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Case Report

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Cervical Radiculopathy Caused by Spinal Epidural Arteriovenous Fistula (SEDAVF) Without Intradural Drainage: A Case Report and Literature Review

Daewon Park [1], **Donghan Kim** [2], **Dong-Hun Kang** [1], **Subum Lee** [1], and **Dae-Chul Cho** [1]

[1]Department of Neurosurgery, Kyungpook National University Hospital, School of Medicine, Kyungpook National University, Daegu, Korea
[2]Department of Neurosurgery, ParkWeonWook Hospital, Busan, Korea

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Address for correspondence:
Dae-Chul Cho
Department of Neurosurgery, Kyungpook National University Hospital, School of Medicine, Kyungpook National University, 130, Dongdeok-ro, Jung-gu, Daegu 41944, Korea.
Email: dccho@knu.ac.kr

ORCID iDs
Daewon Park
https://orcid.org/0000-0003-1103-2908
Donghan Kim
https://orcid.org/0000-0001-7561-4128
Dong-Hun Kang
https://orcid.org/0000-0001-6552-3644
Subum Lee
https://orcid.org/0000-0003-4732-8137
Dae-Chul Cho
https://orcid.org/0000-0002-2899-8015

Conflict of Interest
The authors have no financial conflicts of interest.

ABSTRACT

Spinal epidural arteriovenous fistula (SEDAVF) is a rare vascular malformation. Due to the mass effect of enlarged epidural veins and venous hypertension, progressive radiculopathy and myelopathy are likely to occur. A 33-year-old female presented with right upper extremity weakness for a month. The cause of this symptom was a SEDAVF, which was located near the C5-6-7 foramens and compressed the nerve roots. In the absence of intradural venous drainage, endovascular treatment is often difficult because of the large venous pouch. We performed endovascular trapping of the vertebral artery (VA) and loose packing of the coil material on the AVF to minimize mass effects. Immediately after embolization, the fistula was occluded, but a small new feeder vessel developed a day later. An n-butyl cyanoacrylate embolization was performed, and the fistula was successfully occluded.

Keywords: Cervical spine; Arteriovenous fistula; Therapeutic embolization

INTRODUCTION

Spinal arteriovenous (AV) shunts are direct vascular connections between arteries and veins of the spine that are not through the capillary system.[12] A spinal epidural arteriovenous fistula (SEDAVF) is an entity consisting of a spinal AV shunt that primarily drains into the epidural venous plexus.[6] Spinal vascular malformations develop mainly in the thoracic and lumbar regions.

Although there is no large statistical report on cervical SEDAVFs yet, it is known that the incidence of SEDAVFs in the cervical spine is exceedingly rare.[2] Regarding SEDAVFs in the thoracic and lumbar regions, some recent reports have said that microsurgical treatment is superior to endovascular treatment for initial treatment.[3,7,13] However, a standard treatment according to various angio-architectures of spinal AV shunts has not yet been established.[13] Currently, for SEDAVFs, endovascular treatment is the favored method of treatment, with about two-thirds of studies and case reports reporting the use of endovascular treatment.[1]

In our case, which was a cervical SEDAVF, there was a large abnormal vascular pouch located in the cervical foramen, and it was difficult to achieve AVF occlusion without neurological damage through simple embolization due to mass effect from the embolic agent. As a consequence, we used vertebral artery (VA) trapping, and we would like to share this experience.

CASE REPORT

A 33-years-old female presented with right trapezius pain and right 3rd, 4th, 5th finger extension weakness for a month. There was no other pathological lesion except for a tortuous right VA in the C5–6 foramen and C6–7 extraforaminal zone that compressed the C6 and C7 right nerve root, which was revealed in cervical magnetic resonance imaging (**FIGURE 1**). Therefore, a complete cervical angiography was performed, which demonstrated that the lesion was a direct AV fistula. The left VA was the main supplying artery to brain posterior circulation, and there was small blood stilling in the right VA. The main fistula point was on the V2 segment near the C7 lateral mass (**FIGURE 2**). The lesion had paravertebral multiple venous drainages.

The patient underwent a trans-arterial embolization procedure. To prevent blood feeding from the left VA, we first performed trapping using coiling from the distal part of the right VA. Additional compressive neuropathy was possible if we filled the embolic material in the AVF directly. So, we loosely packed the AVF. Finally, we trapped the proximal VA. On final angiography, there was no AV shunt flow from the right VA angiography (**FIGURE 3**). However, a minor feeding artery from the ascending cervical artery was suspected. One day after angiography, the minor feeding artery became prominent on right subclavian angiography. Therefore, we performed n-butyl cyanoacrylate (NBCA) embolization on a small feeder from the ascending cervical artery, which is a branch of the right subclavian artery (**FIGURE 4**).

After all procedures were completed, the fistula was completely occluded and the patient's symptoms were relieved. There was no sign of recurrence at 1-year follow-up.

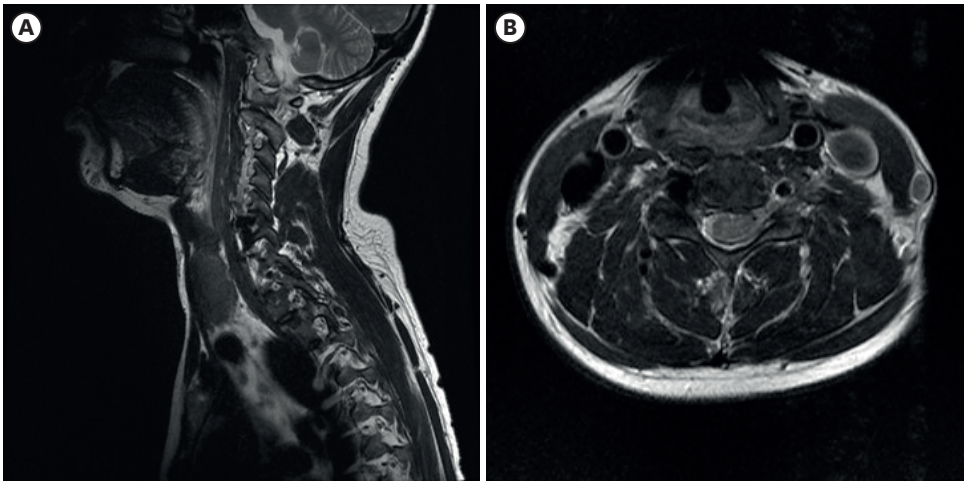

FIGURE 1. T2-weighted cervical spine magnetic resonance shows abnormal flow void on right C5–6. (A) Abnormally dilated vessel on right C5–6 foraminal zone and extraforaminal zone is seen in sagittal T2-weighted image (arrow). (B) In axial T2-weighted image, it is observed that the abnormal vessel structure occupies the way out of the C6 nerve root (arrow).

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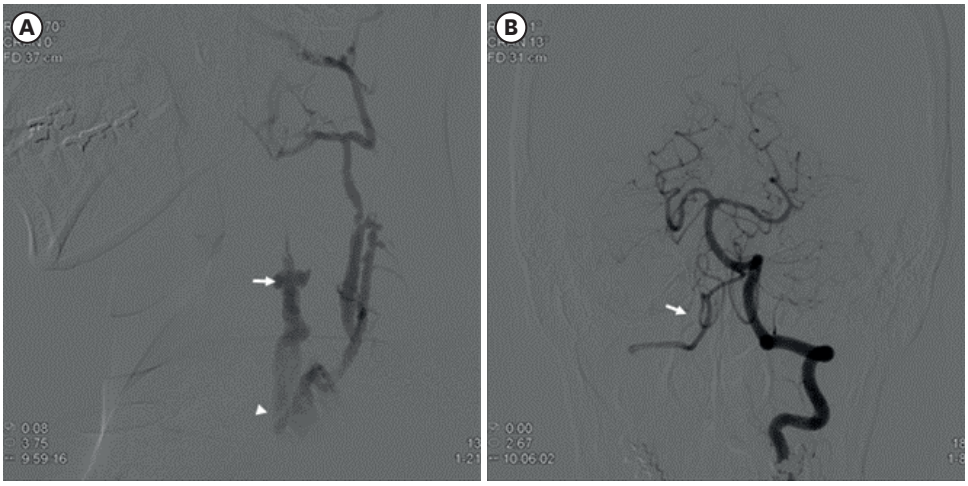

FIGURE 2. Digital subtraction angiography of the right VA. (A) The high-flow fistula occurring directly on the right VA at the C7 lateral mass level (arrowhead) and dilated vertebral vein is observed at the C5–6 level, and this area is thought to be the main cause of the patient's symptoms (arrow). (B) Distal flow in the right VA is poor beyond the fistula, but the brain posterior circulation is well maintained by the left VA, and small blood stilling is occurring to the right VA (arrow).
VA, vertebral artery.

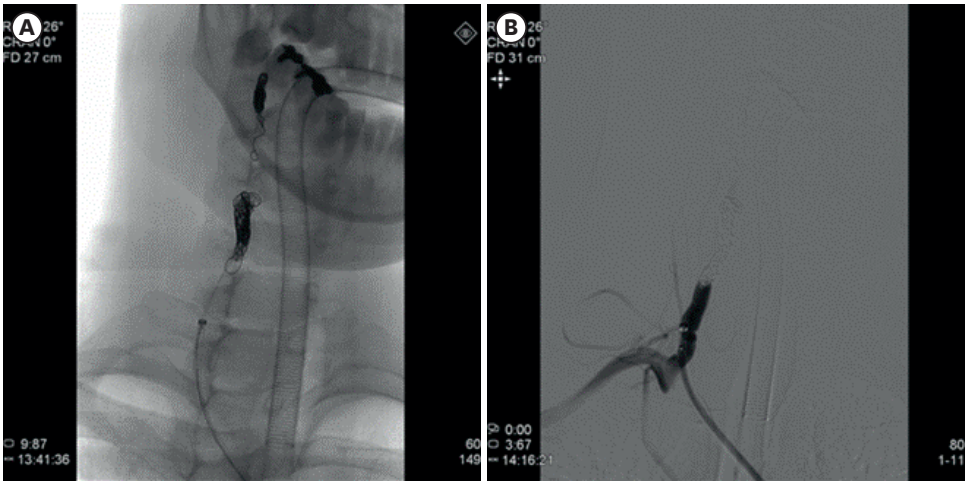

FIGURE 3. (A) The distal end of the left VA was first occluded to prevent backflow of blood from the right VA. Subsequently, the proximal part of the left VA was occluded. (B) On final angiography, arteriovenous shunt flow from the right VA was completely occluded.
VA, vertebral artery.

DISCUSSION

Spinal AV shunts are rare disease entities, which were classified by Di Chiro et al.[4] for the first time in 1967. Subsequently, several attempts have been made to classify the disease through several reports, and in 2002, the first classification of an extradural AVF was made by Spetzler et al.[10] Later, Rangel-Castilla et al.[9] proposed a classification of extradural AVFs according to the presence or absence of intradural venous drainage, which greatly helped to understand extradural AVFs. In that paper, the authors classified spinal epidural AVFs into type A and type B according to the presence or absence of intradural drainage. Our case was a type B AVF, which has no intradural venous drainage. Although standard treatment has not been established, endovascular treatment has recently been preferred for cervical SEDAVFs.[5,8,11]

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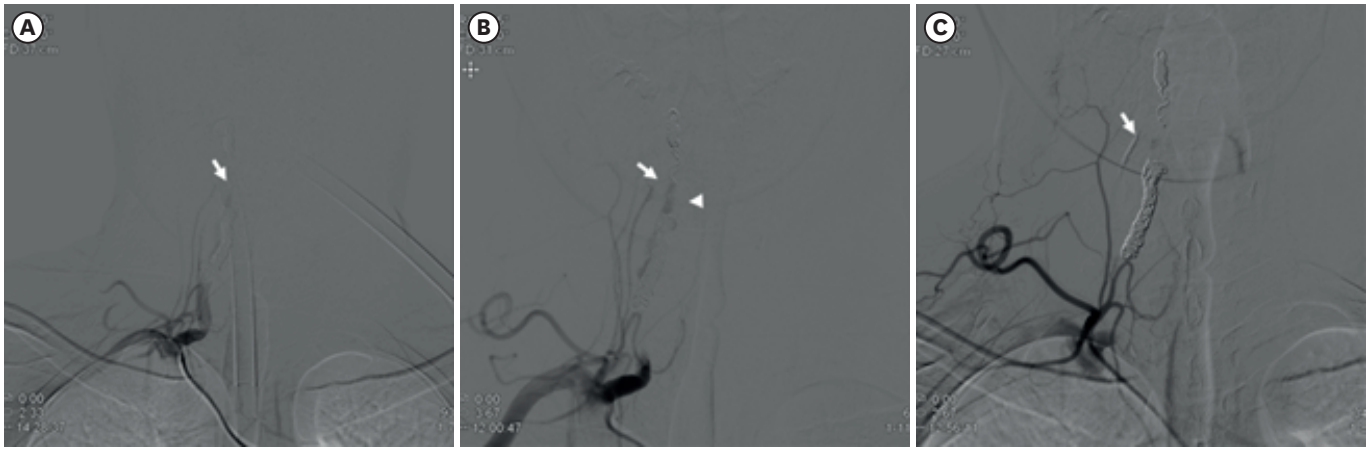

FIGURE 4. (A) Faint minor feeding arteries from the ascending VA were observed on final angiography immediately after trapping the right VA (arrow). (B) One day after angiography, minor feeders became prominent (arrow) and the AV fistula also became more prominent compared to before (arrowhead). (C) n-butyl cyanoacrylate embolization was performed, and small feeding arteries and the AV fistula were no longer observed.
VA, vertebral artery; AV, arteriovenous.

Due to the nature of this disease, which shows high blood flow, it is easy to have multiple complex anastomoses.[11] So, there may be a risk of massive bleeding if treatment is attempted with a microsurgical approach. In addition, if you block the main fistula with the high blood flow, minor feeders that are initially hidden by the main feeders can be newly recruited. Because of this, access to cervical SEDAVFs with a microsurgical approach alone may increase the likelihood of inadequate treatment.

Although there is a tendency toward choosing endovascular treatment, if we consider type A and type B separately, the case of type B, which is our case, primarily shows large venous pouches and often suffers from difficulties in achieving complete obliteration through endovascular treatment.[1] In our case as well, due to extensive venous drainage of the epidural space, there was a high possibility of incomplete obliteration for simple embolization, and there were difficulties in worrying about side effects due to the secondary mass effect of the embolic material.

In this situation, loose packing of the AVF was performed to create a scaffold for embolization. Therefore, mass effect of the coil material could be minimized. Afterward, the AVF was completely blocked by trapping the VA. This method can be considered and tried when the occluded VA is not the main blood supply source for the brain posterior circulation.

Therefore, as an initial therapeutic approach for a cervical SEDAVF, we believe that endovascular treatment will be more effective than a microsurgical approach. In the case of a type B cervical SEDAVF, endovascular treatment is sometimes difficult. However, we believe successful treatment is possible with the use of selective techniques such as VA trapping or AVF loose packing.

CONCLUSION

Cervical SEDAVF is a rare disease, and a standardized treatment has not yet been established. We performed coil embolization as an initial treatment in this case. New feeding artery recruitment occurred the following day. Total occlusion of the lesion was successfully achieved after NBCA embolization of the new feeding artery. Based on this case, we expect

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endovascular treatment to be promising as an initial approach, given the many characteristics of the disease. Further studies are needed to increase the generalizability of these findings.

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