

Episodic astasia-abasia associated with hyperperfusion in the subthalamic region and dorsal brainstem

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Abstract

Astasia-abasia refers to the inability to stand or walk despite possessing good motor strength and conserved voluntary coordination. Although it is usually regarded as a psychogenic disorder, organic causes have been reported. Herein we describe a patient who presented with alcohol-induced episodic astasia-abasia. Interestingly, SPECT performed during an episode showed hyperperfusion in the dorsal brainstem and subthalamic region. These areas roughly coincide with the mesencephalic locomotor region and subthalamic locomotor region, respectively, and it is conceivable that abnormal neural activity in these areas is related to the symptoms in our patient.

INTRODUCTION

Astasia-abasia refers to the inability to stand or walk despite possessing good motor strength and conserved voluntary coordination. Patients are unable to maintain standing posture unassisted and exhibit unusual and dramatic wild lurches in various directions. Although it is usually regarded as a psychogenic disorder, organic causes have been reported.¹⁻⁴ We describe herein a patient who presented with alcohol-induced episodic astasia-abasia associated with hyperperfusion in the subthalamic region and dorsal brainstem.

CASE REPORT

A 45-year-old man with no previous medical history presented with repeated episodes of postural instability that started at the age of 40 years. Most of the episodes occurred after drinking a small amount of alcohol, with 3 bottles of beer (approximately 60 g of alcohol) resulting in difficulties in standing or walking due to instability. This perplexed the patient because he had formerly been boastful regarding his ability to remain fairly sober after drinking more than 20 bottles of beer. The episodes, which lasted 30–40 minutes each, were not accompanied by dizziness, nausea, vomiting, dysarthria, diplopia, motor weakness, sensory disturbances, or headache. Prolonged sitting for

a couple of hours or running for a few minutes provoked similar but less severe symptoms that lasted for less than 1 minute. These symptoms never developed in any other circumstances and the patient denied experiencing any symptoms between episodes. There was no family history of paroxysmal movement disorders.

An initial neurologic examination between episodes revealed no abnormality. The patient was challenged with alcohol to provoke the symptoms. After drinking, while he had no instability in the sitting position, the patient exhibited difficulty in standing up from a chair and swayed laterally while in the standing position, but with no tendency to deviate to one particular side. When walking, he lurched wildly with a slightly wide-based disorderly gait and inconsistent foot positioning (Video 1). He showed no gait ignition failure, freezing of gait, or parkinsonism. A neurologic examination performed during the episode showed no nystagmus, dysarthria, motor weakness, or sensory abnormalities. Extraocular movements were normal. The finger-to-nose test, rapid alternating movement, and heel-to-shin test were normal. His symptoms gradually disappeared after 40 minutes. Similar but less severe symptoms with a duration of 30 seconds were provoked after prolonged sitting for 2 or 3 hours. Results of routine laboratory tests were normal. Sequencing of all 48 exons and flanking introns of the

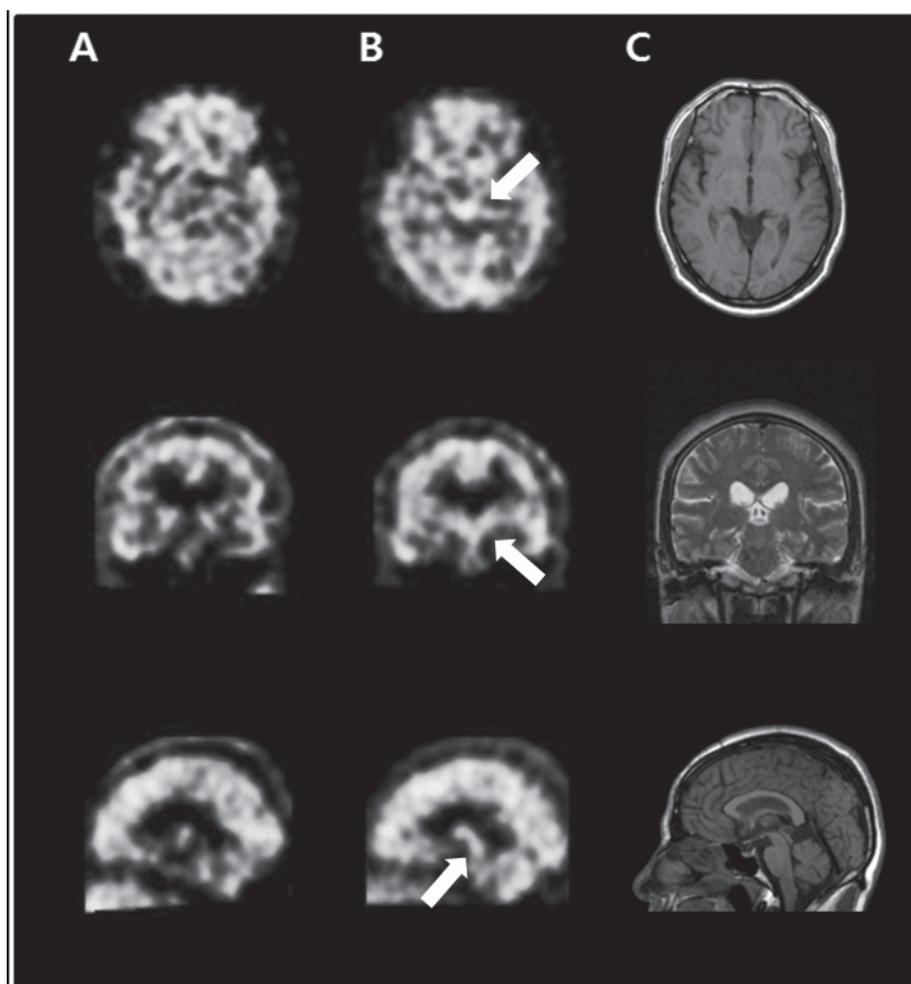


Figure 1. **A:** Findings of ^{99m}Tc -HMPAO SPECT between episodes were normal. **B:** SPECT with injection of ^{99m}Tc -HMPAO during the episode induced by prolonged sitting showed hyperperfusion in the subthalamic region, dorsal midbrain, and dorsal upper pons (Arrows). **C:** T1 (sagittal and axial)- and T2 (coronal)-weighted MRI were unremarkable.

CACNA1A gene revealed no known mutations or novel sequence variants. Brain MRI and MRA were unremarkable. ^{99m}Tc -HMPAO SPECT and ^{18}F -FDG PET performed between episodes were normal. However, SPECT with injection of ^{99m}Tc -HMPAO during an episode induced by prolonged sitting showed hyperperfusion in the subthalamic region, dorsal midbrain, and dorsal upper pons (Figure 1).

Various medications including benzodiazepine, carbamazepine, amantadine, methazolamide, buspirone, levetiracetam, and levodopa were not beneficial.

DISCUSSION

Our patient had recurrent episodes of disturbance in standing and walking without limb weakness.

We consider his condition more likely to be astasia-abasia than ataxia, because it was not accompanied by limb ataxia, nystagmus, or dysarthria, and his stance was only slightly wide-based.

Symptomatic astasia-abasia has been associated with lesions affecting the pontomesencephalic region, thalamus, corpus callosum, or cingulate cortex.¹⁻⁴ Although not referred to as astasia-abasia, postural and gait disturbance without limb ataxia has also been reported in patients with dorsal midbrain infarctions.^{5,6} It has been postulated that astasia-abasia results from disruption of the vestibulocerebellothalamocortical connection⁷ or from damage to the mesencephalic locomotor region (MLR).^{3,6} The dorsal brainstem, and specifically the midbrain tegmentum, is the

anatomical region where these two functional systems overlap. Interestingly, SPECT performed during an episode in our patient showed hyperperfusion in the dorsal brainstem and subthalamic region. These areas roughly coincide with the MLR and subthalamic locomotor region, respectively⁸, and it is conceivable that abnormal neural activity in these areas is related to the symptoms in our patient. Although locomotor regions are usually described in relation to gait freezing, there is evidence that clinical manifestations of derangement of the locomotor regions are not restricted to gait freezing.^{8,9} However, it cannot be excluded that his symptoms are the result of functional impairment of the fibers connecting the vestibulocerebellum and thalamus, which itself is caused by or associated with the hyperperfusion observed in this area.

It is unclear why this patient's symptoms are episodic and provoked only by alcohol intake, prolonged sitting, or brief running, and not in other circumstances. Furthermore, we cannot explain how standing after prolonged sitting causes regional hyperperfusion in our patient. One may argue that the SPECT findings described here are simply a result of postural change, or that they are caused by, rather than being the cause of, his abnormal stance and gait. However, the pattern of regional hyperperfusion that we found on SPECT has never been described in studies on alterations in cerebral blood flow after postural change or alcohol intake^{10,11}, and differs from that of patients with psychogenic astasia-abasia.¹²

Legends to the Video

Video 1. Video taken after drinking alcohol ([http://www.neurology-asia.org/content/15/3/neuroasia-2010-15\(3\)-279-v1.mpg](http://www.neurology-asia.org/content/15/3/neuroasia-2010-15(3)-279-v1.mpg)). The patient swayed laterally in the standing position. He staggered when walking, with his gait pattern being disorderly, slightly wide-based, and having inconsistent foot positioning.

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