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Case Report

# Proprioceptive-induced seizure in diabetic non-ketotic state: A video case report

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ABSTRACT

A 63-year-old diabetic gentleman with proprioceptive-induced seizures is reported. Marked hyperglycemia due to discontinuation of anti-diabetic medication was the precipitating cause. Distinctive clinical feature of this case was recurrent focal motor seizures involving the left upper limb precipitated by volitional movements of the same limb and self-aborted by holding the affected limb with the contralateral limb. This was accompanied by left brachial weakness without corresponding lesions on cranial MRI. Despite rapid achievement of euglycemia, seizures persisted and required anticonvulsant polytherapy for effective control.

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## 1. Introduction

Abnormal glycemic status—both hyper and hypoglycemia predispose to seizures. Hyperglycemia predisposes to seizures by causing neuronal hyperexcitability.[1] Recurrent tonic focal motor seizures can be the first manifestation of non-ketotic hyperglycemia (NKH) of diabetes.[2] We report a patient with proprioceptive-induced seizures related to marked hyperglycemia arising from discontinuation of diabetes therapy.

## 2. Case study

A 63-year-old-gentleman presented with recurrent involuntary movements confined to left upper limb since 4 days. These movements were precipitated by attempted volitional activity of left upper limb, lasting 1–2 minutes, without loss of consciousness, followed by spontaneous recovery. There was accompanying weakness of left upper limb and unsteadiness upon walking. He was a known diabetic since 30 years and

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hypertensive since 3 years. Three months prior to this presentation he had discontinued anti-hypertensives and anti-diabetics and had switched to native treatment.

Clinical examination showed a blood pressure of 170/90 mm Hg. Neurologically he was conscious, alert and oriented. Language functions, speech and cranial nerves were normal. Optic fundi revealed diabetic retinopathy. Motor tone was normal. He had left pronator drift with mild left brachial weakness (Medical Research Council grade 4+/5). Volitional left arm movements were followed by tonic posturing of left hand followed by clonic twitching of the fingers which subsequently spread to the whole arm. Towards the end of the attacks possible atonic component involving left shoulder muscles was noted. He was fully alert and responsive during the attack. These attacks could be self-aborted by pulling the left index finger with the right hand for few seconds (see video attachment). These attacks were stereotypical and brief, lasting for a minute. Sensory examination was normal. Stretch reflexes were normal and plantar responses were flexors bilaterally.

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Serum chemistry revealed elevated blood glucose (770 mg%) and renal parameters (urea 57 mg% and serum creatinine 2.9 mg%). Serum electrolytes showed hyponatremia (sodium 122 m eq/l) with otherwise normal electrolyte profile. Glycosylated hemoglobin level was 11.7%. Urinalysis revealed albuminuria (2+) and glycosuria (3+) but no ketones. Arterial blood gas did not reveal metabolic acidosis. Cerebrospinal fluid analysis and non-contrast MRI of the brain (T1, T2, weighted images, FLAIR and diffusion-weighted images) revealed no abnormalities. Abdominal ultrasonogram revealed bilateral grade I increase in cortical echoes of the renal parenchyma. Ictal EEG was recorded by asking the patient to induce the seizure with volitional motor activity of left upper limb. High voltage sharp wave discharges were recorded over the contralateral hemisphere having central emphasis during the clinical attack. In between the attacks, EEG showed no evidence of on-going encephalopathy.

Intravenous insulin therapy and nebivolol (2.5 mg/day) were instituted. Sodium valproate (600 mg/day, later increased to 1000 mg/day) and levetiracetam (1000 mg/day) were administered for seizures. With physiotherapy measures limb weakness improved over a week. Despite rapid control of diabetes within 24 h, seizures persisted. Clobazam (10 mg/day) was added. No recurrences of seizures were noted after 12 days and he maintained improvement thereafter.

## 3. Discussion

Neurologic abnormalities well known with NKH are focal seizures and hemichorea. Our patient had severe hyperglycemia owing to recent discontinuation of diabetes therapy and had developed renal dysfunction and hyponatremia. While hyponatremia and uremia have been associated with seizures, these are usually generalized tonic-clonic in nature and such seizures are usually accompanied by alteration in sensorium. Our patient had only focal seizures and did not have features of uremic or hyponatremic encephalopathy. Thus the seizures were attributed to the hyperglycemic state per se.

Sabitha and colleagues analyzed seizures in hyperglycemic patients and observed that long duration and frequent focal motor seizures with or without secondary generalization occur in NKH, sometimes as the presenting manifestation of diabetes. They observed that complex partial seizures, visual hallucinations and epilepsia partialis continua were common and movement induced seizures were a rarity.[3] Our patient presented with proprioceptive-induced seizures. Interestingly, such seizures have been described more than 100 years ago by Gowers in 1901.[4] Differential diagnosis for such seizures is paroxysmal kinesogenic dyskinesia where involuntary movements could be precipitated by movements. However characteristic movement disorders like dystonia and choreoathetosis and normal EEG are distinctive of paroxysmal kinesogenic dyskinesia. Such features serve to distinguish it from proprioceptive-induced seizures where the movements are tonic or tonic-clonic and are often accompanied by epileptiform discharges in the EEG. Accompanying the focal seizures in our patient were the distinctive sharp wave abnormalities arising from the contralateral hemisphere.

While the anatomical substrate for chorea in hyperglycemic nonketotic patients is thought to be the contralateral striatum as has been observed radiologically on MRI and functional imaging studies, that which underlies the seizures is only poorly understood. Subcortical T2 hypointensity rather than hyperintensity with contrast enhancement has been seen in patients with seizures associated with non-ketotic hyperglycemia.[5] Our patient had no corresponding lesions on the non-contrast MRI and renal dysfunction precluded contrast administration.

Proprioceptive-induced seizures are reflex seizures commonly triggered by active or passive movements. These consist of mainly simple partial seizures of tonic or clonic semiology. Occasionally, these occur in patients with structural brain lesions resulting in motor deficits. These are considered to represent "system epilepsies" which involve a single sensory-motor network. Some argue that this form of seizures is due to epileptically-enhanced stretch reflex.[6] Classically, rolandic sensory-motor cortex or supplementary motor area is involved in the epileptic network. Hence, the provoking stimulus and the seizure are confined to a single functional cerebral network. In our patient, the posture and contraction of the left hand was a possible trigger which resulted in increased stretch reflex afferent impulses to the hyperexcitable contralateral sensory-motor cortex which resulted in the focal motor seizure, and, towards the end of the seizure, due to physiological increase in the inhibitory cortical mechanism, the seizure was interrupted by atonic component. Our patient could abort the seizure on several occasions by pulling the left index finger. We feel this was due to the modification of the central hyperexcitable circuit by stretching of the finger (opposite action to the initial contraction of the long flexors of the hand) rather than peripheral mechanism due to Golgi tendon receptor induced reflex inhibition as these receptors are stimulated by excessive contraction induced muscle tone rather than stretching the muscles. Gabor showed that these seizures are not prevented by brachial plexus block and thus are independent of proprioceptive afferents.[7] Our case proves the importance of afferent sensory inputs as pulling the left finger stops the

seizures. This is consistent with the work of Chavel and Lamarche.[8] Thus, this form of reflex focal motor epilepsy has possible subtypes and may not represent a single entity.

Agents commonly used in reflex seizures are valproic acid and levetiracetam while lamotrigene and topiramate are alternatives. Valproic acid–levetiracetam combination was initially used in our patient with inadequate clinical response warranting addition of clobazam. Seizures recurred even after normalization of glycemic status suggesting that lowering of seizure threshold due to hyperglycemia persists even after correction of the hyperglycemia.

## Conflicts of interest

All authors have none to declare.

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