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Case Report

CHANTER Syndrome and mesenteric ischemia presenting concurrently, a case report and literature review ☆

Cagri Yurtsever, MD, Jessica Harris, MD, Alex Breeser, MD, Sudha Ramachandran, MD, Leszek Pisinski, MD*

Department of Radiology, Columbia University College of Physicians and Surgeons, Harlem Hospital Center, New York, USA

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ABSTRACT

We present a case of a 70-year-old male who was brought to the hospital with altered mental status and was found to have 2 serious complications of cocaine use which are Cerebellar Hippocampal and Basal Nuclei Transient Edema with Restricted diffusion (CHANTER) syndrome and mesenteric ischemia. CHANTER syndrome is a recently described constellation of radiologic and clinical findings and has a strong association with opiates, and/or other drugs of abuse, including cocaine. Even though CHANTER has many similarities with other ischemic, anoxic, and/or toxic injuries related to substance abuse such as clinical presentation and restricted diffusion on magnetic resonance imaging (MRI); the typical distribution of affected regions in the brain is helpful in differentiating from other injuries. With this study, we aim to emphasize the clues that separate CHANTER syndrome from other acute neurologic problems in the setting of substance use. Our case also suggests that the obstructive hydrocephalus, a known possible complication of CHANTER, is likely seen in the cases with severe and central cerebellar involvement. Additionally, it is not common to see complications in 2 different systems concurrently and a multisystemic approach is crucial to a patient with cocaine use to prevent missed life-threatening consequences throughout the various body systems.

Introduction

Cocaine is a powerfully addictive stimulant and one of the most frequently used recreational drugs. According to the annual World Drug Report, 4% of the global population between ages 15 and 64 years have been suffering from cocaine abuse [1]. Besides the significant number of deaths, cocaine use disorder has been found to be associated with 2.6 million disability-adjusted life years [2]. It leads to a variety

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* Corresponding author.
E-mail addresses: yurtsevc@nychhc.org, yurtsevercagri@gmail.com (C. Yurtsever).

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of chronic manifestations such as loss of smell, nosebleeds, cough, asthma, malnutrition, higher risk for infections including HIV and Hepatitis C. Moreover, exposed individuals may present to the emergency rooms with acute cardiovascular symptoms, acute neurologic symptoms, and/or acute abdominal pain [3–5].

The physiopathology of cocaine on different organs is substantially similar. It inhibits noradrenaline reuptake in the synaptic cleft of sympathetic neurons. It has also been shown to facilitate the discharge of central and peripheral catecholamines which induce thrombus formation through activating and increasing the number of circulating platelets. Another effect of cocaine is to stimulate vasospasm by increasing the level of extracellular dopamine [4–6]. Regardless of the underlying mechanism, the risk of brain ischemia may increase up to 7 times more in cocaine users as compared to the normal population [7].

Although there have been known acute neurologic injuries associated with cocaine and/or other substance abuse, such as acute ischemic stroke, Hypoxic-Ischemic Encephalopathy (HIE), Heroin-associated spongiform leukoencephalopathy (HASL), CHANTER was described recently as a radiologic and clinical syndrome with typical imaging findings [8]. As clinical presentations can overlap, it is crucial to differentiate CHANTER from other substance-use-related acute neurologic injuries, given its distinct clinical course, potential complications, and management, while also acknowledging the heightened risk of obstructive hydrocephalus because of cerebellar edema [8–10]. CHANTER is characterized by bilateral symmetric abnormal restricted diffusion areas involving basal ganglia, hippocampus, and cerebellar hemispheres. The differential diagnosis of acute neurologic disturbances in the setting of substance use is discussed in detail in the discussion.

Cocaine use-related bowel ischemia may have a benign-appearing physical exam and subtle radiological findings and may not be detected at the presentation. It can cause a variety of clinical conditions ranging from less severe non-obstructive mesenteric ischemia (NOMI) to bowel necrosis and even death [11].

Case report

A 70-year-old male with a past medical history of hypertension, diabetes mellitus, hyperlipidemia, and substance use was brought in by emergency medical services after being found unresponsive on the street. At the initial physical exam, the patient was lethargic, oriented to self only, and unable to give adequate history. Additionally, he was found to be hypoxic. There was left lower quadrant tenderness on the abdominal exam. Lab studies revealed acute kidney injury (GFR: 17 mL/min), hyperkalemia (potassium: 6.4 mmol/L) and high anion gap metabolic acidosis (PH Venous: 7.11, anion gap: 19 mEq/L and lactate venous: 3.7 mmol/L). The urine drug screen test was positive for cocaine.

Computed tomography of the abdomen and pelvis (CTAP) was performed and revealed long segment concentric wall thickening of ileal loops along with mild surrounding mesenteric fat stranding and suspicious air densities within the bowel wall (Figs. 1A and B). Findings were suspicious for mesenteric ischemia given the history of cocaine use. CT angiogram of abdomen and pelvis showed negligible stenosis of superior mesenteric artery and there was no thrombosis in superior mesenteric vein. CT Head was positive for hypodensities involving bilateral basal ganglia and cerebellar hemispheres which was likely secondary to cytotoxic edema. (Figs. 2A and B). MRI of the brain revealed areas of restricted diffusion in the bilateral globus pallidus, bilateral hippocampi and bilateral peripheral cerebellar hemispheres. Figs. 3A–C respectively). MR angiography (MRA) revealed no significant stenosis neither in the carotid system nor in the circle of Willis arteries. This constellation of findings was found to be the most consistent with CHANTER syndrome because of the typical distribution of restricted diffusion areas and history of opiate use. There was no evidence of obstructive hydrocephalus in our case. We believe that it is because of mild and peripheral involvement of cerebellar hemispheres in contrast to prior published cases. (Fig. 3A). Follow-up CT also did not reveal obstructive hydrocephalus (Fig. 4).

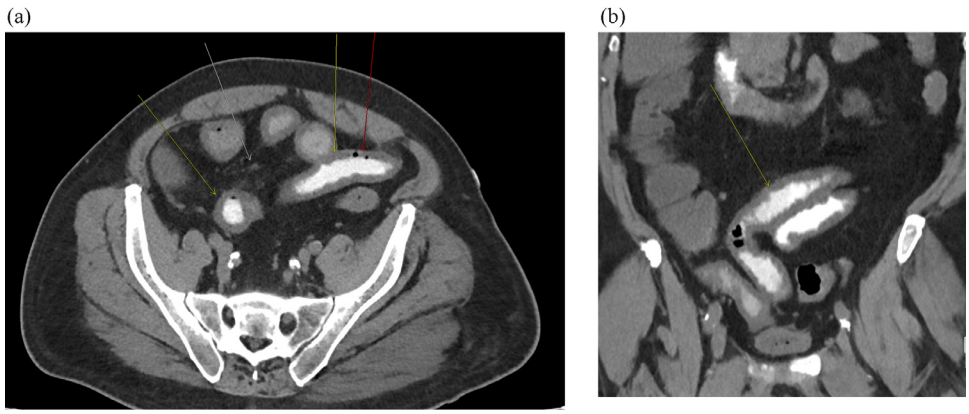

Fig. 1 – Axial (A) and coronal (B) CT abdomen and pelvis revealed long segment concentric wall thickening of ileal loops (yellow arrows) with mild surrounding mesenteric fat stranding (white arrow) and suspicious air densities within the bowel wall (red arrow).

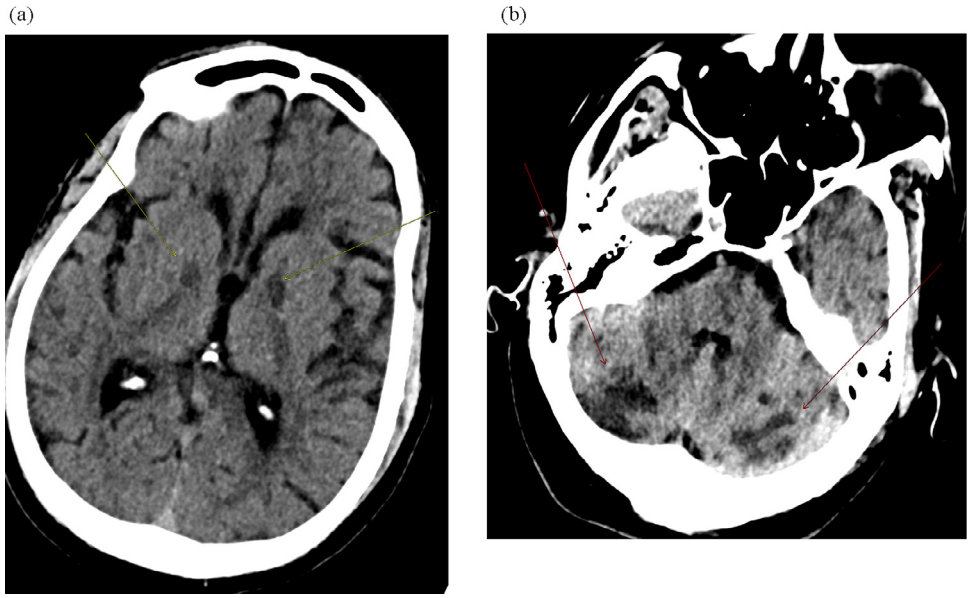

Fig. 2 – CT Head was positive for hypodensities symmetrically involving bilateral basal ganglia (A; yellow arrows) and cerebellar hemispheres (B; red arrows).

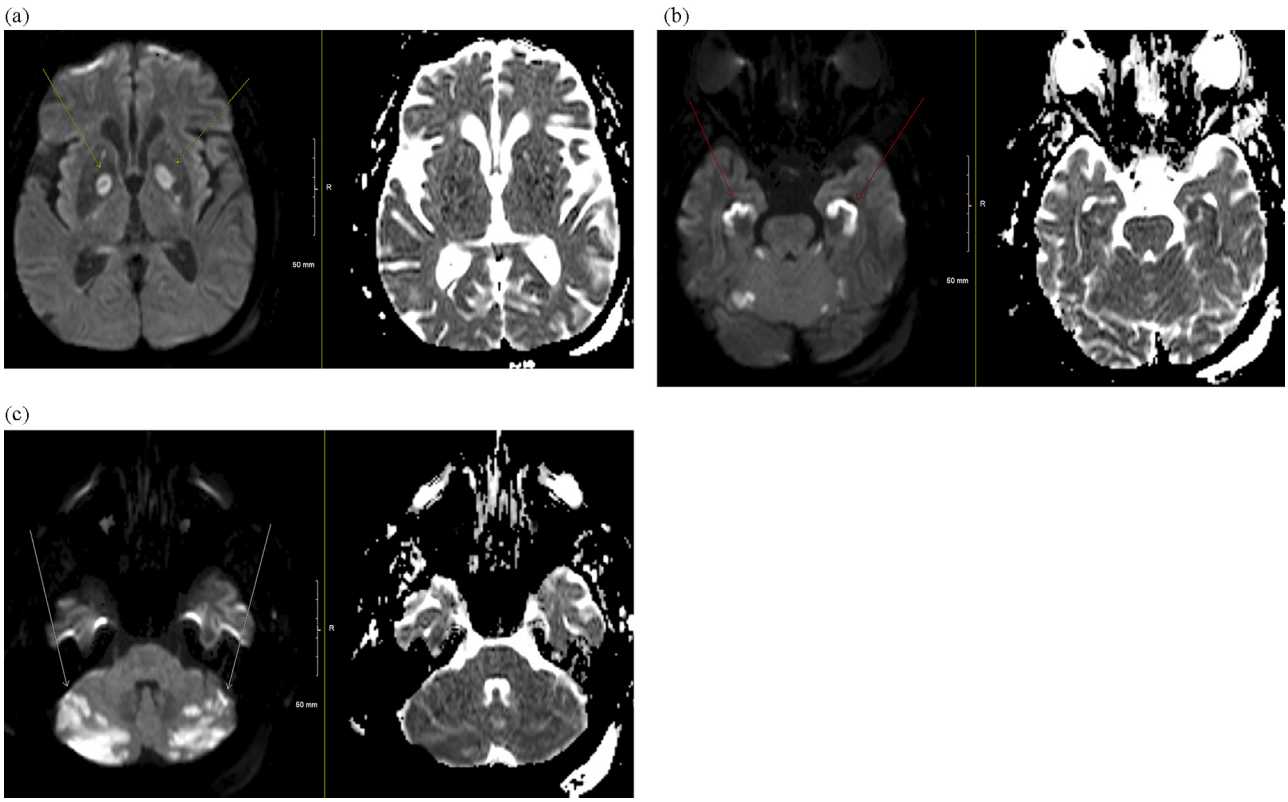

Fig. 3 – MR of brain revealed areas of restricted diffusion in the bilateral globus-pallidus (A; yellow arrows), bilateral hippocampal gyri (B; red arrows), bilateral cerebellar hemispheres (C; white arrows).

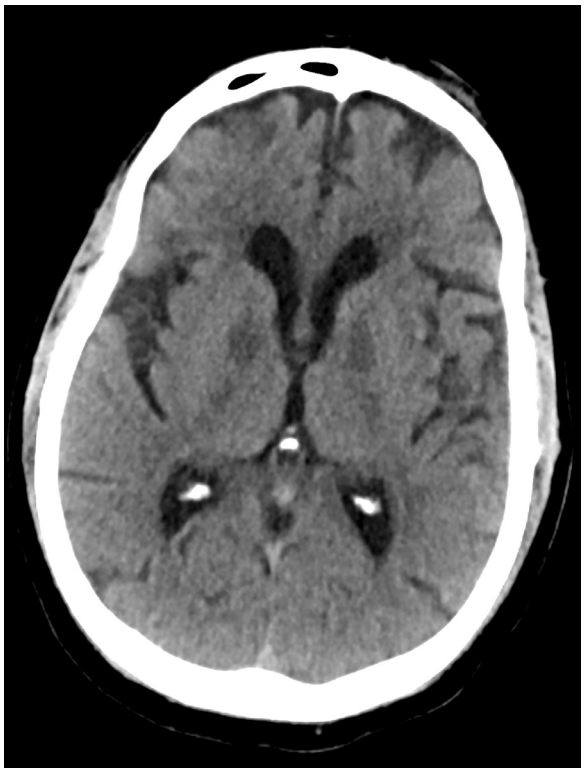

Fig. 4 – Follow-up CT Head did not show evidence of hydrocephalus.

The patient was admitted to the intensive care unit and cared for in the intensive care unit and neurology floor. The patient's mental status improved, was alert and able to move all extremities on the discharge day, and was sent to the subacute rehabilitation center for further treatment. Nonobstructive mesenteric ischemia was managed conservatively, resulting in improved abdominal pain the following day.

Discussion

CHANTER is a novel radiologic and clinical syndrome identified by the presence of restricted diffusion areas in the brain. These areas are present in the basal ganglia, hippocampi, and cerebellar hemispheres with grossly symmetrical involvement. This pattern is commonly observed in patients with a history of opiate use or other substance abuse, including cocaine. Patients usually present with stupor or comatose conditions. There is an increased risk for obstructive hydrocephalus which may need more aggressive treatment and possible surgical intervention. The obstructive hydrocephalus did not develop in our case. We believe that is because the involvement was less severe and more peripherally located in the cerebellar hemispheres in our case, as compared to the prior published cases. It suggests that patients with severe and central cerebellar involvement may have increased risk for hydrocephalus and may need close clinical and radiologic follow-up.

The differential diagnosis of CHANTER syndrome should include acute ischemic stroke, hypoxic ischemic encephalopathy (HIE) and Heroin-associated spongiform leukoencephalopathy (HASL). Acute ischemic stroke can cause areas of restricted diffusion in a vascular territory and occlusion of vasculature may be seen on MRA. In our case, all macrovascular structures within the territories of ischemia looked grossly normal without evidence of hemodynamically significant stenosis or thrombosis. The restricted diffusion areas seen in HIE may include basal ganglia, hippocampus, and cerebellar hemispheres like CHANTER; however other high metabolic demand areas are also affected such as the cerebral cortex, thalamus, and subcortical white matter. HIE can also cause diffuse cerebral edema. Moreover, obstructive hydrocephalus is not a known complication of HIE. HASL is a vacuolating myelopathy that is characterized by extensive, confluent symmetrical white matter involvement with predilection of the occipital and parietal lobes in the setting of inhaling heroin use, in contrast to basal ganglia and hippocampal involvement seen in CHANTER syndrome. There is more subacute injury and T2 hyperintensities seen in the white matter with or without associated restricted diffusion.

Consequently, with this case, we would like to reiterate the importance of distinguishing CHANTER from other substance use-related acute neurologic injuries. Our case also suggests that hydrocephalus is one of the well-known possible complications of CHANTER syndrome that is likely related to central and severe involvement of the cerebellum. These patients may need close follow-up. We do also recommend a multisystemic approach for those patients to prevent missed life-threatening consequences in the various body systems.

Author contribution

All authors contributed to the study conception and design. Material preparation, data collection, and analysis were performed by Cagri Yurtsever and Jessica Harris. The first draft of the manuscript was written by Cagri Yurtsever and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

Patient consent

All relevant patient information was anonymized and the manuscript only includes non-identifiable images. The informed consent for publication has been obtained.

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