

Marginal sinus fistula supplied exclusively by vertebral artery feeders

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Abstract

A 54-year-old woman is reported with severe pulsatile tinnitus. Digital subtraction angiography demonstrated dural arteriovenous fistula of the marginal sinus with feeders arising exclusively from bilateral vertebral arteries. Patient underwent successful transarterial Onyx embolization with complete angiographic and clinical cure.

Keywords

marginal sinus; dural fistula; arteriovenous malformation; vertebral artery; Onyx

Introduction

Intracranial dural arteriovenous fistulas (DAVFs) are acquired pathological connections between extradural arteries and intradural veins or dural venous sinuses. They comprise 10–15% of all intracranial vascular malformations and 35% of all infratentorial vascular malformations [1–4]. DAVFs involving the marginal sinus are uncommon, accounting about 4% of all intracranial DAVFs. Like DAVFs in other locations, they often present with pulsatile bruit or tinnitus in most patients [5].

The marginal sinus encircles the foramen magnum. An adult cadaveric study [6] demonstrated that the sinus communicated anteriorly with the basilar venous plexus in 80% of the specimens and posteriorly with the occipital sinus in all of the specimens. The sinus tapers down as it travels both anteriorly and posteriorly with the maximum vertical height ranging from 7 to 15 mm (mean 10 mm) more laterally. Magnetic resonance angiography (MRA)-based measurements exhibited a mean diameter of the sinus being about 2 mm [7]. The sinus drains ultimately into the sigmoid sinus or jugular bulb [6,8].

Arterial feeders to marginal sinus fistulae (MSF) may arise from vertebral and/or carotid circulations. The arterial supply to all the previously reported cases of MSF involved the external carotid artery. We report the clinical presentation, imaging findings, and endovascular

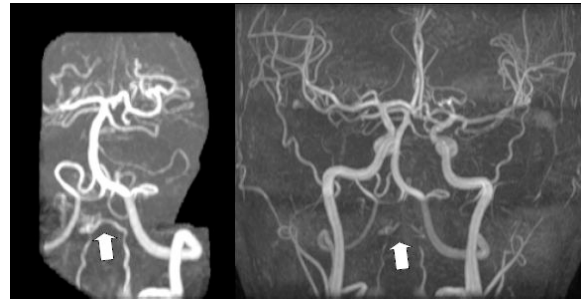


Figure 1. Magnetic resonance angiography demonstrating coalescence of abnormal arteries in the region of marginal sinus (white arrows).

treatment of a patient with an MSF, whose feeders arose exclusively from the vertebral arteries.

Case Report/Technique

A 54-year-old woman reported with a one year history of the bilateral pulsatile tinnitus that was becoming progressively louder. The noise was more pronounced at nighttime resulting in insomnia. MRA revealed a dilated vascular structure at the foramen magnum with convergence of prominent branches from each vertebral artery (Figure 1). Selective catheter angiography of the bilateral carotid and vertebral circulations was performed, and vertebral artery angiograms demonstrated an MSF with arterial feeders arising from V2 segments of the left

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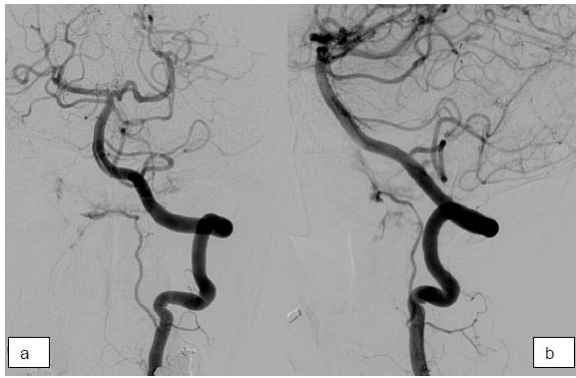


Figure 2.a and b Left vertebral artery arteriogram in anteroposterior (AP) and lateral projections demonstrating the MSF with an arterial feeder arising from the left vertebral artery.

and right vertebral arteries (Figures 2a and b and Figures 3a and b). The MSF are drained via bilateral jugular bulbs. Retrograde cortical venous filling was not demonstrated. After obtaining an informed consent from the patient, we proceeded with treatment of the MSF to relieve her disabling tinnitus. The treatment was performed under general anesthesia with motor and somatosensory evoked potential monitoring.

Initially, we unsuccessfully attempted a transvenous approach to gain access into the marginal sinus. We then proceeded with a transarterial approach with a guide catheter positioned in the proximal left V2 segment. A prominent arterial feeder arising from the ascending portion of the left V2 segment was catheterized, and the microcatheter was advanced into the marginal sinus (Figures 5a and b). Microcatheter injections outlined the marginal sinus as well as retrograde filling of the arterial feeders from the right vertebral artery (Figure 5c). The microcatheter was positioned in the center of the sinus, and we embolized the sinus with ethylene vinyl alcohol copolymer (Onyx 18; Covidien Inc., Irvine, CA) resulting in angiographic obliteration of the fistula (Figure 6a–d). The patient was extubated with normal neurological exam and she reported immediate resolution of the tinnitus. She remained asymptomatic at six-month clinical follow-up.

Discussion

We have described a patient who developed pulsatile tinnitus from MSF with feeders arising exclusively from the vertebral arteries. Most published descriptions of MSF involve single cases or small series. One series [5] reported 14 patients with MSF with arterial supply arising

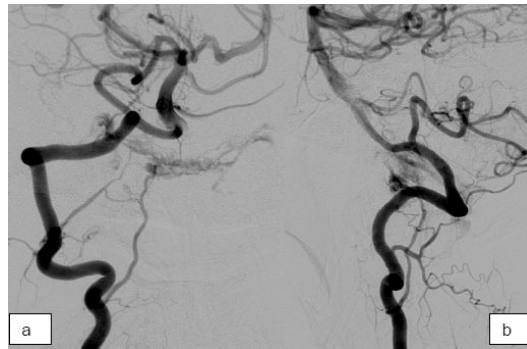


Figure 3.a and b Right vertebral artery arteriogram in AP and lateral projections demonstrating the MSF with arterial feeders arising from the right vertebral artery.



Figure 4. Unsubtracted right vertebral artery arteriogram in AP projection demonstrating MSF around the foramen magnum (white arrow).

from the external carotid artery, internal carotid artery, and the vertebral artery. All patients in the series had at least one external carotid artery feeder; in two patients, branches of the external carotid artery were the only supply to the fistula. Ascending pharyngeal and occipital arteries were involved in 11 and 7 patients, respectively, either solely or in combination. Posterior meningeal arteries and the meningohypophyseal artery supplied the MSF in eight patients.

Several other small series and case reports have documented the external carotid artery involvement in MSF [9–12]. The ascending pharyngeal and occipital arteries seem to be the most common feeders involved in these fistulae. To our knowledge, the current case is the first to demonstrate an MSF without the external carotid artery supply. The MSF in our patient was supplied exclusively by vertebral artery branches.



Figure 5.a and b Marginal sinus fistulogram with a microcatheter via left vertebral artery approach. c Microcatheter injection demonstrating the arterial feeders arising from the contralateral (right) vertebral artery.

A proper treatment requires detailed understanding of the arterial and venous anatomy of this region. Unsubtracted images and venous drainage patterns are necessary to determine the pathological anatomy of an MSF (Figures 4, and 5a and b). DAVFs involving the base of the skull and the upper cervical spine may mimic MSF. For example, fistulae involving the anterior condylar vein in the hypoglossal canal can resemble an MSF [12–14]. Some of these anterior condylar fistulae are supplied solely by the vertebral arteries [15,16]. Unlike MSF, which are midline, the anterior condylar fistulae are situated laterally in the occipital condyles and in proximity to the hypoglossal canal.

Both transarterial and transvenous routes have been used to treat MSF in previous reports [5,9–11]. In our case,



Figure 6.a and b AP and lateral, left vertebral artery angiogram. C and d AP and lateral, right vertebral artery angiogram, demonstrating complete obliteration of the fistula.

we felt that a transvenous approach would be safer and avoid potential compromise of the spinal cord blood supply; when this was not feasible, we used a transarterial approach. When using a transarterial approach, the microcatheter should be positioned as close to the fistulous point as possible. Embolic agents such as Onyx, n-butyl cyanoacrylate, polyvinyl alcohol foam, absolute alcohol, calibrated particles, and silk have all been used in treating DAVFs [17–21]. We routinely use Onyx for transarterial embolizations. To our knowledge, the use of Onyx in the treatment of an MSF has not been described before, though the principles of Onyx embolization are the same as in other dural AVFs. The liquid embolic agent must penetrate across the fistula into the venous side in order to achieve an angiographic cure and minimize the chances of recurrence. Reflux must be minimized to avoid embolization of a normal branch or the parent artery. We elected to use motor and sensory evoked potential monitoring during the procedure as the artery of the cervical enlargement and the cervical radiculomedullary branches usually arise from the V2 segment of the vertebral artery. Other normal branches of the V2 segment include muscular branches, and anterior and posterior meningeal arteries [22].

Conclusion

Recognition of MSF can be challenging. Although the external carotid circulation is typically the major source of supply to these lesions, the present case demonstrates that the posterior circulation can be the sole blood supply. Onyx liquid embolic agent can be used safely to embolize MSF via transarterial route.

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