



Case Report

Cervical extradural arteriovenous fistula with radiculopathy managed endovascularly

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ABSTRACT

Background: Extradural arteriovenous fistulas (AVFs) of the cervical spine are extremely rare, and typically manifest as slowly progressive myelopathy caused by mass effect. This is a unique case of extradural AVF of the cervical spine manifesting with purely radicular symptoms and treated endovascularly with coil and liquid embolization.

Case Description: A 55-year-old woman presented with neck pain and right upper extremity radiculopathy persisting for 9 months. Imaging studies demonstrated an AVF spanning from C4–C6 with extension into the C5–C6 foramen supplied primarily from the deep cervical branch of the costocervical trunk. The patient underwent successful coil and liquid (Onyx) embolization. Six-week postoperatively, the patient's symptoms completely resolved and magnetic resonance imaging and angiographic imaging confirmed complete obliteration of the fistula.

Conclusion: While extradural AVFs typically present with myelopathic symptoms, they may present with solely radicular symptoms and can be successfully treated endovascularly.

Keywords: Arteriovenous fistula, Cervical, Extradural, Radiculopathy, Therapeutic embolization

INTRODUCTION

Spinal arteriovenous fistula (AVF) is a rare vascular malformation with direct anastomoses between the radicular arteries and the venous plexus of the spine without intervening capillaries.^[14] Extradural AVFs, which drain primarily into the epidural venous plexus, are especially rare and account for approximately 1.6% of all spinal arteriovenous malformations.^[3] Furthermore, the incidence of extradural AVFs in the cervical spine is far less common than in the thoracic and lumbar spine, with limited cases having been reported in the literature.^[4] Extradural AVFs typically manifest as slowly progressive myelopathy caused by mass effect of the dilated internal vertebral venous plexus; and only seven cases of cervical extradural AVF manifesting with purely radicular symptoms have been reported in the literature.^[1,7,8,10-12,15] At present, there is no standard paradigm for the treatment of cervical extradural AVFs. We report a rare case of extradural AVF of the cervical spine manifesting as radiculopathy of the right upper extremity treated by coil and onyx embolization.

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CASE REPORT

Preoperative evaluation

A 55-year-old female with no significant medical history was initially evaluated because of the right-sided radiculopathy in a C6 distribution. She complained of constant mild neck pain. Her symptoms had been persistent for approximately 9 months. She had no neurological deficits on physical examination and had no signs of myelopathy. Before evaluation, she had tried physical therapy, acupuncture, oral steroids, chiropractic care, gabapentin, and topical analgesics without relief. In addition, she underwent C7–T1 interlaminar injection of steroids with no change in symptoms.

Initial noncontrast computed tomography (CT) image demonstrated a bony destructive process involving the right C5 pedicle and superior facet, concerning for an aggressive neoplastic process. Follow-up contrast-enhanced magnetic resonance imaging (MRI) demonstrated extradural enhancement along the right side at C4–C6, within the right C5–C6 neuroforamen partially insinuating into the bone. Several MRI sequences also demonstrated flow related artifact within the area of extradural enhancement [Figures 1a and b].

At this time, she had been evaluated by an orthopedic surgeon who initially planned C4–C7 anterior cervical discectomy and fusion. However, because the MRI suggested vascular lesion, neurovascular consultation and further imaging studies were instead pursued. CT angiography (CTA) was then obtained demonstrating multilevel asymmetric venous epidural enhancement spanning C4–C6 with a focal extradural venous pouch at the C5–C6 level [Figures 1c and d]. This further supported the presence of a vascular lesion, specifically an extradural AVF. Because of this, she then underwent outpatient angiography followed by embolization of the cervical foraminal AVF.

Endovascular angiography and intervention

The digital subtraction angiography (DSA) revealed an AVF supplied from the deep cervical branch of the costocervical trunk. There was a smaller contributor from the inferior thyroid branch of the thyrocervical trunk [Figure 2a].

One week after DSA, she underwent embolization of the fistula. A transradial approach was used with a Bernstein select catheter and a 256 cm 027 Headway microcatheter. The microcatheter was wedged in the distal most part of the deep cervical branch. A single coil was deployed at the neck of the venous varix (distal to the fistula) to prevent filling densely packing the varix with embolic material and preserving its mass effect. Onyx 18 was then infused upstream of the coil and allowed to disperse throughout the fistulous network in the typical fashion of “lava flow.” At the conclusion of

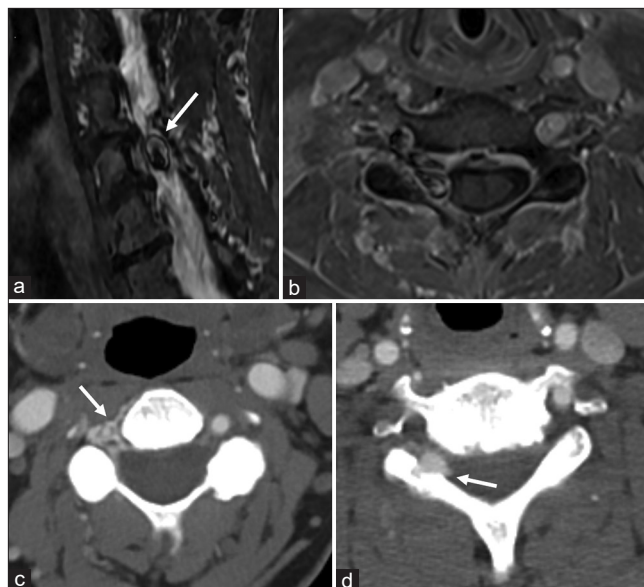


Figure 1: (a) Sagittal short T1 inversion recovery (STIR) magnetic resonance (MR) image demonstrates flow related artifact along the right lateral aspect of the spinal canal at the C5–C6 level producing a lamellated chemical shift appearance (arrow). (b) Axial volumetric T1 fat suppressed post contrast MR image at the level of the C5–C6 foramen demonstrating asymmetric extradural enhancement throughout the right C5–C6 neuroforamen and right lateral epidural space with local mass effect. Flow related artifact is also demonstrated, similar to what was seen on the sagittal STIR. (c) Axial contrast-enhanced computed tomography angiography (CTA) image at the C3–C4 level demonstrates asymmetric enhancement of the venous epidural enhancement surrounding the vertebral artery in the transverse foramen as well as along the right lateral aspect of the spinal canal (arrow). (d) Axial CTA image at the C5–C6 level demonstrates a focal extradural venous pouch, partially insinuating into the adjacent bone (arrow).

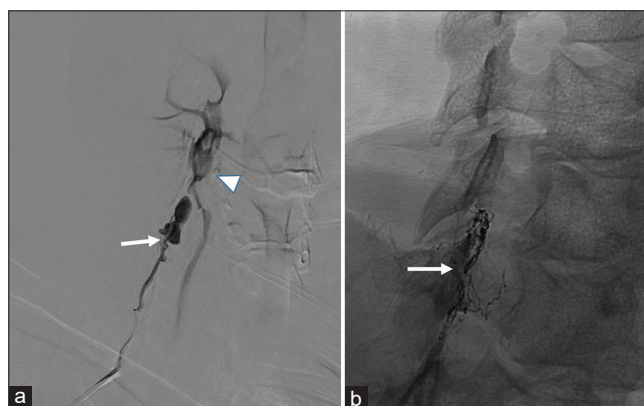


Figure 2: (a) Selective catheter angiography of the deep cervical branch of the costocervical trunk. This early/mid arterial phase shows a fistulous point (arrow) leading to two large dilated venous pouches draining early to the deep cervical venous plexus (arrowhead). (b) Native oblique image at the end of treatment showing both coil and onyx cast (arrow). Notice in this mid arterial phase, the fistula and venous pouches are no longer evident.

the embolization, the fistula appears completely obliterated [Figure 2b].

Postoperative course

Postoperatively, she was started on a short decadron taper over 5-day course. She was discharged to home the following day having met all expected postoperative milestones. At 1-week postoperatively, she had no change in her radicular pain and remained full strength. She continued to take gabapentin for pain relief but was weaning off. An MRI and CTA were repeated at 6-week postoperatively which demonstrated complete obliteration of the C6 AVF [Figures 3a and b]. Her symptoms at this time entirely resolved and she was off of all her preoperative analgesics and muscle relaxants.

DISCUSSION

Spinal AVFs are a rare disease that is direct anastomoses between the radicular arteries and venous plexus of the spine without intervening capillaries.^[14] These lesions are much more common in the thoracic and lumbar regions. Cervical AVFs have only been reported in case reports thus far in the literature and a unified consensus on the most appropriate treatment modality has not been established.^[4]

Spinal AVFs have undergone several classification schemes since they were first described in the literature in 1967 by Di Chiro *et al.*^[6,13] Extradural spinal AVFs were recently defined by Rangel-Castilla *et al.*, according to the presence (Type A) or absence (Type B) of intradural venous drainage.^[12] This variant of fistula is rarer than the classically described spinal intradural AVF but can cause significant neurological deficit. Within this classification schema, the present case falls into the Type B1, indicating compression of the neural structures associated with that particular region. In the literature thus far describing cervical extradural AVFs, there are only seven with purely radicular symptoms.^[1,7,8,10-12,15]

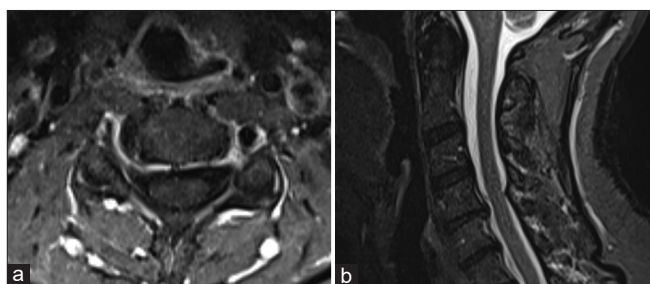


Figure 3: (a) Axial volumetric T1 fat suppressed post contrast magnetic resonance (MR) image and (b) Sagittal short T1 inversion recovery MR image demonstrate no residual venous enhancement or mass effect. The previously visualized flow-related artifact is no longer present.

Management of cervical AVFs has primarily been endovascular with a radiographic goal of complete obliteration of the fistula given the risk of recruitment of new feeding vessels with incomplete treatment. When the embolic material fills the proximal draining vein, the lesion is considered obliterated. A recent case report by Park *et al.* describes the difficulty in treating Type B fistulas that typically have a large venous pouch with endovascular therapy due to the risk of incomplete obliteration.^[5,10] In their case, they loosely packed the AVF with coils to avoid mass effect but provide support for embolic material before trapping the vertebral artery. Rangel-Castilla *et al.* highlight seven patients with eight total extradural AVFs at a single institution who underwent onyx embolization, one of which was located from C2 to T1 with the nidus at C7, supplied by the right inferior thyroid artery with associated with significant cord compression.^[12] The authors used a venous approach (the only lesion treated transvenously) with onyx and coils to prevent distal venous spread of the onyx with complete obliteration of the fistula on angiography at 1 year and without recurrent symptomatology in the patient. Certainly, this lesion could have been treated with other embolic agents. One alternative would have been to use N-butyl cyanoacrylate with an intermediate catheter situated in the parent costocervical trunk flushing Dextrose 5% in water. A vial of Onyx 18 is approximately half the cost (for single-pedicle embolization), avoids the need for an intermediate catheter, and has an appropriate viscosity to by “lava flow” penetrate the fistula to an appropriate depth. Other authors have shown similar results.^[2,9]

The present case is unique in its extradural and cervical location, the associated presenting symptoms being radicular versus myelopathic, and highlights another viable endovascular approach to these rare lesions. It is of clinical interest for vascular neurosurgeons and interventional radiologists to define these lesions and continue to add to the literature of successful treatment using endovascular approaches. In addition, general neurosurgeons and spine surgeons should also be aware of this clinical entity given that a clinical radiculopathy can indicate an underlying AVF and if imaging findings on CT/MRI suggest asymmetric extradural enhancement with flow related artifact, additional imaging studies are warranted before undertaking any decompressive efforts.

CONCLUSION

We describe a rare case of extradural AVF of the cervical spine, manifesting with purely radicular symptoms, treated with endovascular coil and Onyx embolization. Our case provides evidence of the benefits of endovascular approaches to treating extradural AVFs and highlights the potential for such cases to present with radicular symptoms as opposed to

the more commonly seen myelopathic symptoms. This is of significance to both neurosurgeons and orthopedic surgeons, as extradural AVF should be included in the differential diagnosis for patients present with radicular symptoms in their upper or lower extremities.

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.

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