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Case Report

Successful balloon-assisted coil embolization for a diagnostically difficult case of spontaneous vertebro-vertebral arteriovenous fistula

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ABSTRACT

Background: We describe a rare case of idiopathic lower cervical vertebro-vertebral arteriovenous fistula (VVAVF) with compression of the nerve roots and spinal cord, successfully treated with detachable coils utilizing the transarterial balloon-assisted technique without complication of coil mass.

Case Description: A 68-year-old woman was admitted for numbness of the left upper limb and pain in the left neck. Cervical magnetic resonance imaging (MRI) revealed compression of nerve roots and spinal cord by a large vascular lesion. The left vertebral angiography demonstrated a VVAVF draining into the vertebral venous plexus at C5 level. Under general anesthesia, the fistula site was accessed with a microcatheter through the right femoral artery, and successful embolization performed by compactly placing several detachable coils using balloon-assisted technique. The patient made a full recovery, with long-term MRI-documented left vertebral artery patency and no fistular leakage, and without postoperative complications.

Conclusion: Target occlusion utilizing the balloon-assisted technique in this case of VVAVF with compression of nerve roots and spinal cord, was effective in improving neurological symptoms, and achieved long-term occlusion when preservation of the VA was desired.

Keywords: Balloon-assisted technique, Cervical portion of vertebral artery, Endovascular embolization, Radiculopathy, Vertebro-vertebral arteriovenous fistula

INTRODUCTION

Vertebro-vertebral arteriovenous fistula (VVAVF) is a shunt created by abnormally high blood flow between the extracranial vertebral artery (VA), or its muscular/radicular branches, and adjacent veins such as the vertebral venous plexus, venous lakes, and jugular veins, excluding capillaries.[3] It is commonly caused by trauma, but can be congenital, spontaneous, idiopathic, or related to dysplastic syndromes. About 30% of VVAVFs are asymptomatic,[2] but some patients may experience symptoms including tinnitus, vertigo, diplopia, or radiculopathy associated with high-flow arteriovenous shunting, steal phenomenon, or compression by venous dilatation.[2,6,9,10]

Surgical ligation or endovascular closure of the high-flow arteriovenous fistula is the main goal of treatment.[1,2] Embolization by detachable balloons, coils, covered stents, or other agents has been used. Here, we present transarterial coil embolization using microballoon-assisted technique, for complete occlusion of a cervical VVAVF with venous plexus-mediated nerve root compression, while preserving the parent artery without causing coil-mediated nerve root compression, and without complications.

CASE DESCRIPTION

A 68-year-old woman without previous relevant medical or traumatic history consulted the orthopedic surgery department for the left upper limb numbness and left neck pain. Noncontrast brain computed tomography and cervical vertebral X-ray were normal. She was prescribed analgesics and followed for about a year, but her symptoms worsened. The patient underwent cervical magnetic resonance imaging (MRI) and was found to have abnormal blood vessels. Thus, she was referred to our neurosurgery department for further evaluation and management.

Neurological examination revealed left C5 radiculopathy and worsening pain and hyperesthesia in the left arm and scapula. Other neurological findings included left deltoid and biceps brachii weakness. A vascular murmur was also heard in the left neck. Cervical spine MRI revealed an extramedullary structure, demonstrated by flow voids with abnormal vascular morphology, adjacent to the left neural foramen from C4 to C6, with compression of nerve roots and spinal cord [Figure 1]. MR angiography (MRA) [Figure 1] revealed a left VVAVF with left VA ectasia, but could not accurately identify the fistulous point. Subsequently, the left VA digital subtraction angiography demonstrated a high-flow VVAVF connecting the left VA to the vertebral venous plexus, with a single fistulous point at the C5 level of the V2 segment [Figure 2], and large venous plexus with drainage into the anterior internal vertebral venous plexus. The anterior cerebral circulation was normal in all angiogram phases. There was no filling of the fistula from the anterior circulation. The contralateral vertebrobasilar circulation was normal with no steal flow into the fistula, except the VVAVF.

We chose compact microballoon-assisted coil embolization of the left expanded paravertebral venous plexus, that is, target shunt occlusion. Heparin (3000 IU intravenously) was administered preoperatively. Under general anesthesia, a 10 cm 7-French femoral short sheath (TERUMO, Tokyo, Japan) was placed into the right common femoral artery. A 90 cm 7-French Guider Guiding Catheter ST (Stryker Japan K.K., Tokyo, Japan) was placed in the left VA. A 150 cm 2.5-French Renegade-18 microcatheter (Stryker Japan K.K., Tokyo, Japan) was transarterially guided to the left paravertebral venous plexus through the shunt using a 180 cm

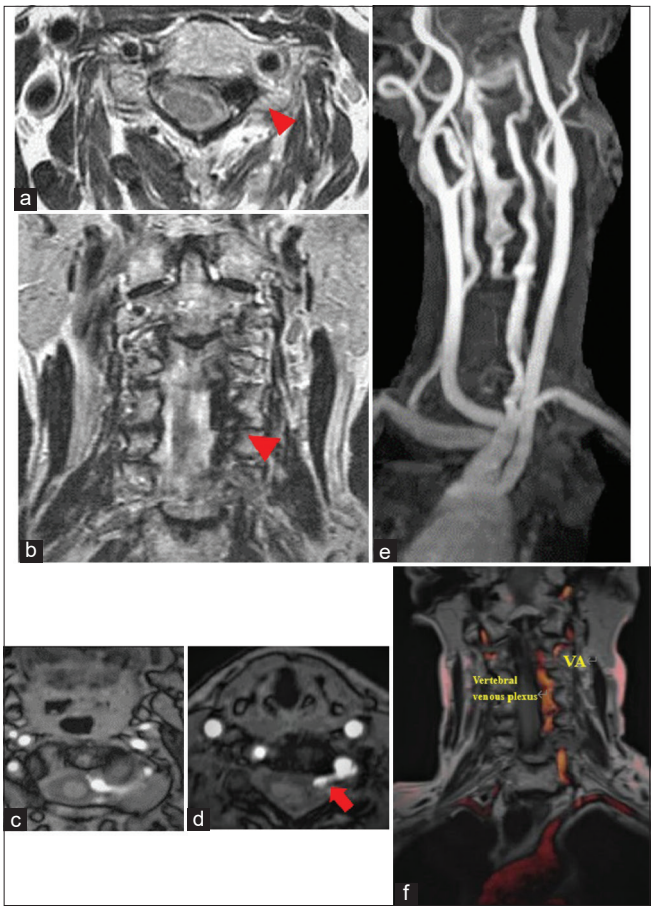

Figure 1: Preoperative MR image. T2-weighted axial (a) and coronal (b) images demonstrated enlarged flow voids of the left anterior internal vertebral venous plexus (arrowheads) at C4 and C3-6. Severe compression of the nerve root and spinal cord from the enlarged epidural venous plexus is also demonstrated. The maximum intensity projection of time-of-flight images demonstrated anterior internal vertebral venous plexus at arterial phase at C2 (c) and C5 (d). The neck MR angiography showed abnormal veins around left vertebral artery (e and f).

0.016-inch GT wire double angle (TERUMO corporation, Tokyo, Japan) through the guiding catheter. In addition, a 4–15 mm HyperGlide microballoon catheter (Medtronic, Tokyo, Japan) was introduced into the left VA shunt point to regulate blood flow, prevent coil migration, and stabilize the microcatheter, so coils would not prolapse into the parent artery. While controlling shunt flow by inflating the microballoon placed in the left VA, the left vertebral venous plexus was embolized tightly and stably over a short distance with the Axium 3D coils 4 × 100 mm and 3.5 × 120 mm (Medtronic, Minneapolis, Minnesota, USA), and Tornado embolization coil 4 × 20 mm and 3 × 20 mm (COOC Medical, Tokyo, Japan). The microcatheter was gradually pulled back while placing the coils, and the fistula was firmly embolized with ED coil 10 ExtraSoft Type R 3.5 × 80 mm, 3 × 60 mm,

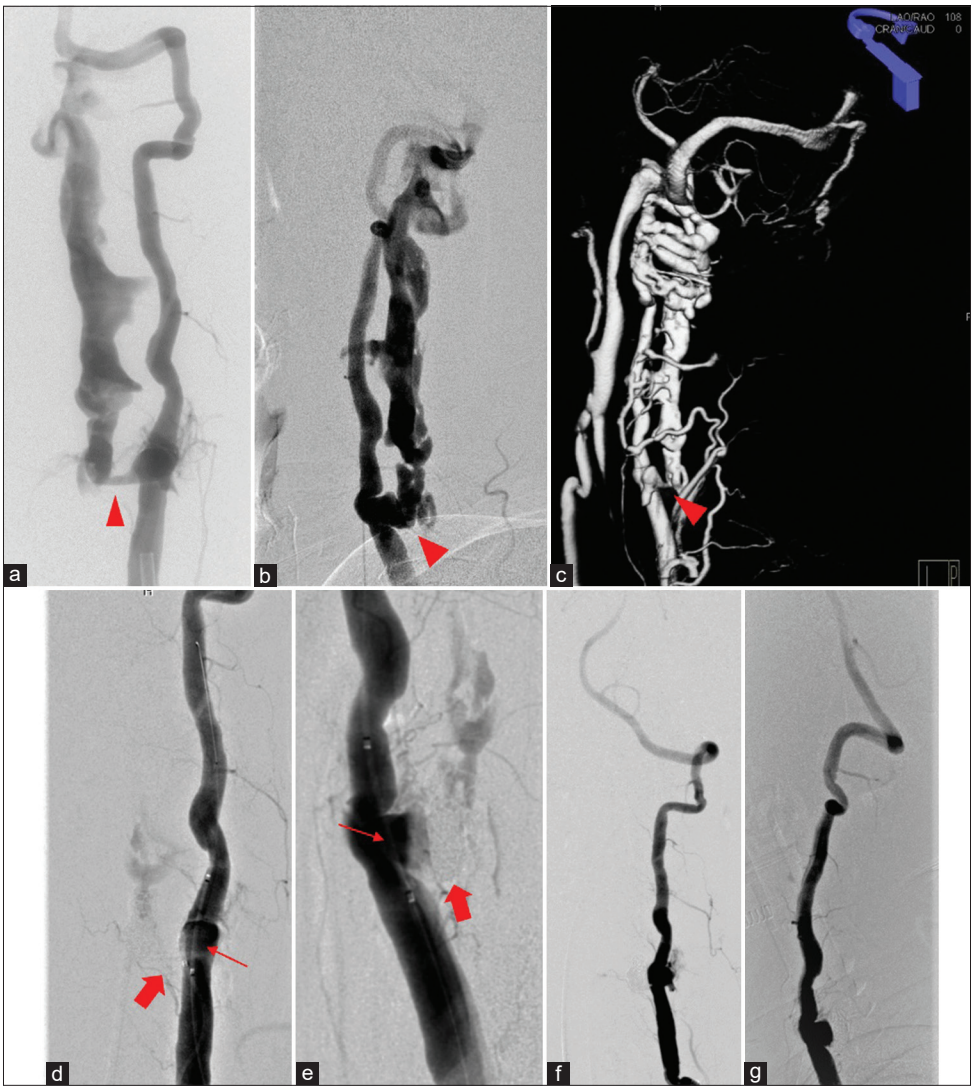

Figure 2: Preoperative and postoperative digital subtraction angiography image. Anteroposterior (AP) view (a) and lateral (b) view of the left vertebral artery (VA) angiography, and oblique view (c) of reconstructed three-dimensional rotational angiography of the left VA demonstrated high-flow vertebro-vertebral arteriovenous fistula (VVAVF) between V2 segment (arrowheads) and dilated vertebral venous plexus. AP view (d) and lateral (e) view of angiogram showed that the coils (arrows) were placed in the left vertebral venous plexus just distal to the shunt, using a microballoon catheter (small arrow). The AP view (f) and lateral view (g) of the left VA angiography obtained immediately after coil embolization demonstrated the disappearance of the VVAVF.

3 × 30 mm and 1.5 × 20 mm (Kaneka Medics, Osaka, Japan). The final angiogram from the left VA demonstrated complete disappearance of the VVAVF, with stable coils [Figure 2]. Immediately after the procedure, the vascular murmur disappeared and we did not prescribe antithrombotic therapy. The patient was discharged 3 days postprocedure.

Pain and numbness in the left upper limb were diminished at 1 month after the procedure. More than 4 years after the procedure, the patient remained well, without any complaint attributable to the endovascular embolization, and equally importantly, her symptoms, such as numbness and weakness, had improved. MRI 4 years postprocedure revealed that the flow void on the left side of the spinal canal had completely disappeared and compression of C4-6 nerve roots and spinal cord released [Figure 3]. Likewise, no recurrence of the shunt was confirmed by MRA, with patency of the right VA [Figure 3e].

DISCUSSION

In this report, we describe a rare case of idiopathic lower cervical VVAVF with compression of the nerve roots and spinal cord, successfully treated with detachable coils utilizing the transarterial balloon-assisted technique, with improved neurological symptoms.

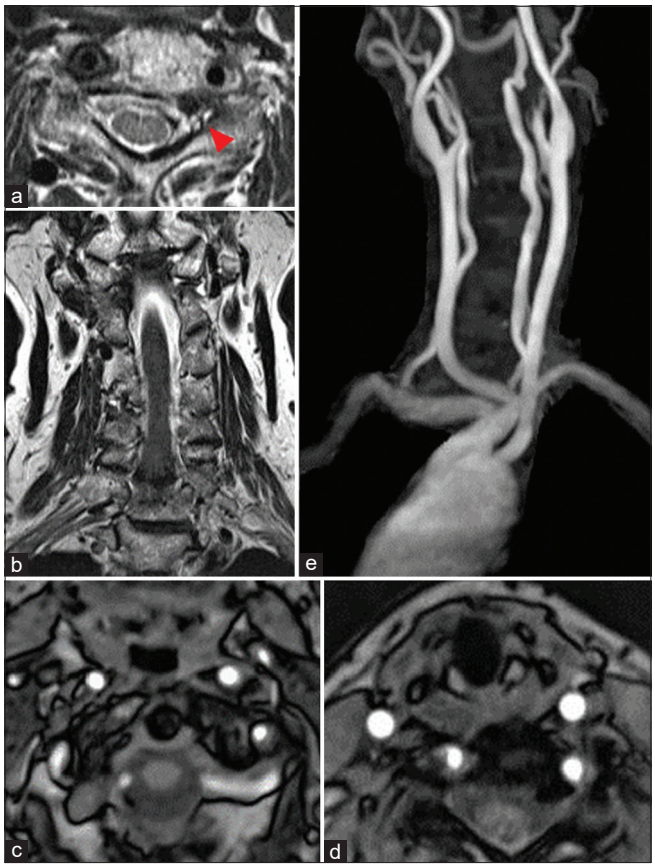

Figure 3: Postoperative MR image. The T2-weighted axial (a) and coronal (b) images 4 years after embolization showed reduction of flow voids of the left anterior internal vertebral venous plexus (arrowhead) at C4, and improvement of compression of nerve roots and spinal cord. The MIP images demonstrated no recurrence of the fistula at C2 (c) and C5 (d). The neck MR angiography obtained 4 years later demonstrated that the fistula was completely occluded (e).

Cervical VVAVFs are uncommon vascular lesions involving an abnormal communication between the extradural VA and surrounding venous structures, including the epidural venous plexus and/or jugular venous system. Common symptoms of VVAVFs include pulsatile tinnitus and vertebrobasilar ischemic symptoms such as vertigo and diplopia, and rare symptoms include congestive heart failure and spinal dysfunction including radiculopathy and myelopathy.[2] Radiculopathy results from direct compression and distortion of the nerve root caused by dilated draining veins and venous plexus in the neural foramina and spinal canal.[9] Due to the rarity of its clinical presentation, the morbidity and mortality of this disease are not well established but are thought to be high if left untreated.[5]

Traumatic, iatrogenic, and spontaneous VVAVFs have different underlying demographics, with important treatment considerations. The spontaneous form is more commonly seen in young women because of the increased prevalence of underlying connective tissue disorders.[1] Spontaneous VVAVFs are most commonly located between C1 and C2,[6] because the increased strain on this segment from the vessel due to physiologic movements in this location, coupled with a possible intrinsic weakness of the vessel wall in this redundant segment of the artery, might be predisposing factors. Spontaneous VVAVF formation in this location is often associated with an underlying congenital abnormality and often associated (25%) with neurofibromatosis type 1.[7]

In our difficult to diagnose case, there was no underlying connective tissue disease or history of trauma. Our patient was atypical, because to her relatively advanced age, having a VVAVF at the lower cervical level despite being idiopathic, and lack of symptoms other than C5 radiculopathy. However, as symptomology of VVAVFs varies according to fistula site and flow pattern, even in her case, physicians should consider this diagnosis.

Therapy is based on signs and symptoms, angioarchitecture, and cause of the fistula. Treatment is indicated if retrograde, intracranial, or spinal cord venous drainage is present. For patients with clinical symptoms, treatment is especially essential. For asymptomatic fistulas, some have suggested that the advantages and disadvantages of treatment should be carefully evaluated. However, most published reports suggest that treatment should be recommended as this condition will further develop and produce corresponding neurological dysfunction.[7] Delay of treatment also allows time for the fistula to recruit additional feeding vessels that may make future treatment more difficult.[5] If left untreated, these fistulas can produce symptoms of vertebrobasilar insufficiency through vascular steal phenomena, aneurysm formation with subsequent thromboembolism due to abnormal flow patterns, compressive myeloradiculopathy due to progressive engorgement of the cervical epidural veins, or catastrophic intramedullary hemorrhage due to intramedullary venous hypertension.[5] Our patient was also diagnosed with VVAVF earlier and needed earlier treatment.

The goal of treatment should be occlusion of the fistula site. Although VVAVFs have been conventionally treated surgically, endovascular treatment has increased due to high risk of surgical complications including intraoperative bleeding. Various endovascular techniques can be employed to occlude the fistula such as transarterial detachable balloons;[4] stent grafts; transarterial/transvenous coiling; liquid embolics such as n-Butyl cyanoacrylate, or Onyx; and trapping the parent vessel with coils.[1] Endovascular parent artery occlusion including VVAVF is probably the simplest and most reliable treatment, but this method is difficult to perform, especially when fistulas are located in the dominant VA side. It was reported that 19% of patients whose vertebral arteries were sacrificed with coils showed neurologic symptoms.[9] Placement of stent grafts may be preferred in

these cases because it preserves flow through the treated vessel, though potential in-stent stenosis and incomplete closure due to inadequate vessel wall apposition are risks with this technique. It was reported that there were advantages of endovascular treatment using a covered stent for VVAVF because the parent artery could be preserved and the risk of exacerbation of radicular symptoms resulting from migration of the embolic material and a mass effect associated with coil embolization could be avoided,[8] although this treatment requires long-term antiplatelet therapy.

In this case, we aimed to occlude the fistula while preserving the parent artery, but avoid coil-mediated compression of nerve roots and requirement for antithrombotic therapy. In our patient, when embolizing the intervertebral vein in the intervertebral foramen and the internal vertebral venous plexus in the spinal canal, even if spinal nerve compression by the shunt pulsation or dilated vein is released, symptoms caused by spinal cord compression could remain or worsen with coil compression. Therefore, it was necessary to shorten the embolization distance and minimize coil compression. Stent grafts or covered stents can be placed to preserve the VA, but not only are there risks of long-term antiplatelet use, additional treatment for incomplete occlusion is difficult. Since the VA passes through the foramen transversarium, neck movement is transmitted directly to the VA stent. Therefore, we thought that the balloon-assisted technique would be safer because repeated neck flexion and extension may cause stent damage. We should also consider shunt flow and the blood vessels involved during coil embolization. Regarding extent of shunting, only the fistula needs to be occluded unless it is a high flow shunt, but with a high flow shunt, there is risk of coil migration. In our patient, target embolization using balloon-assisted technique was suitable considering the proximal location and sufficient space although this was a single high-flow shunt. Balloon-assisted coil embolization is straightforward, but in this case, it was particularly useful because the microballoon could control fistula flow, the catheter placed at the VVAVAF was stabilized by controlling the microballoon, and the balloon prevented coil migration. The fistula was successfully treated by target coil embolization and VA patency maintained. Therefore, balloon-assisted technique was effective in controlling the high-flow VVAVF, as first-line treatment. However, targeted therapies for different VVAVFs types require clinical classification for individualized treatment.

CONCLUSION

Our case demonstrated successful transarterial target coil embolization with balloon-assisted technique, of a spontaneous high-flow VVAVF, with MR demonstrable reversible compression of nerve roots and spinal cord. This method improved symptoms, when preservation of the VA was necessary without stents.

Declaration of patient consent

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

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Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Aljobeh A, Sorenson TJ, Bortolotti C, Cloft H, Lanzino G. Vertebral arteriovenous fistula: A review article. World Neurosurg 2019;122:1388-97.
2. Beaujeux RL, Reizine DC, Casasco A, Aymard A, Rüfenacht D, Khayata MH, *et al*. Endovascular treatment of vertebral arteriovenous fistula. Radiology 1992;183:361-7.
3. Briganti F, Tedeschi E, Leone G, Marseglia M, Cicala D, Giamundo M, *et al*. Endovascular treatment of vertebro-vertebral arteriovenous fistula. A report of three cases and literature review. Neuroradiol J 2013;26:339-46.
4. Briganti F, Tortora F, Elefante A, Volpe A, Bruno MC, Panagiotopoulos K. An unusual case of vertebral arteriovenous fistula treated with electrodetachable coil embolization. Minim Invasive Neurosurg 2004;47:386-8.
5. Edwards MK, Christenson EN, Corliss BM, Polifka AJ, Allen BR. Vertebral arteriovenous fistula: An unwelcome thrill. Case Rep Emerg Med 2017;2017:8386459.
6. Merland, JJ, Reizine, D, Riche MC, George B, Guimaraens L, Laurent A, *et al*. Endovascular treatment of vertebral arteriovenous fistulas in twenty-two patients. Ann Vasc Surg 1986;1:73-8.
7. Nikolopoulos P, Krokidis M, Spiliopoulos S, Lioupis C, Gkoutzios P, Katsanos K, *et al*. Endovascular treatment of an iatrogenic vertebrojugular fistula with a balloon-expandable covered stent: Case report and review of the literature. Perspect Vasc Surg Endovasc Ther 2012;24:149-54.
8. Okata T, Ishii A, Sadamasa N, Kai Y, Ishibashi R, Ando M, *et al*. Successful endovascular treatment with covered stent for iatrogenic vertebral arteriovenous fistula. J Neuroendovasc Ther 2018;12:29-37.
9. Shirakawa M, Nishioka T, Yamashita K, Maeda Y, Arita N. Traumatic vertebro-vertebral arteriovenous fistula manifesting as radiculopathy. Case report. Neurol Med Chir (Tokyo) 2008;48:167-70.
10. Vinchon M, Laurian C, George B, D'arrigo G, Reizine D, Aymard A, *et al*. Vertebral arteriovenous fistulas: A study of 49 cases and review of the literature. Cardiovasc Surg 1994;2:359-69.

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