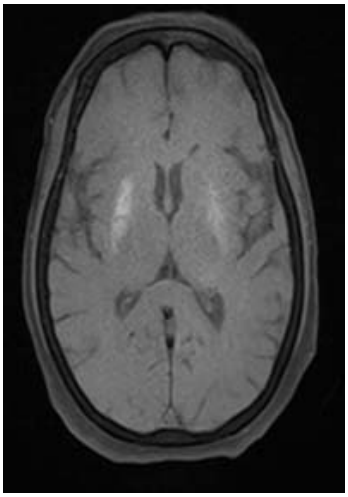

Fig 1 — MRI Brain in T1 weighted image shows Bilateral hyperintensities in putamen & caudate nucleus

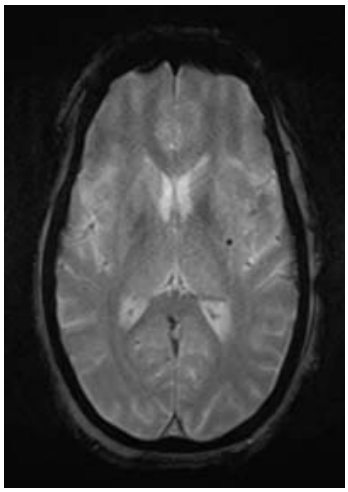

Fig 2 — MRI Brain in T2 weighted image shows Bilateral Hyperintensities in caudate nucleus and putamen

The movement disorders commonly associated with Hyperglycaemia can be Hemichorea-hemiballismus [HB-HC], Monoballismus, Myoclonus, Hemifacial spasm, paroxysmal kinesogenic dyskinesia (PKD) and several partial seizures[2,3]. Overall, these are usually more frequently observed in a background of non Ketotic hyperglycaemia ratherthan that with ketosis[1,2]. However, Paraballism-Parachorea that is ballismus and choreiform involvement of both lower limbs as a primary manifestation is rare, as is diaphragmatic Myoclonus.

Ballismus are large amplitude wild flinging incessant purposeless movements that are typically seen affecting one half of the body. The pathologic abnormality lies in the subthalamic nucleus and its afferent or efferent connections. Very rarely can they be Bilateral /involving both legs which is known as PARA BALLISMUS. As early as 1965 Hemiballismus was described as (1) Hemiballismus, typical (well localized lesion in the contralateral sub thalamic nucleus) (2) Hemiballismus, atypical (involving connections of the subthalamic nucleus, usually internal capsule) (3) Para-ballism (bilateral ballistic activity usually as a part of encephalitic sequelae with corpus luyii seemingly normal)[4].

Chorea is described as an involuntary irregular, random, non-rhythmic, purposeless movements, caused by involvement of the caudate nucleus. Variable in their distribution, they can affect a single extremity, one half of the body (hemichorea) or be generalized. Characteristically involving the distal extremities, it may also affect the proximal parts, lower extremities,trunk,face, tongue lips and Pharynx de Jong[5].

Diaphragmatic myoclonus also known as belly dancers' dyskinesia, is a form of segmental myoclonus caused by rhythmic, involuntary contractions of the diaphragm resulting in undulating, rhythmic movements of the abdomen. Its generator source is believed to lie in the rostral medulla. It can be due to central causes such as Encephalitis and extra pontine myelinosis. Peripheral causes include Phrenic nerve injury /irritation, Spinal cord lesions. They may also be drug induced or psychogenic, however, majority of the cases are idiopathic[6]. Diaphragmatic Myoclonus has been reported only once before as a reversible manifestation of uncontrolled hyperglycemic state[7,8]. Often mistaken for hiccups, it is a less recognized phenomenon,the identification of which is thus crucial for suspecting and treating underlying hyperglycaemia[9].

In this case we observed Para-ballismus and para-chorea.Bilateral chorea/ballismus has been reported twice before in a background of non Ketotic Hyperglycaemia.

References

1 Chua, CB., Sun, CK., Hsu, CW — Diabetic striatopathy: clinical presentations, controversy, pathogenesis, treatments, and outcomes. Sci Rep 10, 1594 (2020). https://doi.org/10.1038/s41598-020-58555-w

2 Awasthi D, Tiwari AK, Upadhyaya A, Singh B, Tomar GS — Ketotic hyperglycemia with movement disorder. *J Emerg Trauma Shock* 2012; **5(1):** 90-1. doi:10.4103/0974-2700.93095

3 Jagota P, Bhidayasiri R, Lang AE — Movement disorders in patients with diabetes mellitus. *J Neurol Sci* 2012; **314:** 5 11.

4 Carpenter MB — Pathologie des Ballismus. *Arch Neurol* 1965; **13(5):** 566. doi:10.1001/archneur.1965.00470050114017

5 Dejong'S the Neurologic Examination (2013) 7th Edition (English, Hardcover, Campbell W. W.)

6 Rathore C, Prakash S, Bhalodiya D. Belly dancer's dyskinesia: A rare movement disorder. *Neurol India* 2018; **66, Suppl S1:** 156-7

7 Dubey S, Chatterjee S, Mukherjee D, Ghosh R, Sengupta S, Lahiri D, Pandit A — "Dancing belly" in an old diabetic lady. *J Family Med Prim Care* 2020; **9:** 2580-2.

8 Bendi VS, Matta A, Torres-Russotto D — Bilateral chorea/ballismus: detection and management of a rare complication of non-ketotic hyperglycaemia. Case Reports 2018; 2018: bcr-2018-224856.

9 Milburn-McNulty P, Michael BD, Woodford HJ — Hyperosmolar non-ketotic hyperglycaemia: an important and reversible cause of acute bilateral ballismus. *Case Reports* 2012; 2012:bcr1120115084.