Bangladesh Journal of Medicine (BJM)
ISSN : 1023 - 1986
eISSN : 2408 - 8366

CASE REPORT

HYPERKINETIC MOVEMENT CAUSED BY DIABETIC STRIATOPATHY: A CASE REPORT

AJAY KUMAR AGARWALLA[1], AMINUR RAHMAN[2], SHARIF AHMED[1], MD ALAMGIR HOSSAIN[3], MAHBUBUL HAKIM MISHU[4], SHAHJADA MOHAMMAD DASTEGIR KHAN [5,] BIPLAB PAUL[5], PALLAB KANTI SAHA[5], MD DAHARUL ISLAM[6]

Abstract:

Hyperkinetic movement like chorea is rare in uncontrolled diabetic patients. Increased level of glucose affects the basal ganglia known as diabetic striatopathy. Synonym of this condition includes hemichorea-hemiballismus, chorea-hyperglycemia-hemiballismus syndrome, non-ketotic hyperglycemic hemichorea. We report a 65-year-old woman with uncontrolled diabetes who presented with unintentional ballistic movements of his right upper and lower extremities making her disabled. Magnetic resonance imaging of her brain demonstrated findings suggestive of diabetic striatopathy. Antipsychotics (eg. Haloperidol) helped to control her chorea-like movements. Strict glycemic control was advised to prevent recurrence.

Keywords: *Hyperglycemia, Striatopathy, Hemichorea- hemiballismus syndrome, Chorea-hyperglycaemia-basal ganglia syndrome*

Date of submission: 12.03.2025 *Date of acceptance: 19.04.2025*
DOI: https://doi.org/10.3329/bjm.v36i2.81137.

Citation:*Agarwalla AK, Rahman A, Ahmed S, Hossain MA, Mishu MH, Khan SMD. Hyperkinetic Movement Caused by Diabetic Striatopathy: A Case Report. Bangladesh J Medicine 2025; 36(2): 136-138.*

Introduction:

Diabetic Striatopathy (DS) is a rare cause of acute onset hemichorea-hemiballismus and it becomes a good differential when a patient is diabetic with uncontrolled blood sugar and presents with abnormal movement1. To establish a diagnosis of DS, other acute causes of movement disorder need to be excluded which include stroke, and Sydenham's chorea[2]. Basal ganglia lesions of different etiology can present in a similar way (3). Magnetic resonance imaging (MRI) is the key mode of investigation linked with the medical history for accurate diagnosis[2].

Case Report:

This 65-year-old right-handed diabetic woman presented with involuntary movement of her right upper and lower limbs for 5 days. Her past medical history was unremarkable except for uncontrolled diabetes for about 20 years and an ischemic stroke 10 years back. She was not on her regular follow-ups and was not compliant with her medication.

Her abnormal movements started insidiously but deteriorated rapidly. Her movement persisted when she was awake but dampened when sitting still in one place. Initiating movement of any sort made her

1. Assistant Registrar, Department of Neurology, Sir Salimullah Medical College Mitford Hospital, Dhaka-1100, Bangladesh.
2. Professor, Department of Neurology, Sir Salimullah Medical College, Dhaka- 1100, Bangladesh.
3. Registrar, Department of Neurology, Sir Salimullah Medical College Mitford Hospital, Dhaka- 1100, Bangladesh.
4. Resident, Department of Neurology, Sir Salimullah Medical College Mitford Hospital, Dhaka- 1100, Bangladesh.
5. Assistant Professor, Department of Neurology, Sir Salimullah Medical College, Dhaka- 1100, Bangladesh.
6. Professor, Department of Medicine, Sir Salimullah Medical College, Dhaka- 1100, Bangladesh.

Address of Correspondence: Prof. Dr. Aminur Rahman**,** Professor and Head, Dept. of Neurology, Sir Salimullah Medical College, Dhaka-1100, Bangladesh**,** E-mail: draminur@yahoo.com, https://orcid.org/0000-0001-9261-7734.

abnormal movement worse. Her relatives observed very little to no jerks in her limbs during her sleep. Her movements became violent very quickly and seriously affected her physically and mentally. She could not able to hold herself quiet at one place. It was also tiring due to exhaustion caused by the movements.

The findings of her physical examination showed hemiballismus localized to her right upper and lower extremities. Her blood pressure record showed 136/ 80 mm Hg, her pulse was regular at 80 beats/ min, and other vital parameters were unremarkable. A neurological examination revealed that she was alert, oriented, cooperative, and communicating well. Her cranial nerve functions were intact including normal fundoscopy examination and normal speech. Her gait was violent, not following any specific pattern and we were unable to perform a Romberg's text on her. Regarding her limb examinations, there was no pronator drift. Tone, power, and deep tendon reflexes were normal. The plantar reflex was bilaterally flexor. Her sensory findings were also unremarkable.

Her blood parameters revealed hyperglycemia of 15 mmol/L and corresponding urine sugar was 4+, HbA1c 13.3%, S. creatinine 1.4 mg/dl. Serum electrolytes were within normal limits. Her blood lipid profile revealed dyslipidemia (Cholesterol 180 mg/dl, LDL 80 mg/dl, TG 470 mg/dl). We have done brain imaging on her, which showed hyperintensity at the left lentiform nucleus in the T1 sequence (Figure:1). There was also a hypo-intensity at pons suggestive of an old infarct. There was no evidence of acute infarction, intracranial hemorrhage, parenchymal mass lesion, mass effect, midline shift, or sign of herniation.

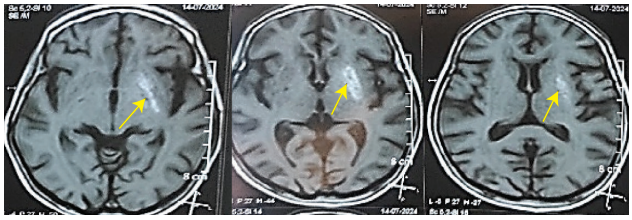

Fig.-1: *Sagittal T1 weighted magnetic resonance imaging of the brain*

Her violent movements were treated with haloperidol 2.5 mg twice daily and hyperglycemia was managed with basal-bolus insulin regimen. The patient responded very well to the mentioned therapy and dramatic improvement of symptoms was observed.

Discussion:

DS is a rare condition that was first coined a decade back for patients of abnormal movement in patients of hyperglycemia[1]. Because of the uncommon nature of the condition, there are only a few case reports and the nomenclature is not consistent in the articles due to a poor understanding of this condition[2,3]. Any lesion at the basal ganglia can produce similar symptoms, however, high blood glucose in the absence of an infarct or mass lesion makes the condition unique and labeled as DS. This condition is often known by other names like hyperglycemic nonketotic hemichorea/ hemiballismus, diabetic hemichorea/hemiballismus, and hyperglycemic basal ganglia syndrome[4]. Only a clinical picture is not sufficient to make this diagnosis[5].

On review of the available literature, we have found that most patients with hemichorea/ hemiballism had uncontrolled diabetes and their imaging showed evidence of contralateral basal ganglia involvement[6]. This condition is mostly treatable with typical or atypical antipsychotics combined with control of blood glucose.

A meta-analysis published in 2020 showed that putamen is the most involved striatal area (about 78.6%), and MRI is way more sensitive (95.33%) than CT (78.9%) for identifying the lesion[7]. This analysis also recognized the benefit of antipsychotic medication combined with glycemic control compared with those managed with glycemic control alone, showing a respond rate of 76.25 in earlier cohort compared with 25.6% in the later cohort[7,8].

There are few analyses done pathologically and radiologically as attempts to identify the mechanism behind this. Though definite pathogenesis has yet not been discovered, the proposed mechanism includes disruption of the blood-brain barrier, demyelination, mineral deposition, petechial hemorrhage, and microinfarction with astrocytosis[8].

DS is less common in non-Asian descent, making it highly underdiagnosed among them. This article could raise awareness among clinicians to think of DS as a differential whenever they encounter a patient with acute chorea/hemiballism-like movement disorder in a hyperglycemic patient. This will alert the clinicians to screen those subjects for DS changes on MRI. As this is a treatable condition and has a great impact on patients' quality of life, early diagnosis is important.

Conclusion:

One uncommon neurological symptom in DM patients is DS. Despite the paucity of treatment guidelines and the possibility of potentially life-threatening consequences, it is curable and has a favorable prognosis if treated appropriately.

Conflict of Interest:
The authors stated that there is no conflict of interest in this study

Funding:
This research received no external funding.

Consent:
For the purpose of publishing this case report and any related photos, the parents are written informed consent was acquired.

Acknowledgments:
The authors were grateful to the staffs of the Department of Neurology in Sir Salimullah Medical College Mitford Hospital, Bangladesh.

References:

1. Oh SH, Lee KY, Im JH, Lee MS. Chorea associated with non-ketotic hyperglycemia and hyperintensity basal ganglia lesion on T1-weighted brain MRI study: a meta-analysis of 53 cases including four present cases. Journal of the neurological sciences. 2002 Aug 15;200(1-2):57-62. https://doi.org/10.1016/S0022-510X(02)00133-8. PMid:12127677
2. Dubey S, Biswas P, Ghosh R, Chatterjee S, Kanti Ray B, Benito-León J. Neuroimaging of diabetic striatopathy: more questions than answers. European neurology. 2022 Sep 20;85(5):371-6. https://doi.org/10.1159/000524936. PMid:35717942
3. Das L, Pal R, Dutta P, Bhansali A. "Diabetic striatopathy" and ketoacidosis: report of two cases and review of literature. Diabetes research and clinical practice. 2017 Jun 1;128:1-5. https://doi.org/10.1016/j.diabres.2017.03.008. PMid:28431304
4. Mozzini C, Ghirardi R, Pagani M. Diabetic striatopathy: case report and possible new actors. Case Reports in Neurological Medicine. 2022;2022(1):4176419. https://doi.org/10.1155/2022/4176419. PMid:36578653 PMCid:PMC9792229
5. Arecco A, Ottaviani S, Boschetti M, Renzetti P, Marinelli L. Diabetic striatopathy: an updated overview of current knowledge and future perspectives. Journal of Endocrinological Investigation. 2024 Jan;47(1):1-5. https://doi.org/10.1007/s40618-023-02166-5. PMid:37578646 PMCid:PMC10776723
6. Lucassen EB, Delfyett WT, Stahl MC. Persistent hemichorea and caudate atrophy in untreated diabetic striatopathy: a case report. Case reports in neurology. 2018 Jul 9;9(3):299-303. https://doi.org/10.1159/000484201. PMid:29422853 PMCid:PMC5803691
7. Xu Y, Shi Q, Yue Y, Yan C. Clinical and imaging features of diabetic striatopathy: report of 6 cases and literature review. Neurological Sciences. 2022 Oct;43(10):6067-77. https://doi.org/10.1007/s10072-022-06342-y. PMid:35965280 PMCid:PMC9376124
8. Lin YT, Chen SC, Yip PK, Wang V. Magnetic resonance imaging volumetric analysis for diabetic striatopathy with two episodes of hemichorea-hemiballism syndrome: a case report. Medicine. 2019 Sep 1;98(38):e17249. https://doi.org/10.1097/MD.0000000000017249. PMid:31567992 PMCid:PMC6756614