A cervical dural arteriovenous fistula in a patient presenting with radiculopathy

Case report

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A 51-year-old man presenting with radiculopathy with a rare cervical dural arteriovenous fistula (AVF) is reported. Angiography revealed that the cervical dural AVF was fed mainly by the left C-3 and C-4 radicular arteries and drained into the internal vertebral venous plexus with no communication with intradural structures. The dural AVF was treated surgically after embolization therapy. Although the AVF showed mass effect on computerized tomography (CT) scanning, abnormal vessels, which were suspected to drain the AVF, were observed intraoperatively to compress the left C-4 and C-5 nerve root sleeves. After resection of these abnormal epidural vessels, monoparesis of the left proximal upper extremity was markedly improved. In this patient, dynamic CT scanning was useful in the initial diagnosis, and the preoperative embolization therapy was very effective.

KEY WORDS • cervical dural arteriovenous fistula • arteriovenous malformation • radiculopathy • embolization • surgery • dynamic computerized tomography

S _pineal _dural arteriovenous fistulas (AVFs) are usually found in the thoracolumbar region in middle-aged men and are rarely found in the cervical region. In contrast to those with thoracolumbar dural AVFs who usually present with myelopathy only, patients with cervical dural AVFs frequently present with myelopathy or subarachnoid hemorrhage. To our knowledge, there is only one previously reported case of cervical dural AVF presenting with radiculopathy. We report the unusual intraoperative findings in our patient and review the reported cases of cervical dural AVF.

**Case Report**

This 51-year-old man was admitted to our service complaining of weakness of the left upper extremity, with onset on the day following golf practice 2 months prior to admission.

**Examination.** Neurological examination revealed paresis of the left deltoid, biceps and brachioradial muscles, and atrophy of the left deltoid muscle, as well as hypoesthesia and hypealgesia of the C4–6 dermatomes in the left upper extremity. The deep tendon reflexes of the left biceps and brachioradial muscles were diminished, and those of the bilateral lower extremities were exaggerated without Babinski’s sign. Cervical bruit was not recognized, although the patient was aware of a pulsatile murmur. Plain computerized tomography (CT) scanning revealed a slightly high density mass at the level of C3–C4 extending from the left ventrolateral epidural space posteriorly (Fig. 1). Considering the irregular shape of the epidural mass, we suspected a well-developed epidural venous plexus, and dynamic CT was performed. On dynamic CT scan, the peak Hounsfield unit value of the epidural mass was noted in the arterial phase (Fig. 2), and the mass was suspected to be an arteriovenous malformation (AVM). Angiography disclosed a cervical dural AVF fed by the C2–4 radicular arteries branching from the left vertebral, costocervical, and thyrocervical arteries, draining into the internal jugular vein and muscular veins via the internal vertebral venous plexus without communication with intradural structures (Fig. 3). Abnormal epidural vessels that were suspected to be the mass lesion observed on the CT scan were recognized at the C2/3–4/5 level.

**Embolization.** We planned to treat this lesion with a combination of preoperative embolization and surgical resection. The left C-3 radicular artery, the main feeding vessel of this fistula, was occluded with N-butyl cyano-
acrylate and straight platinum coils. After embolization therapy, the blood flow through the fistula was markedly reduced (Fig. 4), and the epidural mass visualized on CT scanning was diminished in volume, followed by neurological improvement and disappearance of the subjective pulsatile murmur. After embolization, the symptoms showed a tendency to improve; however, surgery was performed as scheduled 12 days after embolization to obtain further improvement and to avoid the risk of recanalization.

Operation. The patient was placed prone and a left hemilaminectomy of C2–5 with foraminotomy of C2/3, C3/4, and C4/5 was performed. The C-4 and C-5 nerve root sleeves were covered by abnormal vessels and could not be identified (Fig. 5). After dissection, they were found to be compressed by a few abnormal vessels, and they were exposed and released by resection of these vessels. Both of the nerve root sleeves were flattened, suggesting forceful compression by the surrounding abnormal vessels. These vessels were suspected to drain the dural AVF because one vessel arose directly from the dura mater at the level of C-3. Microscopically, no portion of the AVF or micro-AVM on the dura mater could be found near the level of C-3, perhaps due to the effect of embolization therapy. After the abnormal epidural vessels were resected extensively, the dura mater was incised at the level of C-3 and the intradural structures were inspected. No swelling of the spinal cord or enlargement of the coronal venous plexus of the dorsal spinal cord was recognized, and there was no other abnormal finding.

Postoperative Course. The patient was discharged 1 month postsurgery after an uneventful course. Neurologically, weakness of the left upper extremity was much improved and sensory disturbance in this extremity had resolved at the time of discharge. One year postsurgery, only slight paresis of the left deltoid muscle remained.

Discussion

The clinical characteristics, treatment, and outcome in 12 reported cases of cervical dural AVF or dural AVM, including our case, are summarized in Table 1. A cervical dural AVF is usually fed by a radicular artery from a vertebral artery and, via the fistula or micro-AVM on the dura mater, discharges the blood flow into the internal vertebral venous plexus. Cervical AVF with direct shunt between the vertebral artery and the internal vertebral venous plexus, without passage through the dural lesion, is thought to be another clinical entity that may be of traumatic, iatrogenic, congenital, or spontaneous origin. Although a few cases reported as spontaneous cervical AVF may in fact have been cervical dural AVF, these cases are...
excluded from the Table. The ages of the 12 patients with cervical dural AVF ranged from 9 to 62 years (mean 47.3 years), and there was marked male preponderance (nine males and three females). Myelopathy (eight cases) and hemorrhage (five cases) were common symptoms. The treatments for these lesions consisted of embolization only (six cases), surgery only (three cases), embolization and surgery (two cases, including ours) and observa-

In routine thoracolumbar spinal dural AVF, myelopathy occurs due to congestion of the spinal cord caused by the postshunt blood reflux through the radiculomedullary vein into the coronal venous plexus, which plays an anatomical role in venous drainage of the spinal cord. The radiculomedullary vein is also found in the cervical spine, and eight of the cases of cervical dural AVF had intradural communication, including five presenting with myelopathy. In all five, except one in which myelopathy was suspected to have been caused by intradural hemorrhage, myelopathy was considered to have been due to the same mechanism as the thoracolumbar dural AVF. There is another type of cervical dural AVF that lacks intradural communication, and four cases of this type have been reported including ours. In three cases, including ours, in which myelopathy was present without intradural communication, the myelopathy was considered to have been due to the mass effect of the draining veins. Willinsky, et al., reported a case of cervical dural AVF, which they diagnosed initially as spinal tumor because of the mass effect noted on preoperative CT scan and identified as AVF intraoperatively.

Only one case of cervical dural AVF presenting with radiculopathy has been reported previously. The findings were similar to those in our case, in that no intradural communication was found but improvement of symptoms was noted after embolization therapy; however, the cause of the radiculopathy in that case was not described.

Conclusions

The treatment selected for common thoracolumbar dural AVF with intradural communication is usually either surgery or embolization therapy, although recently surgery has been recommended more often because
recanalization frequently occurs after embolization. In our case, in spite of improvement of symptoms after embolization, surgical resection was performed as initially planned because of the persistent nature of the symptoms. The diagnosis of cervical dural A VF should be considered and dynamic CT scanning, which has been found to be useful in the differential diagnosis of spinal tumors, should be performed.

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**References**

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