Dural arteriovenous fistula as a late complication of upper cervical spine fracture

Case report

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The authors report an unusual case of a dural arteriovenous fistula (DAVF) in the cervical spine after a C1–2 fracture. The patient presented with a delayed epidural hematoma and quadripareisis. The DAVF was successfully treated by coil embolization and the patient made a full recovery. The possibility of a DAVF as a late complication of an upper cervical spine fracture should be considered when a patient presents with a spinal epidural hematoma.

Key words • dural arteriovenous fistula • cervical spine • fracture

Spinal DAVF is a well-known entity that typically presents with progressive myelopathy. The DAVF is often situated in the thoracolumbar region and is mainly found in middle-aged men. A spinal DAVF is almost always congenital in origin and rarely acquired, mainly due to nonhemorrhagic venous (hypertensive) congestion. A cervical DAVF is uncommon but is prone to bleed for unknown reasons. To our knowledge, a post-traumatic cervical spine DAVF complicated by epidural hematoma after a C-1 and odontoid fracture has not been previously reported.

Case Report

Presentation and Examination. This 23-year-old man was admitted after diving in shallow water. His medical history was unremarkable. Retrograde and antegrade amnesia were observed; the patient complained of modest pain in the craniocervical region, but results of neurological examination were normal.

Cervical spine radiography and CT scanning revealed a mild anterior atlantoodontoid luxation with an associated odontoid Type I fracture and a fracture through both atlantal posterior arches (Fig. 1 left).

Treatment. A halo brace and jacket were used to immobilize the neck. One week later, cervical CT scanning demonstrated good alignment of the fractures. Neurologically the patient remained stable, and the neck pain resolved.

After 7 months the halo brace was removed because of progressive consolidation at two of the three fracture sites and because of the normal neurological status. As a precaution, a Miami J hard collar (Jerome Medical, Moorestown, NJ) was prescribed for the next 1.5 months, allowing some sagittal motion but controlling torque.

Second Presentation. Suddenly, more than 8 months after the trauma, while sitting in the car as a passenger, the patient developed a rapidly progressive upper- and lower-limb weakness and paresis. On admission the patient was found to have quadripareisis with bilateral proprioceptive sensory deficits. Cervical CT scanning revealed an acute posterior epidural hematoma from C-1 to C-5, causing limited cord compression. To avoid compromising stability and because spontaneous clinical improvement occurred, the hematoma was not evacuated, and after 24 hours the neurological symptoms had resolved. Magnetic resonance imaging demonstrated the epidural hematoma with limited cord compression and with evidence of some intramedullary C3–4 edema (Fig. 1 right). Vertebral artery angiography revealed a serpiginous and slightly dilated DAVF at the posterior C-1 level. This DAVF was fed by a radiculomeningeal branch arising from the distal part of the left vertebral artery draining into enlarged medullary veins on the anterior surface of the spinal cord (Fig. 2).

Operation. After selective navigation involving a microcatheter placed into the DAVF, the patient underwent Guglielmi Detachable Coil endovascular occlusion; two different-sized screws (2/2 and 2/1) were placed, and these were followed by a short 2-mm-diameter fiber coil. The procedure resulted in total obliteration of the arterial supply to the DAVF (Fig. 2).

Abbreviations used in this paper: AVF = arteriovenous fistula; CT = computerized tomography; DAVF = dural AVF; MR = magnetic resonance.
Postoperative Course. Follow-up MR imaging demonstrated a significant decrease in size of the epidural hematoma. The patient remained neurologically stable and was discharged home.

Discussion

The unique phenomenon in our case was the association of cervical spine trauma and the development of a DA VF with the late-onset complication of an epidural hematoma. The details in this case do not fit the typical demographic pattern of spinal DA VF, which is usually found in the thoracolumbar region. Theoretically, the existence of the DA VF prior to the trauma, although an unlikely coincidence, cannot be entirely excluded because no previous neuroimaging studies had been performed. Spontaneous DA VF has been reported as congenital malformation in relation with fibromuscular dysplasia and neurofibromatosis. We hypothesize that, in our patient, the abnormal vascular communication developed de novo secondary to the cervical trauma and, thus, should be considered as an acquired posttraumatic DA VF. Several authors have reported the formation of AVFs in the cervical spine as a direct complication of infection, trauma without fracture, or an iatrogenic procedure. It is likely that the fractures at C-1 and C-2 caused microtears of the arterial wall of the affected radiculomeningeal artery and produced an AVF, or that a pseudoaneurysm formed and evolved into an AVF. Another possibility is that the primary event was a thrombosis or thrombophlebitis of the perimedullar veins that caused pathological growth of dural arteries during the organization and recanalization of the intraluminal thrombus, establishing an abnormal shunt. Finally, it is also possible that a thrombus itself caused elevation of the venous pressures due to impaired venous drainage and resulted in spontaneous occurrence of an AVF. It is known experimentally that venous hypertension without thrombosis can result in an AVF by releasing angiogenic factors in rats. In our case the abrupt neurological deterioration was caused by the epidural hematoma–induced spinal cord compression and myelopathy. Venous hypertension is believed to represent one of the

![Fig. 1.](image1.png) Left: Axial CT scan with bone window settings demonstrating the complex C-1 and C-2 fractures (arrows). Right: Sagittal T₂* gradient echo MR images demonstrating the acute posterior intraspinal epidural hematoma (arrow).

![Fig. 2.](image2.png) Selective left vertebral artery angiograms (left, center) and lateral cervical radiograph (right). Note the presence of the DA VF (left, arrow) and the result following the endovascular occlusion with coils (center, arrow). Note also the position of the coils on the lateral radiograph (right, arrow).
major responsible mechanisms for bleeding. At the time of diagnostic angiography no acute bleeding was identified. Likely, the drop in pressure in the DAVF and the compressive effect of the epidural hematoma played an important role in stopping the bleeding. The patient underwent endovascular coil embolization to prevent rebleeding and irreversible loss of function as well as to enable progressive recovery. Surgical ligation or clip placement was another treatment option. Our main reason for choosing endovascular therapy was the patient’s history of traumatic C-1 and C-2 fractures, where direct surgery would compromise stability and necessitate occiput–C2 fusion. The DAVF was successfully obliterated using microcoils. It is important to bear in mind the possibility of a DAVF as a late complication of a spinal trauma. A spontaneous spinal epidural hematoma is highly suggestive of a DAVF in a patient who has suffered previous trauma.

References


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