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Case Report

Dural Arteriovenous Fistula of the Cervical Spine Presenting with Subarachnoid Hemorrhage

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Summary: We describe a case of dural arteriovenous fistula (DAVF) presenting with subarachnoid hemorrhage (SAH). The diagnosis of DAVF was based on spinal angiography. A review of the literature revealed that five of 13 previously reported DAVFs of the cervical spine were accompanied by SAH. SAH has not been observed in DAVFs involving other segments of the spinal canal.

Spinal dural arteriovenous fistulas (DAVFs) are usually located in the thoracolumbar region. This type of vascular malformation is predominantly found in middle-aged men with the typical presentation of progressive myelopathy (1). Spinal DAVFs in the cervical spine are rare, and, when present, usually are seen with myelopathy (2). In patients with DAVF in the posterior fossa with spinal venous drainage, there appears to be a relationship between clinical presentation and pattern of drainage (3). Patients had no myelopathy when venous drainage was limited to the cervical cord; myelopathy was present when drainage descended toward the conus medullaris (3). We report an unusual case of DAVF in the cervical spine accompanied by subarachnoid hemorrhage (SAH) and review the literature on cervical spinal DAVFs.

Case Report

A 50-year-old right-handed man with no previous pertinent medical history came to the emergency department with complaints of severe nuchal rigidity, headache, and nausea. The headache began acutely when the patient extended his neck while stretching at work earlier on the day of admission. A CT scan of the head revealed SAH in the basal cisterns and fourth ventricle and mild hydrocephalus. He was neurologically intact. Findings at cerebral angiography (consisting of bilateral common carotid and left vertebral artery injections) were normal with the exception of mild atheromatous changes in the carotid bifurcations. The left vertebral artery was selected because it was the dominant vessel. The right vertebral artery was not selectively catheterized because the left vertebral arteriogram resulted in retrograde filling of the right vertebral artery and the right posterior inferior cerebellar artery (PICA). MR images of the cervical, thoracic, and lumbar spine obtained to look for a spinal source of hemorrhage were unremarkable. The patient was observed in the hospital for 4 days

prior to discharge. He remained neurologically stable and his headache and neck pain improved. A follow-up CT scan of the head showed some resolution of the subarachnoid blood and a decrease in the size of the ventricles.

The patient returned as an outpatient 4 days after discharge from the hospital for repeat cerebral and spinal angiography to exclude cerebral aneurysm(s) and spinal vascular malformation, respectively. A right vertebral arteriogram revealed a DAVF in the cervical spine. The fistula was fed by muscular branches of the distal right vertebral artery at the C1 level, which shunted into an enlarged medullary vein on the ventral surface of the spinal cord. This abnormal vein drained toward the head (Fig 1A and B). MR imaging and MR angiography of the craniocervical junction level did not show abnormal vascularity or hemorrhage within the spinal cord but the MR angiogram did reveal flow-related enhancement in tiny serpentine structures on the ventral surface of the upper cervical spinal cord. The patient subsequently underwent right occipital craniectomy, right C1 laminectomy, and electrocautery obliteration of the DAVF without complications. A follow-up angiogram 1 month after surgery showed no residual fistula (Fig 1C and D).

Discussion

DAVF in the upper cervical spine is unusual; our patient came to medical attention because of sudden onset of headache and neck pain resulting from SAH. Although spinal DAVF is a rare cause of intracranial SAH, this entity must be considered when initial angiographic findings for nontraumatic SAH are negative. Our case also demonstrates the usefulness of selective angiography of both vertebral arteries in the search for a cause of SAH. DAVFs or pial arteriovenous malformations of the cervicomedullary junction have been reported to be a frequently unrecognized cause of SAH and may go unnoticed unless both vertebral arteries are studied (4). Therefore, to do a complete angiographic workup of SAH, one must visualize all segments of all intradural arteries, including both vertebral arteries. Visualization of both PICAs from a unilateral vertebral arteriogram should be considered inadequate if no source for the SAH has been found.

The majority of patients with spinal DAVFs are middle-aged men who initially present with pain, weakness, and sensory disturbances (1, 5, 6). The symptoms are usually gradual in onset and slowly progressive and have been attributed to increased venous pressure, which leads to passive congestion in the spinal cord. Most spinal DAVFs are found in the thoracolumbar region. Spinal DAVFs in the cervical region are rare and most of these also pres-

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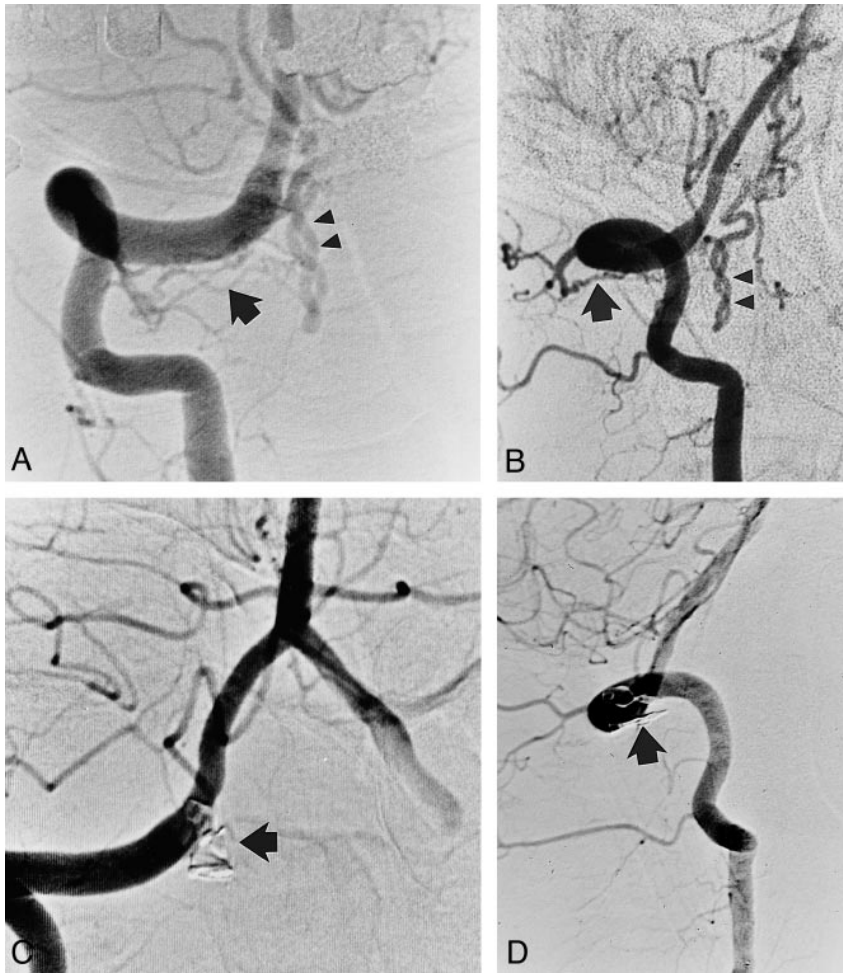


FIG 1. 50-year-old man with severe nuchal rigidity, headache, and nausea.

A and B, Anteroposterior (A) and lateral (B) angiograms show a DAVF fed by multiple small muscular branches of the distal vertebral artery (arrows) at the C1 level. There is early opacification of an enlarged tortuous medullary vein (arrowheads) on the ventral surface of the spinal cord draining cephalad.

C and D, Follow-up anteroposterior (A) and lateral (B) right vertebral angiograms show no residual DAVF. Metallic surgical clips are present at the site of the fistula (arrows).

ent with myelopathy (2). Our case fits the typical demographic pattern of patients with spinal DAVF but with the atypical clinical presentation of SAH. He did not have myelopathy. Kohno et al (2) reviewed the literature and found 12 reported cases of cervical DAVF. Of these cases, one had isolated cervical radiculopathy, six had isolated myelopathy, one had myelopathy and intradural hemorrhage, one had myelopathy and SAH, and three had isolated SAH. MR imaging is helpful to exclude cord hemorrhage and abnormal intramedullary vessels, but as is the case for other dural vascular malformations, is not sensitive for their detection.

Similar to progressive myelopathy, hemorrhage from spinal DAVFs is thought to be caused by venous hypertension. Venous hypertension occurs when arterialized blood enters the medullary vein and reaches the valveless coronal venous plexus and radial vein (7). To our knowledge, only four cases of cervical DAVF presenting with SAH have been reported (8–11). Cahan et al (8) reported a case of a 19-year-old man with SAH in whom angiography showed a high-flow fistula from the right C5 radicular artery to both the epidural and coronal venous systems that was treated with a single detachable silicone balloon. This case was considered unusual in that the patient was younger than most patients with

this disease, that the high-flow lesion was analogous to a single-hole vertebrovertebral fistula, and that venous drainage was both intradural and extradural.

Willinsky et al (9) reviewed four patients with dural vascular malformations in the cervical spine. Two were at the foramen magnum and two were in the lower cervical region. One of these patients was a 36-year-old man with SAH who had blood in the cisterna magna and weakness of the right arm and leg. The initial cerebral angiogram was negative. Over the next 7 months, the patient developed left arm pain, progressive quadraparesis, and loss of bowel and bladder functions. A repeat angiogram finally revealed a DAVF on the right C8 nerve root sleeve. Initial attempts at embolization of the arterial feeders with *n*-butyl cyanoacrylate and surgical ligation of the proximal vertebral artery failed. Cure was achieved with surgical clipping of the intradural draining vein.

Morimoto et al (10) described a 61-year-old man who had severe occipital headache, neck pain, and bilateral shoulder and arm dysesthesia 24 days before a definitive diagnosis was made. Findings at initial head CT, cerebral angiography, and myelography were negative. A lumbar puncture disclosed bloody CSF with elevated protein and sugar levels. A second angiogram including injections of both

carotid and vertebral arteries revealed a DAVF at the C5 level with arterial feeders emanating from the right vertebral artery. This lesion was effectively treated with surgical ligation of the arterialized draining vein.

Ikeda et al (11) reported a 62-year-old woman with severe headache and loss of consciousness in whom a head CT scan revealed SAH. No abnormal vascular lesions were found on the first cerebral angiogram. The second cerebral angiogram showed a DAVF at the C2 level. The fistula was fed by a branch of the distal right vertebral artery and drained by the epidural vertebral venous plexus and a medullary vein with varix formation. Cure was achieved with surgical ligation of the feeding artery and coating of the draining varix complex.

Five (38%) of 13 reported patients with DAVF in the cervical spine (including our case) were noted to present with SAH. This is in contradistinction to DAVFs located more inferiorly. Berenstein and Lasjaunias (1) reviewed 172 cases of spinal DAVF that were all located in the thoracic, lumbar, or sacral areas. None of these patients had SAH either as the initial symptom or the symptom at the time of diagnosis. Why this difference exists is unclear. Nevertheless, it appears that DAVFs in the cervical region are more prone to present with SAH than those in thoracolumbar regions. Myelopathy remains the dominant presenting symptom in both groups of DAVF.

Of the four previously described cases of cervical DAVF presenting with SAH, three had negative findings on the initial cerebral angiogram (9–11). Repeat angiography in these patients ultimately revealed DAVFs. It is unclear whether the bilateral carotid and vertebral arteries were studied on the initial angiograms, but Morimoto et al (10) did mention that their second angiogram included injections of all four vessels. It is also interesting to note that these cases had DAVFs supplied by branches of the right vertebral artery (C8, C5, and C2 levels, respectively). These lesions would certainly be missed on “routine” cerebral angiograms,

in which a right vertebral injection would frequently be omitted if there was retrograde filling of the distal right vertebral artery to the level of the PICA on a selective left vertebral injection.

Conclusion

The proper angiographic protocol for evaluation of a suspected cervical DAVF should include selective injections of both external carotid and vertebral arteries and both thyrocervical and costo-cervical trunks.

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