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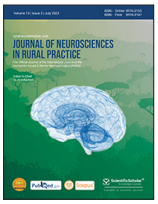

Case Report

Chronic pulsatile tinnitus and continuous vertigo due to very delayed diagnosis of single slow-flow dural arteriovenous

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ABSTRACT

Tinnitus and vertigo are classic symptoms of inner ear disease. Dural arteriovenous fistulas (DAVF) are a rare type of acquired intracranial vascular malformation whose symptoms mimic inner ear disease, but what distinguishes it from other tinnitus is the characteristic of DAVF is pulsatile and heartbeat-synchronous. We present a 58-year-old male with chronic left-sided pulsatile tinnitus (PT) for 30 years and continuous vertigo for 3 years that took numerous consultations to establish a diagnosis after the onset of symptoms. Delay in diagnosis is caused by normal magnetic resonance imaging and an unrecognized subtle mass in the left temporal region by time-of-flight magnetic resonance angiography (TOF-MRA) at the screening test. As we know, TOF-MRA could not provide a clear picture to establish a slow-flow DAVF. Cerebral angiography, a gold standard diagnostic, revealed a Borden/Cognard Type I single slow-flow DAVF in the left temporal region. The patient was treated with superselective transarterial embolization. After 1 week of follow-up, the symptoms of vertigo and PT were completely resolved.

Keywords: Chronic pulsatile tinnitus, Vertigo, Dural arteriovenous fistula, Slow-flow, DAVF

INTRODUCTION

Tinnitus and vertigo are classic symptoms of inner ear disease. Heartbeat-synchronous pulsatile tinnitus (PT) indicates, it is predominantly vascular, making it treatable when the causative vascular anomalies are found and resolved.[1] PT can cause distress and impairment across cognitive, functional, and mental issues such as depression.[2]

Seeking the etiology of PT is essential.[3] Dural arteriovenous fistula (DAVF), the most known cause of PT, accounts for 10–15% of all intracranial vascular malformations worldwide.[4] About 13.6% of patient with DAVF has only PT as an initial symptom. In daily practice, detecting DAVFs using non-invasive imaging remain challenging, especially for slow-flow DAVFs.[4-6] This case report presents a delayed diagnosis case of a single slow-flow DAVF with a longstanding history of PT and vertigo undetected by magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA). Symptoms completely resolved after superselective embolization.

CASE REPORT

A 58-year-old male presented with continuous left-sided tinnitus for 30 years without hearing loss. He described tinnitus as a constant rhythmic sound that synchronized with his heartbeat. Three years before the presentation, the patient was experiencing worsening vertigo. Vertigo was not affected by positional changes but was sometimes accompanied by nausea and vomiting, lasting for hours. The patient claimed that the symptoms affected his quality of life (QoL) with a Tinnitus Handicap Inventory (THI) score of 70.

There were no focal neurological deficits. Audiological examinations showed that his peripheral hearing was normal. MRI showed no abnormalities, yet time-of-flight MRA (TOF-MRA) showed a dilated vessel from the left external carotid artery (ECA) branch on the left temporal region [Figure 1a]. Due to the typical symptoms of vascular lesion and a dilated vessel, concern was raised for a potential DAVF, and we continued with six-vessel cerebral DSA plus ECA branches injection.

A routine six-vessel angiography revealed no abnormal findings in the arterial phase. However, injection of the left

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ECA confirmed the presence of a Borden/Cognard Type I dural fistula in the left middle cranial fossa with a feeder from the internal maxillary artery through the accessories branch of the meningeal artery, with venous ectasia with a size of 4.8 mm × 8.2 mm [Figure 1b and 1c]. The diagnosis of DAVF, fed by the left ECA and draining into the left inferior petrosal sinus [Figure 1d], was therefore made.

Due to the severity of tinnitus and the dizziness impacting the patient's QoL, we performed a superselective embolization using the Seldinger technique through transfemoral access. The Magic microcatheter 1.5–F was positioned as close as DAVF [Figure 2a]. N-butyl-2-cyanoacrylate (n-BCA) and iodized oil (Lipiodol) with a concentration of 1:1 50% were chosen as liquid embolic agents because it was a slow-flow DAVF [Figure 2b]. Post-embolization angiogram showed no more fistula and patent flow of the artery branches [Figure 2c].

During the 1-week follow-up, vertigo and PT were completely resolved with a THI score of 8. We considered performing TOF-MRA in the 6-month follow-up.

DISCUSSION

Tinnitus and vertigo are classic symptoms of inner ear disease due to membranous labyrinth involvement.[7] It is fundamental to recognize between inner and non-inner ear causes of vertigo and tinnitus. Heartbeat-synchronous PT, means the pulsation pattern, occurs with the same rhythm as the heartbeat, indicating it is predominantly vascular in origin.[1]

DAVF, as the most known cause of PT, may have a wide range of symptoms and signs, depending on the hemodynamic properties and the fistula's location. In numerous cases, these conditions are negligibly symptomatic or indeed asymptomatic.[4] Symptoms of vertigo can also occur in

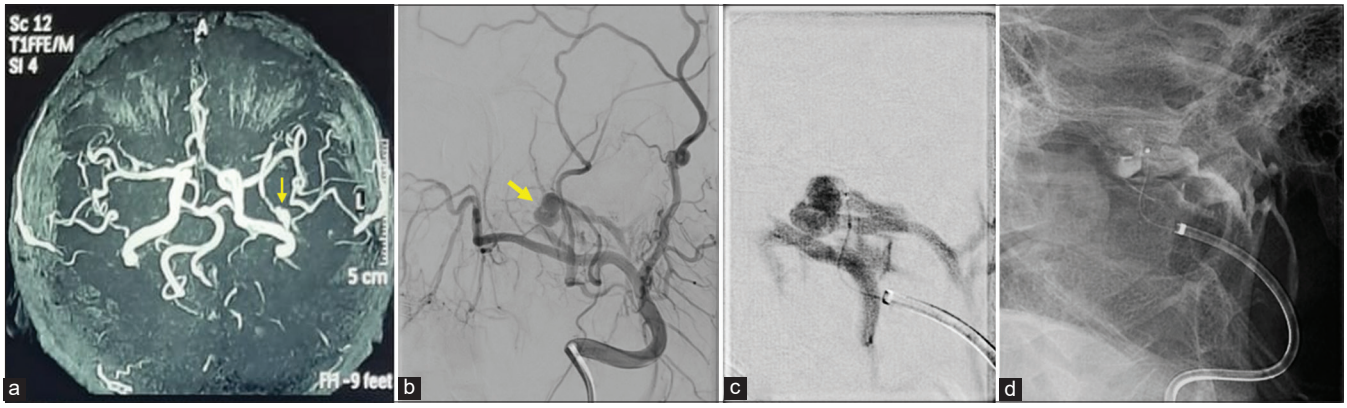

Figure 1: (a) AP view of time-of-flight magnetic resonance angiography showed a dilated vessel from the branch of external carotid artery (ECA) (yellow arrow). (b) The lateral angiographic view of the left ECA shows a dural fistula with primary feeding from the accessory branch of the meningeal artery (yellow arrow). (c) Superselective microcatheter injection of accessories meningeal artery. (d) Late anterograde venous flow to inferior petrosal sinus.

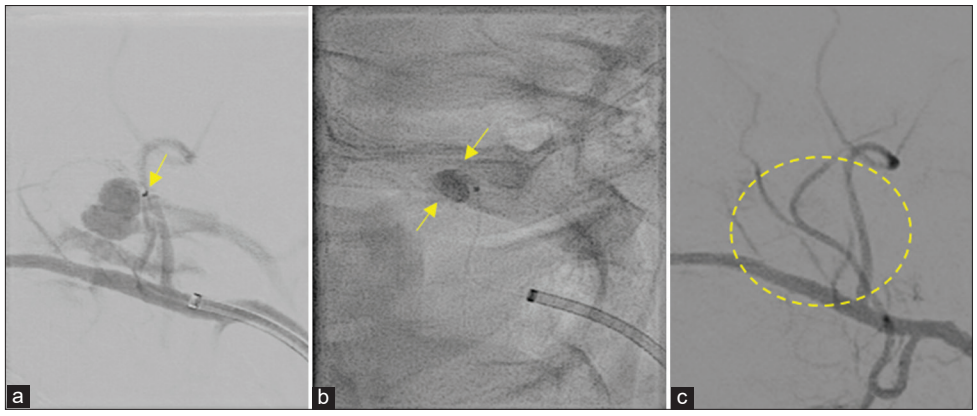

Figure 2: Transarterial curative embolization. (a) The microcatheter was positioned as close as dural arteriovenous fistula, the tip of the microcatheter (yellow arrow). (b) N-butyl-2-cyanoacrylate and iodized oil (Lipiodol) with concentration 1:1 50% were injected through the microcatheter to embolize the fistulous point (yellow arrow). (c) Post-embolization showed no more fistula (circle) and patent flow of the artery branches.

patients with DAVF; one of the underlying mechanisms may be related to the location of the semicircular canals adjacent to sinus drainage, especially the petrosal sinus. As in our case, he came with complaints of PT and vertigo.

Initial non-invasive imaging is helpful in detecting vascular causes and brain tissue abnormality.[4] Unfortunately, DAVFs frequently show only subtle findings on structural imaging. False negatives also occur, with some tiny and low-flow fistulas occult on TOF-MRA, so sensitivity is imperfect compared with DSA.[6,8] Regardless of a negative imaging result, DSA protocol with injection into six-vessel and ECA branches should be performed in patients with unexplained symptoms and imaging signs that propose a possible vascular lesion.[8] It is required for determining fistula angioarchitecture, fistula location, anatomy of the ECAs and their dural branches, degree of dural sinus stenosis, and identifying high-risk features, including cerebrovascular disease and venous outflow obstruction.[4,5]

Patient features, symptom severity, and risk of significant sequelae should all be considered when treating DAVF. The patient presented with progressive clinical presentations that significantly reduced his QoL, so we treated him with endovascular embolization. Endovascular therapy, the first-line established treatment of choice, aims to eliminate the site of fistulization between feeding arteries and draining veins.[4] We used liquid embolic n-BCA agent into the fistula through transarterial. Embolization with NBCA-lipiodol allows embolization at the desired target and requires only a drop of glue. Embolization with other materials, such as polyvinyl alcohol, is uncommon in DAVF cases. Meanwhile, using Onyx material has a reflux risk and the amounts needed are large, making the risk greater. A previous study showed that a favorable outcome of embolization using acrylic glue still allows for a low-cost, quick, and effective treatment with rates of 83%.[9]

Diagnostic delay is common in symptomatic DAVFs with unspecified neurological symptoms. Patients rely on those subjective symptoms and consult with physicians from various departments.[10] Furthermore, in this case, treatment of DAVF led to substantial symptom improvement and should not be delayed.

CONCLUSION

DAVFs symptoms can mimic inner ear disease. Physicians, including GPs, otorhinolaryngologists, and neurologists, should thoroughly evaluate patients with PT and vertigo to rule out the possibility of DAVF through thorough history taking, physical examination, and audiological and neurological evaluations. If clinical suspicion of a DAVF is high, six-vessel DSA plus ECA branches injection should be performed as a gold standard regardless of negative findings on non-invasive vascular imaging.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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