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Video Vignette

Involuntary Movements During Treatment for Hyperglycemia

Yuya Ando, MD, Masato Kadoya, MD, PhD *, Tsutomu Kodera, MD, PhD

Department of Internal Medicine, Japan Self-Defense Forces Central Hospital, Tokyo, Japan

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Case Presentation

An 89-year-old woman with type 2 diabetes mellitus presented with occasional drowsiness. She had lost >5 kg within 1 week, and her family informed that she had become increasingly less responsive. She was not engaging in excessive soft drink consumption or taking laxative medication. However, she had started taking prednisone 10 mg/day for pruritus 1 week prior to presentation. At the initial assessment, she was fully conscious without any abnormal vital signs. However, a glucometer showed >600 mg/dL. A subsequent blood test revealed a glycosylated hemoglobin level of 11.8% and plasma osmolarity of >300 mmol/L. Blood gas analysis showed slight metabolic acidosis with ketones in her urine. The patient was admitted to the hospital with a diagnosis of hyperosmolar hyperglycemic syndrome. Subcutaneous insulin injections were administered to control the blood glucose level, which was successfully tapered within 2 days. Despite further improvements in blood characteristics on day 3, she started to experience the following involuntary facial movements: (1) closing of the eyes and (2) clamping of the mouth (Fig. *A*, photo). On day 4, her extremities also showed involuntary movement (Fig. *B*, photo): gross movements in the right upper and lower limbs. She remained fully conscious even during such movements. These involuntary movements were recorded (photo) after obtaining informed consent from the patient and her family.

What is the diagnosis?

Answer

A neurologic examination revealed normal muscle tone and strength and no abnormal pyramidal tract signs. Additionally, her cranial nerves and sensation were preserved. Laboratory test results showed unremarkable findings except for a slightly elevated blood glucose level. Neither cranial magnetic resonance imaging (MRI) nor nonenhanced computed tomography (CT) revealed any pathogenic abnormalities in the cerebrum, including the basal ganglia. Electroencephalography findings were also normal. Therefore, although the symptoms occurred during the off-peak period and in the absence of any CT or MRI abnormalities, the patient was diagnosed with diabetic striatopathy (DS). Her involuntary movements gradually disappeared over a few weeks with haloperidol medication (up to 3.75 mg/day), and she was discharged. DS is a hyperglycemic condition associated with chorea/ballism that has been found to occur predominantly in older woman with poorly controlled type 2 diabetes mellitus.[1] Although the majority of patients with DS show striatal hyperdensity on CT or hyperintensity on T1-weighted MRI, a small number of patients develop chorea without evidence of striatal involvement in imaging data.[1] The definition and classification of DS are still controversial because of the rarity and low visibility of this condition. Here consistent with the classification system proposed by Dubey et al,[2] we defined our case as “clinically isolated DS” because of the absence of any radiological abnormalities. Previous studies have indicated a probable pathogenesis of microvascular hemorrhage and suggested that chorea/ballism can occur despite successful glycemic control via the same process by which diabetic retinopathy can worsen following intensive control of the blood glucose level.[1,3] Haloperidol is the most commonly used medication for the treatment of chorea[1] and was effective in our case. DS should be recognized as a clinically important complication in individuals with hyperglycemia, although further studies are needed to elucidate the underlying pathogenesis.

Abbreviations: CT, computed tomography; DS, diabetic striatopathy; MRI, magnetic resonance imaging.

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* Address correspondence to Dr Masato Kadoya, Department of Internal Medicine, Japan Self-Defense Forces Central Hospital, 1-2-24 Ikejiri, Setagayaku, Tokyo, Japan.
E-mail address: kadoya-masato@hotmail.co.jp (M. Kadoya).

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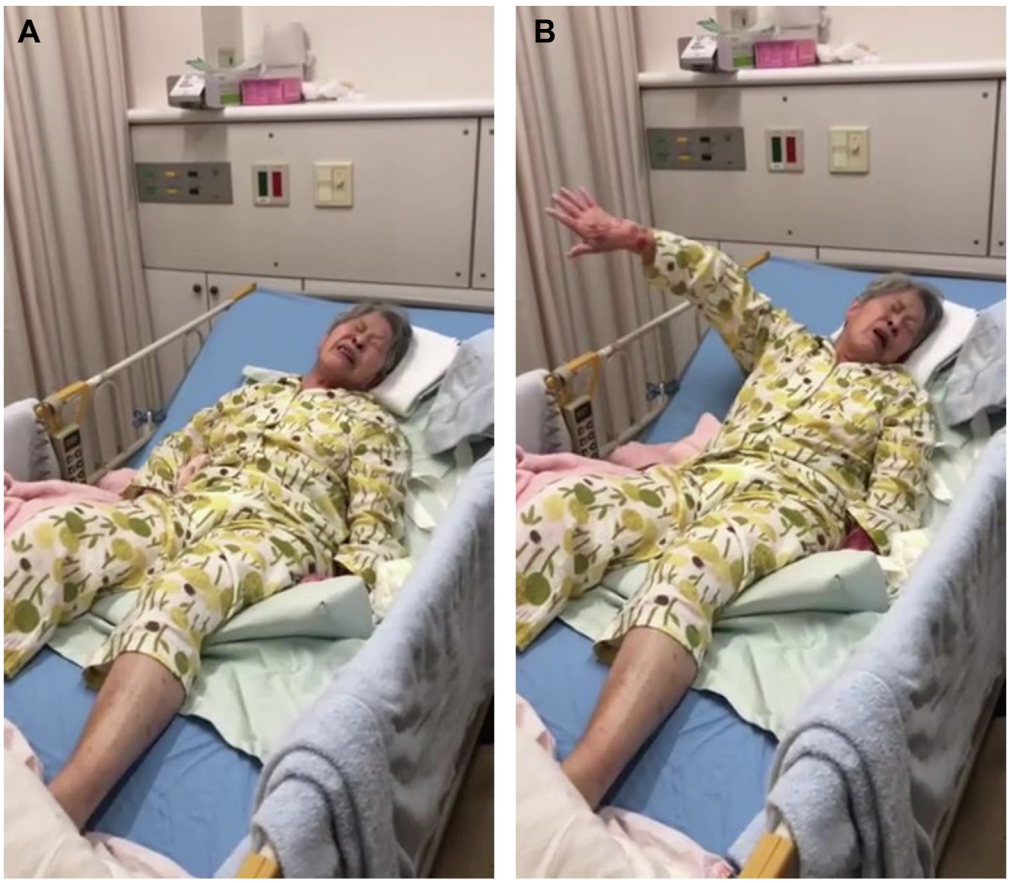

Fig.

Disclosure

The authors have no multiplicity of interest to disclose.

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References

1. Chua CB, Sun CK, Hsu CW, Tai YC, Liang CY, Tsai IT. "Diabetic striatopathy": clinical presentations, controversy, pathogenesis, treatments, and outcomes. *Sci Rep*. 2020;10(1):1594.
2. Dubey S, Biswas P, Ghosh R, Chatterjee S, Kanti Ray B, Benito-Leon J. Neuroimaging of diabetic striatopathy: more questions than answers. *Eur Neurol*. 2022;85(5):371–376.
3. Abe Y, Yamamoto T, Soeda T, et al. Diabetic striatal disease: clinical presentation, neuroimaging, and pathology. *Intern Med*. 2009;48(13):1135–1141.