

Case Report



Stent-Assisted Coil Embolization of a Vertebro-Vertebral Arteriovenous Fistula Secondary to Oriental Acupuncture: A Case Report

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Conflict of Interest

The author has no financial conflicts of interest.

ABSTRACT

A vertebro-vertebral arteriovenous fistula is an abnormal communication between the vertebral artery and adjacent venous structures. Trauma is the most common cause of vertebral arteriovenous fistulas; however, the fistulas can also occur spontaneously. We herein report a case of a traumatic vertebro-vertebral arteriovenous fistula that occurred following the application of oriental acupuncture in the posterior neck region. A 64-year-old previously healthy female patient took over-the-counter medicine for cervical pain that occurred several months before the acupuncture session but showed no improvement. She had undergone oriental acupuncture treatment in the posterior lower neck region 1 month before visiting our hospital. After the treatment, she gradually developed tinnitus, accompanied by dizziness. Abnormal aneurysmal dilated vessels were observed in the right vertebral artery on computed tomography angiography. The patient was immediately admitted and underwent diagnostic angiography. She subsequently underwent stent-assisted coil embolization, which gradually resolved her dizziness and tinnitus.

Keywords: Arteriovenous fistula; Vertebro-vertebral fistula; Acupuncture; Complications

INTRODUCTION

A vertebro-vertebral arteriovenous fistula (VVAVF) is a rare condition involving abnormal direct communication between the extracranial vertebral artery (VA) and the adjacent vertebral venous plexus, without intervening capillaries.^{7,14)} Spontaneous VVAVF occur in patients with known predisposing factors, including neurofibromatosis I, Ehlers–Danlos syndrome, and fibromuscular dysplasia.⁶⁾ However, the most common cause of VVAVF is blunt or penetrating trauma to the neck.⁶⁾ Injury during the insertion of a central venous catheter or blunt cervical trauma that may result from chiropractic exercise or traffic accidents have frequently been reported.⁶⁾

Oriental acupuncture has been used for thousands of years in Asian countries. It is defined as the use of needles placed transcutaneously along a set of defined points to treat diseases and their symptoms. In Asian countries, many people undergo oriental acupuncture treatment to treat conditions ranging from mild pain to incurable disease.⁸⁾ The frequency

of severe complications has been extremely low.⁵⁾ Nevertheless, various adverse effects have been reported, such as infections, internal organ or tissue injury, and adverse reactions.¹³⁾ Traumatic injury includes various organs and tissues, but few cases of vascular injury due to oriental acupuncture have been reported²⁾. Furthermore, no cases about VVAVF caused by oriental acupuncture have been described in the literature.

We report a case of VVAVF that occurred after oriental acupuncture. We describe the patient's symptoms and diagnosis and discuss the treatment and prognosis.

CASE REPORT

A 64-year-old previously healthy female patient presented to the outpatient neurosurgery department due to left side pulsatile tinnitus and dizziness. No neurological deficits were noted. Three months previously, the patient had undergone medication and physical therapy for cervical neck pain. She had undergone oriental acupuncture treatment in the posterior lower neck region 1 month before visiting our hospital. After the treatment, her cervical neck pain improved. However, the patient complained of gradual dizziness and pulsatile tinnitus developing in the left side thereafter. The patient underwent magnetic resonance angiography (MRA) evaluation for diagnosis at another hospital. MRA showed an approximately 6.2×5.5 mm sized saccular vascular dilatation with a venous structure in the right VA V2 segment (**FIGURE 1A**). When the patient visited our hospital, she reported that the tinnitus had worsened. Neck auscultation revealed a slight murmur in the right lower cervical level.

The patient immediately underwent conventional angiography. On digital subtraction angiography, the right VA exhibited a dilated aneurysm measuring 8.6 mm (highest width) \times 7.2 mm (height) in the right V2 segment (**FIGURE 1B**), thereby forming an abnormal fistula

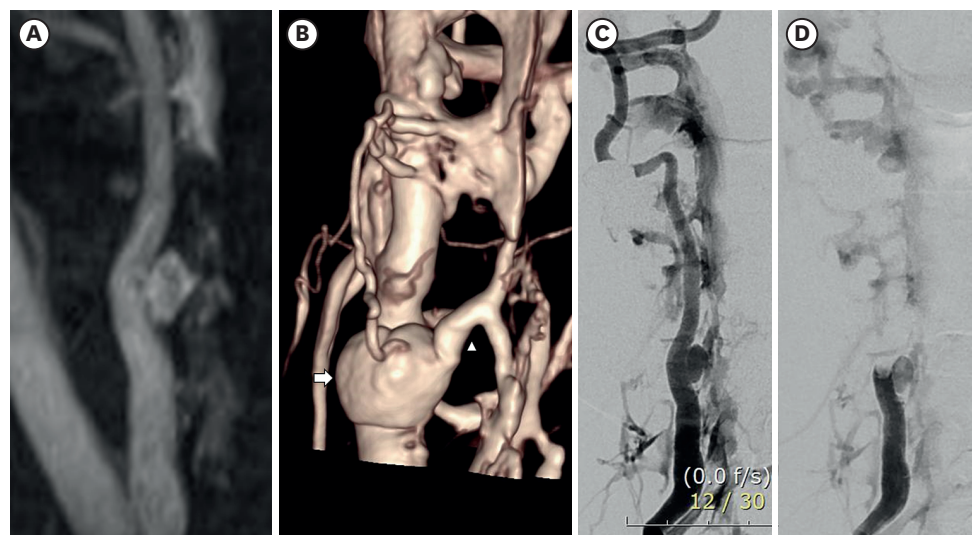


FIGURE 1. Pre-operative radiologic findings. (A) A 64-year-old female presented with progressive dizziness and tinnitus due to VVAVF on magnetic resonance angiography. (B) DSA revealed a broad-necked small aneurysm (white arrow) with fistula (small arrow) portion in the proximal right VA. (C) Rapid shunting blood flow occurred in the paravertebral venous plexus. (D) Upon slow injection of a contrast medium (2 mL per second with total 5 mL), most of the contrast medium accumulated in the paravertebral venous plexus through the fistula. VVAVF: vertebro-vertebral arteriovenous fistula, DSA: digital subtraction angiography, VA: vertebral artery.

with a paravertebral venous plexus. The fistulous portion was accompanied by a dissected aneurysm portion with a flap. Shunting of blood flow rapidly occurred in the paravertebral venous plexus (**FIGURE 1C**). Upon slow injection of a contrast medium (2 mL per second with total 5 mL), most of the contrast medium accumulated in the paravertebral venous plexus through the fistula (**FIGURE 1D**).

Endovascular treatment of VVAVF

Under general anesthesia, a 6F sheath was introduced into the right femoral artery following a standard Seldinger technique. An Envoy guiding catheter (CERENOVUS, Irvine, CA, USA) was then placed into the right proximal VA.

Subsequently, a Prowler select plus 21 straight microcatheter (CERENOVUS) was placed at the distal right VA through the dissection segment using a Synchro 14 microguidewire (Stryker, Portage, MI, USA). The other headway 17 preshaped 45° microcatheter (Microvention, Tustin, CA, USA) was guided into the small sac of the dilatated aneurysm carefully using a Synchro microguidewire (Stryker Neurovascular, Cork, Ireland). After making a 4.0×120 mm frame using an Orbit Galaxy detachable coil (CERENOVUS), an Enterprise stent (CERENOVUS) 4.5×23 mm was deployed to cover the broad aneurysm neck (**FIGURE 2A**). Subsequently, Axium prime 3D detachable coils (Medtronic, Minneapolis, MN, USA) measuring 3.0×100 mm, 2.5×60 mm, 2.0×40 mm, and 1.5×40 mm were inserted into the aneurysm (**FIGURE 2B**). After deployment of coils in the sac, the fistula was still seen after 10 minutes. Double overlapping stent-assisted coil embolization was successfully performed using an LVIS stent (Microvention) 4.5×23 mm in size, which resulted in complete aneurysmal occlusion (**FIGURE 2C**). No fistulous segments or venous structures were observed on the final vertebral angiogram (**FIGURE 2D**), which gradually resolved her dizziness and tinnitus.

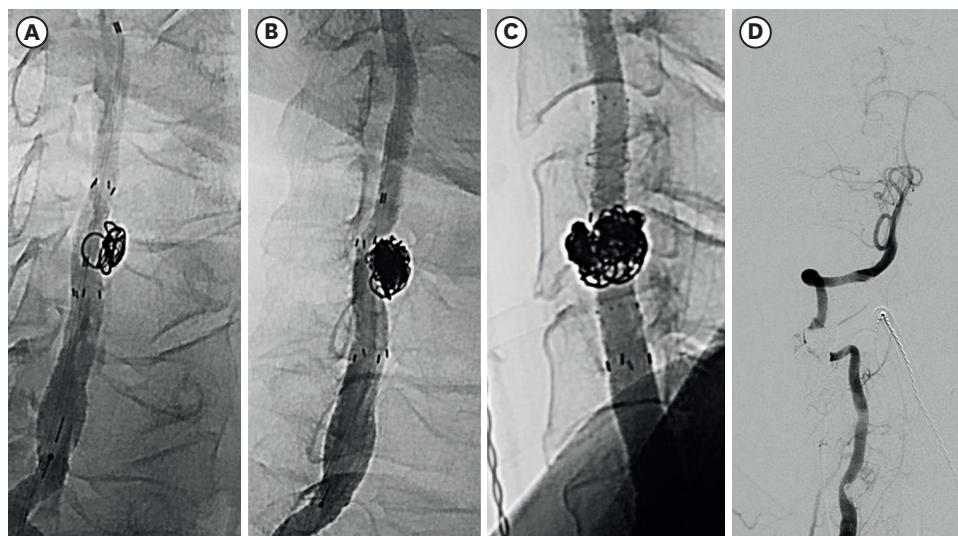


FIGURE 2. Cerebral angiography of stent assisted coil embolization for vertebro-vertebral fistula. (A) After making frame into the sac, a stent was deployed to cover the broad aneurysm neck. (B) Because the fistula was seen after several coils were deployed into the sac, the microcatheter was passed through the deployed. (C) Double overlapping stent-assisted coil embolization was successfully performed. (D) No fistulous segments or venous structures were observed on the final vertebral angiogram.

DISCUSSION

VVAVFs are rare occurrences and arise spontaneously or following underlying connective tissue disorders. However, the most common cause of VVAVF is neck trauma and iatrogenic injuries, such as percutaneous internal jugular vein cannulation and pedicle screw fixation after vertebral fracture.⁹⁾ This case showed that VVAVFs can also be caused by acupuncture treatment used in oriental medicine.

Many studies have reported side effects after treatment with oriental acupuncture. Acupuncture can cause infection and internal organ or tissue injury, such as pneumothorax, heart damage, and peripheral nerve injury.¹³⁾ Studies on serious injuries that occur after acupuncture treatment are rare. Of nine central nervous system injury cases, 2 involved intracranial hemorrhage, 3 epidural hematoma, 1 subarachnoid hemorrhage, 1 cerebrospinal fluid fistula, and 2 direct injury to the brain.¹³⁾ A systematic review of vascular injuries caused by acupuncture reported that of 31 cases, 3 led to death and involved severe complications, including cardiac tamponade, aortoduodenal fistula, ischemia, venous thrombosis, pseudoaneurysm, and compartment syndrome.³⁾ Peuker and Grönemeyer¹²⁾ reviewed four studies on the association between lesions in blood vessels and acupuncture. A pseudoaneurysm of the costocervical artery was reported in 1994. Another case report of an aneurysm of the posterior wall of the popliteal artery was reported in 1996. One case involved deep vein thrombophlebitis after acupuncture in the upper calf region, with leg pain developing 48 hours after the treatment. A further case involved anterior compartment syndrome in the upper calf after acupuncture. However, no complications in the VA have been published till now.

VVAVF symptoms depend on the location of the lesion and the flow pattern. About half of the patients with a VVAVF are asymptomatic, but other patients develop symptoms, such as tinnitus or vertebrobasilar insufficiency.¹⁾ The patient in this case presented with tinnitus and dizziness. Tinnitus is the most frequent symptom of VVAVFs, whereas dizziness is likely caused by the steal phenomenon that occurs with VVAVFs. Particularly, vertebral angiography revealed that the steal phenomenon was outstanding through the slow injection of a contrast medium.

In VVAVFs, accurately distinguishing whether the velocity of shunt flow is high or slow is important.¹¹⁾ A slow-flow VVAVF can be managed using serial radiological angiography and conservative therapy. However, a high-flow VVAVF should be treated by complete obliteration of the fistulous area because of progressive symptoms and signs of arterial steal. No definite guidelines have been established for managing VA injuries combined with high-flow VVAVF.¹¹⁾ Here, we performed endovascular treatment because of progressive tinnitus.

The depth of insertion of acupuncture needles varies from a few millimeters to several centimeters. Because the needle often lies in the organs and associated nerves, muscles, and vessels, acupuncturists must have an accurate knowledge of anatomy to avoid causing direct trauma. All iatrogenic injuries could be avoided if acupuncturists gain better anatomical knowledge and practice more careful treatment.¹²⁾

Although the depth from the skin to the transverse process while applying pressure could greatly vary individually, an ultrasonographic study revealed that it was 9.5 ± 2.7 mm on the left side and 9.7 ± 2.5 mm on the right side in men, whereas it was 8.0 ± 2.2 mm on the left side and 8.2 ± 2.0 mm on the right side in women.⁴⁾ The mean interforaminal distance of the

transverse foramen in axial cervical images ranged from 25.59±1.12 mm to 29.73±1.41 mm in men and from 24.03±1.17 to 28.17±1.25 mm in women.¹⁰⁾ Additionally, the transverse foramen diameter ranged from 6.5±0.87 mm to 6.63±0.67 mm in men and from 5.81±0.70 to 5.89±0.65 mm in women.¹⁰⁾ When performing invasive procedures with a cervical posterior approach, approaching from at least up to 18 mm lateral to the midline and no deeper than up to 10 mm is recommended to avoid damage to the VA while applying pressure.

The main limitation of our case report was the difficulty in obtaining evidence that the VVAVF was directly damaged by the acupuncture needle. However, we found a high probability of a causal relationship based on the congruence between the treatment site and VVAVF lesion and the immediate onset of symptoms after acupuncture treatment.

CONCLUSION

VVAVFs can be cured through endovascular treatment based on the patient's symptoms and blood flow after accurate diagnosis by angiography. Accurate knowledge of the anatomical locations of blood vessels in patients requiring invasive treatment, including acupuncture, is important to prevent complications.

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