

Cervical Spinal Epidural Arteriovenous Fistula with Coexisting Spinal Anterior Spinal Artery Aneurysm Presenting as Subarachnoid Hemorrhage—Case Report

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Hemorrhagic presentation of spinal epidural arteriovenous fistulas (AVFs) is rare in patients with cervical spinal vascular lesions. The present report describes a patient with cervical spine epidural AVFs associated with anterior spinal artery aneurysm at the same vertebral level presenting with subarachnoid hemorrhage. A 54-year-old man presented with sudden onset of headache. Computed tomography of the head showed subarachnoid hemorrhage. Diagnostic angiography revealed an epidural AVF located at the C1-2 level that was fed mainly by the dorsal somatic branches of the segmental arteries from the radicular artery and anterior spinal artery. This AVF drained only into the epidural veins without perimedullary venous reflux. Further, there was a 4-mm anterior spinal artery aneurysm in the vicinity of the fistula that was thought to be the cause of the hemorrhage. Endovascular transarterial fistulas embolization from the right radicular artery was performed to eliminate the AVF and to reduce hemodynamic stress on the aneurysm. No new symptoms developed after the treatment and discharged without neurological deficits. The aneurysm was noted to be reduced in size after the treatment and totally disappeared by 1 year later, according to follow-up angiography. Anterior spinal artery aneurysm from a separate vascular distribution may coexist with spinal epidural AVFs. In the setting of spinal subarachnoid hemorrhage, comprehensive imaging is indicated to rule out such lesions. **Key Words:** Aneurysm—anterior spinal artery—epidural arteriovenous fistula—subarachnoid hemorrhage—transarterial embolization.

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Spinal epidural arteriovenous fistulas (AVFs) are rare cervical spine vascular lesions, and they are generally thought to present with benign clinical symptoms, such

as radiculopathy.¹⁻³ In some cases, spinal epidural AVF can cause acute paraplegia due to reflux into medullary veins with venous congestion.⁴⁻⁷ In patients with craniocervical dural AVF, subarachnoid hemorrhage has been reported to occur because of a direct venous drainage pattern in an intracranially or intramedullary fashion.⁸⁻¹² However, hemorrhagic presentation of cervical spinal epidural AVF without intramedullary venous drainage is very rare. The present report describes a case of a patient with cervical spine epidural AVFs associated with anterior spinal artery aneurysm at the same vertebral level who presented with subarachnoid hemorrhage.

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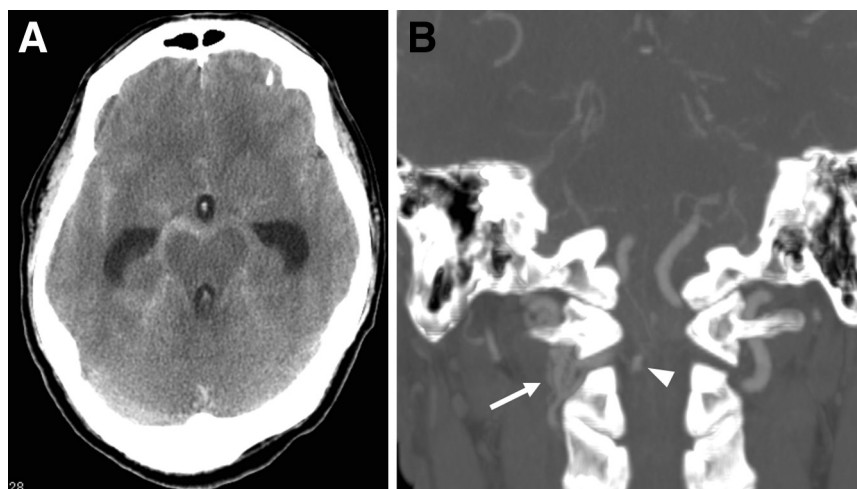


Figure 1. (A) Axial computed tomography (CT) scan of the head showing subarachnoid hemorrhage and intraventricular hemorrhage. (B) Coronal CT scan of the cervical spine with contrast demonstrating anterior spinal artery aneurysm (arrowhead) and dilated epidural veins (arrow).

Case Report

A 54-year-old man presented with sudden onset of severe headache and vomiting. A computed tomography scan of the head revealed a subarachnoid hemorrhage, mainly in the posterior fossa distribution and secondary hydrocephalus (Fig 1, A). Subsequent computed tomography angiogram of the head and neck to access for a vascular etiology revealed a vascular abnormality at the right C1/2 level and aneurysmal dilatation within the spinal canal at the C1/2 level (Fig 1, B). Magnetic resonance imaging demonstrated a signal defect and was strongly supportive of these findings.

Comprehensive cerebral and spinal angiography was subsequently performed. Injection of the vertebral artery (VA) revealed an abnormally enlarged right radicular artery at the C1/2 level and multiple thin feeding arteries from the bilateral VA to the fistula (Fig 2, A and B). Furthermore, an anterior spinal artery aneurysm (4 × 2 mm in diameter) was located anteriorly within the spinal canal in the vicinity of the fistulas at the level of the C1/2 disc. The fistulas directly drained to the epidural veins and paravertebral venous plexus. Perimedullary venous reflux was not identified (Fig 2, C). These findings were consistent with a spinal epidural AVF at the C1/2 level and an anterior spinal artery aneurysm at the same level origin of the subarachnoid hemorrhage.

We planned endovascular transarterial embolization to eliminate the fistulas and to reduce hemodynamic stress to the aneurysm, because it was deemed too difficult to access the anterior spinal artery aneurysm after discussing the case with the neurosurgical and endovascular teams. The endovascular procedure was performed 7 days after admission after obtaining written informed consent from the patient.

Under local anesthesia with systemic heparinization to maintain the activated clotting time at levels above 2- to 3-fold of the baseline value, a 7-Fr guiding catheter was inserted into the right femoral artery and guided into the right VA. Marathon flow directed microcatheters

(Covidien, Dublin, Ireland) were introduced into the main feeder as close to shunt points as possible through each guiding catheter. Right VA angiography revealed the aneurysm had enlarged (5 × 4 mm in diameter) compared with the aneurysm size seen during initial angiography (Fig 3, A). Transarterial embolization was performed from the main feeder right radial artery with a mixture of n-butyl cyanoacrylate and lipiodol. Avoiding retrograde distal migration of embolic material through other feeding arteries, balloon inflation was performed using a Hyperform balloon (Covidien) at the distal orifice of the main feeder of the right radial artery during embolization. Although transarterial embolization did not achieve complete occlusion of the fistulas due to the presence of other small feeding arteries, blood flow to the fistula was markedly reduced. Blood supply from anterior spinal artery to the fistula markedly reduced and there was an almost total disappearance of anterior spinal artery aneurysm without aneurysmal stagnant appearance (Fig 3, B).

The postoperative course was uneventful. No new neurologic symptoms developed, and ischemic lesions were not identified on diffusion-weighted images. Selective angiography performed 15 days later confirmed poor depiction of the anterior spinal artery aneurysm and fistulas. Selective angiography a year after the embolization revealed complete occlusion of the AVF and the aneurysm, preserving anterior spinal artery flow (Fig 3, C).

Discussion

Spinal epidural AVFs are vascular anomalies that preferentially affect cervical portion and that are rare compared with spinal dural AVF. Several cases of cervical spinal epidural AVFs have been published in the literature, which are summarized in Table 1. Most cases are associated with perimedullary venous reflux causing congestive myelopathy.⁸⁻¹³ Hemorrhagic presentation with subarachnoid hemorrhage has only been reported

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