



Exercise Headache Associated With an Arteriovenous Fistula of the External Carotid Artery

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Dear Editor,

Exercise headache is a headache provoked by exercise or exertion.¹ Symptomatic causes of exercise headache include subarachnoid hemorrhage, arterial dissection, reversible cerebral vasoconstriction syndrome, and, less commonly, space-occupying lesions, sinusitis, Chiari malformation, and pheochromocytoma.¹ We report a patient with recurrent exercise-induced and coital headaches associated with an arteriovenous fistula between the external carotid artery and the external jugular vein.

A 27-year old male presented with a 15-year history of severe left occipital headaches induced by physical exercise (e.g., anaerobic leg exercises at a gym) and a 3-year history of headaches during coitus. The exercise-induced headaches peaked within 10 minutes, and then subsided over a couple of hours. He described a left occipital throbbing without associated nausea or light or noise sensitivity. The intensity was 8/10 at about 10 minutes after the onset. The headaches during coitus increased with the degree of sexual excitement. Coughing and other Valsalva (straining) maneuvers did not provoke the headache. These headache attacks occurred over periods lasting 2 to 3 weeks, separated by pain-free periods lasting 6 months or longer. There was no history of neck trauma or surgery.

The patient visited his nearest hospital and was started on nortriptyline, which was not effective at preventing the headaches; however, they did respond to acetaminophen. He underwent brain magnetic resonance (MR) imaging, which produced normal findings. However, computed tomography (CT) angiography of the head and neck demonstrated an arteriovenous fistula between the left external carotid artery and the left external jugular vein (Fig. 1A). He visited our hospital for a second opinion, where he underwent conventional angiography, which confirmed the fistula (Fig. 1B and C). Coil embolization was performed, and the final conventional angiography showed no residual filling of the fistula (Fig. 1D). He was followed up for 2.5 years, during which he was free of exercise-induced and coital headaches.

Most arteriovenous fistulas in the head and neck region occur as a result of trauma, iatrogenic injury, or extensive infection of the neck. Congenital arteriovenous fistulas between the external carotid artery and jugular vein are rare, and they are characterized by symptoms and signs such as pulsatile mass, pulsatile tinnitus, facial pain, vertigo, and dyspnea.^{2,3} It is not clear why the carotid-jugular fistula caused the exercise headaches in the present case. Halbach et al.⁴ investigated six patients with arteriovenous fistulas of the internal maxillary artery, and found that the most common initial symptom was bruit, which was exacerbated by exercise.

Two theories on the pathophysiology of secondary exercise headache in our patient may be proposed. First, increased blood pressure during exercise may result in venous or arterial distension, which may cause engorgement of the arteriovenous fistula and mechanical compression of adjacent pain-sensitive structures.^{4,5} Second, increased intra-abdominal and intrathoracic pressures during exercise may be transmitted through the venous system into an

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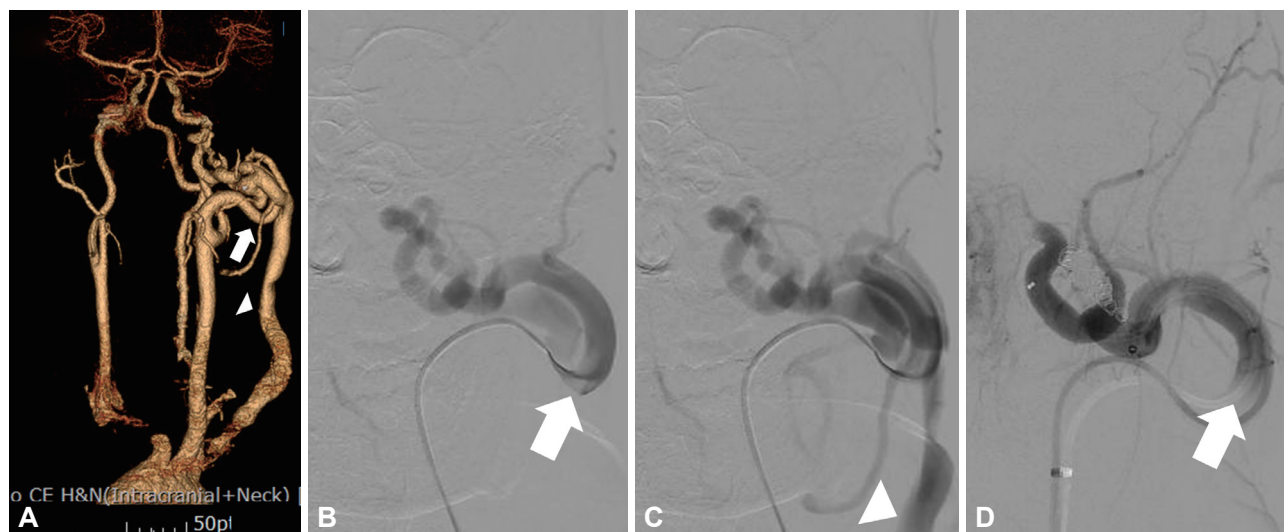


Fig. 1. CT and conventional angiogram of the patient. A: CT angiogram of the head and neck shows enlarged left common and external carotid (arrow) arteries and left external jugular vein (arrowhead). The left external carotid artery supplies an arteriovenous fistula with venous drainage into the left external jugular vein. B and C: Early- (B) and late- (C) arterial-phase anteroposterior views from a selective external carotid angiogram show a fistula between the left external carotid artery (arrow) and the left external jugular vein (arrowhead). D: Angiogram of the same artery (arrow) after embolization shows no residual filling of the fistula.

arteriovenous fistula or the intracranial venous sinuses, causing distension of the fistula or the venous sinuses.⁶

A particularly interesting finding in our patient was that his headaches occurred in clusters separated by headache-free remission periods, which is a characteristic of cluster headache.⁷ Symptomatic causes of cluster headache include an arteriovenous malformation and an aneurysm of the middle cerebral artery.^{8,9} Although it is unclear why our patient's headaches occurred in clusters, they may be explained by the threshold hypothesis.⁷ We hypothesize that there is a threshold for precipitating exercise headache and that the blood flow through the arteriovenous fistula or the threshold might change over time under the influence of unknown factors.

The European Headache Federation guideline from 2015 recommends that patients with recurrent exercise headaches undergo investigations including brain MR imaging and MR angiography, and carotid and vertebral MR angiography when there are red flags.¹⁰ It was interesting that the headaches in our patient were not associated with any red flags other than their precipitation by physical activity.

Direct surgical ligation of the fistula, coil embolization, or detachable balloon embolization has been the conventional treatment for carotid–jugular fistulas.^{2–4} Our patient's exercise headache resolved after the fistula was completely obliterated, suggesting that the fistula was the cause of his exercise headache.

In conclusion, carotid–jugular fistula can be a structural cause of recurrent exercise headache, and CT angiography or MR angiography can be helpful in the diagnosis. Successful coil

embolization may result in resolution of exercise headache.

Ethics Statement

This report was approved for an Institutional Review Board (IRB) exemption by the IRB of the Nowon Eulji Medical Center (IRB file number 2021-08-008). The IRB exemption includes a waiver of the need to obtain signed informed consent.

Availability of Data and Material

Data sharing not applicable to this article as no datasets were generated or analyzed during the study.

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Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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