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# Transient Opsoclonus in A Patient with Hyperosmolar Non-Ketotic Coma

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**Abstract**

Opsoclonus is a rare movement disorder manifested by quick, involuntary, and jerky eye movements. On the other hand, hyperosmolar nonketotic coma (HONK) can present with a range of movement disorders, including focal seizures, myoclonus, and opsoclonus. Despite opsoclonus being commonly linked to neurological conditions, its association with metabolic abnormalities, particularly hyperglycemia, is rarely acknowledged. Here, we report an interesting case of a 60-year-old man who presented with transient opsoclonus associated with hyperosmolar non-ketotic coma. This case highlights the significance of taking metabolic factors into account when evaluating and managing opsoclonus. However, additional research is required to explain the underlying mechanisms and optimal management strategies for this uncommon association.

**Keywords:** ocular motility disorder; hyperglycemic hyperosmolar non ketotic coma; middle aged; coma

## Introduction

Opsoclonus is a rare neurological disorder characterized by rapid, involuntary, and chaotic eye movements called saccades. It can occur in isolation or as part of a broader syndrome named opsoclonus-myoclonus. Dr. Digre [1] investigates opsoclonus, an uncommon condition affecting eye movements marked by erratic, unpredictable saccades in multiple directions. In her study, she reviews 55 documented instances of opsoclonus in adults and adds three additional cases she personally observed. The instances can be classified into three primary categories. The most substantial group includes patients with suspected viral encephalitis, usually younger individuals who often experience complete recovery without enduring consequences. The second group encompasses patients whose opsoclonus is linked to a paraneoplastic syndrome. These individuals typically belong to an older age group and tend to have a less optimistic prognosis. The last group, characterized by various other disorders, consists of individuals with diverse presumed causes such as stroke and trauma. Other contexts [2] in which opsoclonus manifests in adults is due to head trauma or drug intoxication like amitriptyline. Moreover, documented cases [3] of opsoclonus following vascular events are primarily associated with motor deficits. The exact pathological mechanism of this syndrome remains a subject of debate, and while some authors have implicated the cerebellum and upper brainstem, the precise anatomical basis remains unclear. To our knowledge this is the third reported case of transient opsoclonus in patients with hyperosmolar non ketotic coma. This particular case stands out because the syndrome rapidly improved once the blood sugars and metabolic acidosis were addressed through therapy. We hypothesize that the significant metabolic dysfunction of neurons in the critical area alone can lead to the syndrome.

## Case Presentation

A 60-year-old man presented to the Emergency Department with fever and vomiting for eight days. The fever was sudden in onset, undocumented, intermittent in character, and not associated with rigors or chills; meanwhile vomiting occurred four to five times per day, was non-projectile and non-mucoid but contained food particles. One week later, his symptoms were followed by epigastric pain, irritability, and altered level of consciousness. His past medical history was significant for hypertension and hepatitis C which was treated five years back. He was currently taking Amlodipine and NSAIDs. On admission, the vitals were: Temperature afebrile, blood pressure 173/95 mmHg, heart rate 125 beats/minute. The patient's oxygen saturation was initially 93% on room air, necessitating the administration of 12 liters/minute of oxygen following which his saturation improved to 98%. His random blood sugar was 350 mg/dl. Upon

 

examination, he was extremely lethargic and tachypneic and followed single-step commands. There was no facial asymmetry. On further assessment there were multidirectional, conjugate, rapid, involuntary eye movements or jerks of both eyes. The movements occurred in the horizontal, vertical, and oblique directions, at times with a rotating component. Full ductions and versions were present. Pupils were 3 mm and reactive to light, bilaterally. There were no involuntary movements or jerking observed in limbs which might suggest presence of myoclonus. Motor examination revealed normal bulk, tone with power of 5/5 in all four limbs, reflexes were diminished, plantars were flexor. Sensory and cerebellar examination was limited due to his condition. Chest examination showed bilateral basal crepts. No heart murmur was audible.

Laboratory workup revealed hemoglobin 12.6g/dL, total leukocyte count (TLC) 8×109/L, platelets 265×109/L, urea 150 mg/dL, creatinine 4.5mg/dL, creatinine clearance 22, serum osmolality 458 mOsm/liter. Liver function tests showed ALP 256 IU/L, GGT 211 U/L, and SGPT 36 U/L. ABGs showed a pH 7.40, pCO2 25 mmHg, HCO3 16.1 mEq/L, pO2 108mmHg after management which became pH 7.32, pCO2 34.8 mmHg, HCO3 18.1 mEq/L and pO2 120mmHg. Anion gap was normal. Urinalysis showed glucose of 1000 mg/dl, no ketones with numerous pus cells. A CT scan head was done which was unremarkable. Echocardiogram was normal. Ultrasound abdomen revealed an echogenic liver and kidneys, and spleen size 13.9cm suggest chronic liver and kidney disease.

He was managed on lines of hyperosmolar non-ketotic coma, septic encephalopathy due to urinary tract infection and deranged renal parameters due to dehydration. By the next morning after appropriate medical therapy, intensive fluid replacement and vigilant control of blood sugars on insulin infusion, the patient was alert and responsive. His ophthalmological examination became normal.

## Discussion

Different kinds of myoclonus are being documented recently in the context of hyperosmolar hyperglycemic syndrome. While chorea and ballism are well-recognized acute potentially reversible movement abnormalities as the presenting manifestation of non-ketotic hyperglycemic episodes, the generated hyperkinesias typically completely resolve with early diagnosis and appropriate treatment of the metabolic disturbances of hyperglycemic hyperosmolar condition. The patients may experience various movement problems including blepharospasm, oromandibular dystonias, and hemifacial spasm along with myoclonus however, opsoclonus hasn't been frequently reported [4]. The early neuronal effects of fluid and electrolyte imbalances bring about functional changes rather than structural ones which poses a positive patient outcome on prompt correction of fluid and electrolyte disturbances. There is a correlation between a major symptom complex and abnormalities in fluid and electrolytes that facilitates early diagnosis and treatment [5]. The resolution of opsoclonus following correction of sugar levels and fluid replacement in our case provides evidence supporting Hyperosmolar Hyperglycemic Nonketotic Syndrome (HONK) as the underlying cause of this movement disorder which has not been frequently reported in medical literature.

In addition to our case, very few studies have reported metabolic disturbances as a significant contributing factor. One such case6 described opsoclonus accompanied by irregular myoclonic jerks in the face and extremities, and altered level of consciousness, which resolved following normalization of the patient's sugar levels. Another case7 in literature was documented wherein the patient regained consciousness, and both opsoclonus and myoclonus subsided when the plasma glucose concentration was reduced from 1400 mg/dl to 500 mg/dl. Our case lacks the myoclonus component commonly seen with opsoclonus. His neuroimaging was normal at the time of admission which ruled out the structural brain etiology related with opsoclonus. We started managing the acute concerning clinical issues in his case and which simultaneously improved his consciousness and opsoclonus. Therefore, we concluded that although many mechanisms may be responsible for opsoclonus as discussed above, HONK seems most likely in our case.

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