CASE PRESENTATION

Diabetes is associated with an increased risk of late complications in the central nervous system. Certain neurological symptoms may also occur acutely in hyperglycemia. We describe here a case of hyperosmolar non-ketotic syndrome and reversible hemichoreic dyskinesia with morphological changes in the basal ganglia of the CNS.

An 85-year-old woman was admitted to the emergency room with a 2-week history of blurred vision, dizziness, dysarthria, polyuria, and polydipsia. Eleven years earlier, she had suffered a stroke and had a moderately impaired renal function. Her non-fasting P-glucose was 57.5 mmol/L (1035 mg/dL), P-sodium 120 mmol/L, P-potassium 5.5 mmol/L, and P-urea 13.5 mmol/L. P-osmolality was calculated to 311 mmol/L (reference 285-295 mmol/L). Arterial blood gas analysis and B-ketones were normal, and B-HbA1c was 136 mmol/mol (14.6% DCCT).

The patient was admitted to the intensive care unit with a diagnosis of hyperosmolar non-ketotic syndrome (HNKS). A computerized tomography (CT) scan of the brain was done due to facial asymmetry and showed increased attenuation in the putamen and in caput nuclei caudati (Figure 1A). On day seven, another CT scan was done due to uncoordinated movements in the patient's left extremities and revealed slightly more attenuation in the previously described areas (Figure 1B).

Six weeks later, the patient again sought emergency care due to more intense involuntary movements. Interestingly, the CT scan showed complete resolution of the previously described abnormalities (not shown) but magnetic resonance imaging (MRI) showed a high signal in the right putamen on T1-weighted images (Figure 1C). The patient was given haloperidol for symptom relief. Two months later, her symptoms disappeared. Six months later, the MRI was normalized.
2 | DISCUSSION

The case illustrates an unusual phenomenon, hemichorea/hemiballismus associated with hyperglycemia as first described by Bedwell.\(^1\) The symptoms consist of involuntary, unilateral movements in connection with, or shortly after, hyperglycemia. They occur throughout the waking time and cease during sleep. Typical imaging findings are unilateral high-attenuating areas in basal ganglia on CT scans or increased signal on T1-weighted images in MRI.\(^2\)

Onset of symptoms both in connection with the acute hyperglycemia but also later after initiating glucose-correcting treatment has been described. A 15-years survey of all chorea/ballismus patients at the Mayo clinic showed that hyperglycemia as the underlying cause was noted in only 1%, indicating the rarity of the condition.\(^3\)

The causes behind the condition are currently not clear and there are several theories regarding the underlying pathophysiological mechanisms. One theory suggests that hyperglycemia causes the neurotransmitter GABA (gamma aminobutyric acid) to be consumed as an energy source in the affected areas and thus reduces the ability of inhibitory GABA signaling in the thalamus, which would then lead to involuntary movements on the contralateral side.\(^4\)

Findings of morphological changes of the caudate and putamen are not uncommon and may have several causes.\(^5\) It is thus very difficult to conclusively prove a cause-and-effect relationship between the hyperglycemia and the symptoms and morphological changes in our patient. However, the chain of events over time, from the appearance of morphological changes and resulting symptoms upon onset of hyperglycemia to the symptoms and morphological changes being resolved upon restoration of euglycemia, is suggestive of such an effect.

This case reports an unusual neurological complication of severe hyperglycemia. The normalization of the neurological symptomatology with the return to euglycemic conditions supports the reversible, essentially metabolic, cause of the symptomatology although a precise mechanism of disease remains unknown.

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Å.S. is the guarantor of this work and, as such, had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

CONFLICT OF INTEREST
No potential conflicts of interest relevant to this article were reported.

AUTHOR CONTRIBUTIONS
AKK: was involved in the care and management of the patient, and all authors took part in the writing and reviewing of the article. Written consent for publication was obtained from the patient.

ETHICAL APPROVAL
The patient gave informed consent to this publication.

DATA AVAILABILITY STATEMENT
Data are available in the patient's medical record.

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**Figure 1** A, Brain CT scan 2 d after admission for hyperglycemia shows slightly increased attenuation in the basal ganglia on the right side. The patient had no clinically noticeable dyskinesia at the time. B, Brain CT scan 6 d after admission for hyperglycemia shows pronounced attenuation in the basal ganglia on the right side. The patient had at the time started to show symptoms of left-sided involuntary movements. C, Brain MRI scan in T1 sequence just short of 2 mo after the original hyperglycemia episode shows, despite moderate movement artifacts, increased signal in the previously described areas of the basal ganglia on the right side.
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