Hemodynamic analysis of associated extracranial atraumatic vertebral artery aneurysm and arteriovenous fistula

Case report

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A case of atraumatic arteriovenous (AV) fistula of the extracranial vertebral artery associated with an atraumatic aneurysm of the contralateral extracranial vertebral artery is reported. The fistulous lesion was excised after distal and proximal ligation of the vessel. Subsequently, the contralateral aneurysm underwent spontaneous dissolution. Seven cases of extracranial vertebral AV fistulae associated with ipsilateral vertebral artery aneurysms (four traumatic and three as part of vascular dysplastic syndromes) have been reported previously.

KEY WORDS • aneurysm • arteriovenous fistula • arteriovenous malformation • vertebral artery

EXTRACRANIAL vertebral artery disease may be occlusive, fistulous, or aneurysmal. Occlusive lesions are largely atheromatous. Traumatic arteriovenous (AV) fistulae and aneurysms of the extracranial vertebral artery are unusual, but occur more commonly than atraumatic lesions. An atraumatic extracranial vertebral AV fistula associated with a contralateral atraumatic extracranial vertebral artery aneurysm is the subject of this report.

Case Report

This 31-year-old man presented with the recent onset of progressive pulsatile right-sided tinnitus which decreased when he turned his head to either side. There was no history of trauma or associated systemic illness.

Examination. Physical examination revealed a bruit on the right side of the neck below the mandible which was unaffected by temporary carotid artery compression. Hearing and equilibrium were normal. Angiography demonstrated a high-flow AV fistula of the right vertebral artery at the C-1 level (Fig. 1 left) with prominent drainage into the epidural and paravertebral venous plexuses, the internal jugular vein, and the basilar venous plexus. There was a 7-mm aneurysm of the left vertebral artery at C2–3 (Fig. 1 right).

Operation. The patient underwent suboccipital craniectomy and upper cervical laminectomy. The right vertebral artery was ligated proximal and distal to the 2-cm vertebral AV fistula. The fistula was subsequently excised.

Postoperative Course. After surgery, the tinnitus disappeared and the neurological examination was normal. Repeat studies 6 months later demonstrated complete obliteration of the fistulous lesion, occlusion of the right vertebral artery (Fig. 2), and disappearance of the left vertebral artery aneurysm (Fig. 3).

Discussion

Atraumatic extracranial vertebral artery aneurysms may arise as solitary lesions from inflammatory processes, atherosclerotic changes, or spontaneous dissection. Congenital extracranial vertebral artery aneurysms have been reported unassociated with systemic illness or in conjunction with Ehlers-Danlos syndrome, neurofibromatosis, and fibromuscular dysplasia.

Atraumatic AV fistulae of the extracranial vertebral artery have been found in combination with fibromuscular dysplasia and neurofibromatosis. Spontaneous vertebral AV fistulae have been described in cases with
Atraumatic vertebral artery aneurysm and AV fistula

Fig. 1. Preoperative angiography studies. Left: Right vertebral artery angiogram, anteroposterior view, demonstrating an arteriovenous fistula of the vertebral artery with the abnormal vascular communication at the level of the C-1 vertebral body. Right: Left vertebral artery angiogram, anteroposterior view, showing a 7-mm aneurysm at the junction of the C-2 and C-3 vertebral bodies and pointing anteriorly and laterally. There is reflux down the right vertebral artery to the level of the fistulous communication at the C-1 vertebral body, where rapid shunting occurs into the draining veins.

Fig. 2. Postoperative right vertebral artery angiogram, anteroposterior view, revealing complete occlusion of the artery at the junction of the C-1 and C-2 vertebral bodies.

Fig. 3. Postoperative left vertebral artery angiograms, anteroposterior view (left) and lateral view (right), demonstrating complete disappearance of the aneurysm and only minimal residual irregularity of the vessel wall (arrows).

Atherosclerosis and prior infection. Congenital lesions have been reported in patients with a history of birth trauma or without such evidence. The origin of these congenital fistulae has been attributed to vertebral artery development from longitudinal anastomoses between six pairs of dorsal embryonic intersegmental arteries and its intimate association with draining epidural and muscular plexuses.

The finding of an extracranial vertebral artery aneurysm associated with an AV fistula is rare. Review of the literature reveals four such instances in which the lesions were known to be traumatic in nature and occurred homolaterally. Three cases of atraumatic extracranial vertebral artery aneurysm have been reported associated with a fistula in patients with syndromes of diffuse vascular dysplasia. There is no previous report of atraumatic extracranial vertebral AV fistula with an atraumatic aneurysm of the contralateral extracranial vertebral artery (Table 1).

The association of intracranial aneurysms and infundibula with intracranial arteriovenous malformations (AVM's) is well described. Their simultaneous occurrence has been attributed to coincidental development or to a common defect in angiogenesis. Paterson and McKissock and later Miyasaka, et al., suggested that hemodynamic changes, especially turbulent flow produced by shunting through AVM's, were responsible for the development of associated aneurysms. The location of these dilatations on vessels anatomically related to the AVM and their atypical location on vessels feeding the malformation was attributed to disturbed flow patterns of the associated AVM. Shenkin, et al., reported the production of aneurysms contralateral to internal carotid artery ligation, which similarly supports this theory.

Laboratory studies have shown that increased local blood flow leads to distension and degeneration of the arterial walls with subsequent aneurysm formation. In AVM's, diminished vascular resistance allows increased flow through feeding arteries with stretching of the vessel walls. This may lead to weakening of a previously undetected defect within the vascular wall and formation of an aneurysm.

There are few instances of aneurysms of spinal cord vessels without associated AVM's. The high incidence
of rupture of spinal AVM's in patients with aneurysms arising from spinal cord vessels reflects the abnormal hemodynamics of these malformations.46 Miyamoto, et al.,28 confirmed the presence of spinal aneurysms on the feeding vessels of spinal AVM's and demonstrated their disappearance with obliteration of the abnormal shunt.

Reports suggest that obliteration of the AV shunts and their high-pressure high-volume flow has been effective in reducing or completely eliminating associated aneurysms.5,29,51 This has been attributed to gravitational layering of thrombus51 or to intimal thickening and contraction of arteries with slower blood flow.5

In the present case, blood flowing in the right vertebral artery was shunted into the AV fistula at the C1-2 level with delayed flow into the intracranial portion of the artery. This was due to blood traveling the path of least resistance, which the fistula provided. Injection of the left vertebral artery (Fig. 1 right) revealed good filling of the intracranial segment of the vessel and rapid reflux down the right vertebral artery to the point just above the fistulous communication. Increased flow in the contralateral vertebral artery was necessary to adequately fill the vertebral artery distal to the fistula. This hyperdynamic turbulent state may have weakened the vessel wall, predisposing to dilatation and subsequent aneurysm formation. Ligature of the right vertebral artery and elimination of the fistula negated the need for increased flow through the left vertebral artery. Once flow rates returned to normal, the hemodynamic stress diminished and the left vertebral artery aneurysm disappeared.

### Table 1

<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
<th>Patient's Age &amp; Sex</th>
<th>Symptom of Bruit</th>
<th>Location of Fistula</th>
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<td>Deans, et al., 1982</td>
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References

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