Spontaneous subarachnoid hemorrhage first from an intracranial and then from a spinal arteriovenous malformation

Case report

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A patient presented with spontaneous subarachnoid hemorrhage (SAH) from a cerebral arteriovenous malformation (AVM) which was later totally removed at surgery. The patient presented again with a new SAH from a spinal AVM that was also totally removed at surgery. Coexistence of spinal and cerebral arteriovenous malformations are exceedingly rare and hemorrhage from each is not previously reported. This case emphasizes the importance of investigating the spinal canal in otherwise unexplained spontaneous SAH.

KEY WORDS • arteriovenous malformation • intracranial AVM • intraspinal AVM • spontaneous subarachnoid hemorrhage

It has been reported that an intracranial source has not been identified in 13% to 36.6% of all cases of spontaneous subarachnoid hemorrhage (SAH). The following case indicates that possibly the spinal canal should be studied more frequently in such cases. This case is unique in that the patient harbored an arteriovenous malformation (AVM) intracranially and intraspinally, each of which bled separately, and each of which was successfully removed.

Case Report

This 47-year-old man first presented to us in November, 1971, after the acute onset of a severe bifrontal headache, followed by loss of consciousness and a generalized seizure. Two similar episodes had preceded this.

First Admission. Aside from neck stiffness there were no neurological deficits. There were no cutaneous hemangiomas and no cranial or spinal bruits. Angiography revealed an AVM in the upper anterior portion of the left temporal lobe (Fig. 1 left) supplied by the posterior temporal branch of the middle cerebral artery and drained by the superficial middle cerebral vein and the superior anastomotic vein (Fig. 1 right).

First Operation. At surgery, December 6, 1971, two red dilated veins were seen on the cortical surface leading away from a widened fissure. The overlying dura and adjacent arachnoid and brain were stained dark red and yellow. The malformation was encountered about 5 mm beneath the surface. The lesion was totally excised as confirmed by postoperative angiography (Fig. 2). The postoperative course was uncomplicated and
FIG. 1. Preoperative angiogram. **Left:** Arterial phase showing the arteriovenous malformation (AVM) fed by branches of the middle cerebral artery deep in the medial upper anterior portion of the temporal lobe. **Right:** Venous phase showing the venous drainage from the AVM.

The patient was discharged with no neurological deficit.

**Second Admission.** The patient remained well until January, 1976, when he was readmitted with a spontaneous SAH. He reported the sudden onset of neck pain followed by headache. Again on examination there was neck stiffness, but no neurological deficit demonstrable. Cerebral panangiography revealed no evidence of a vascular lesion. A myelogram demonstrated an irregular tortuous filling defect extending from T-10 to L-1 (Fig. 3). Angiography with selective catheterization of the segmental arteries from T-8 to L-2 failed to visualize this malformation, thus precluding any attempt to treat by selective embolization.

**Second Operation.** A laminotomy was performed in January, 1976, from T-11 to L-1 exposing large tortuous red veins floating freely in the subarachnoid space. They arose from a small fistulous tangle of vessels densely incorporated in the cord surface over an area about 1 cm in diameter at T-12. The adjacent tissue was stained yellow. The fistula was completely excised along with the adjacent portions of the large looping veins that extended up and down beyond the extent of

FIG. 2. Postoperative angiogram showing complete obliteration of the arteriovenous malformation with preservation of the normal circulation.

FIG. 3. Myelographic picture including T-11, T-12, and L-1.
Spontaneous SAH from intracranial and spinal AVM’s

the laminotomy. The residual segments of these veins became normal in color and reduced in caliber. The connected neural arches were wired back in place. The patient was up and walking the day after surgery and at no time demonstrated any neurological deficit.

Discussion

Co-existence of an intracranial and intraspinal AVM has been reported in only five patients. Hash, et al.9 reported a 24-year-old man who presented with low-back pain and headache. The diagnosis of SAH was confirmed by lumbar puncture. Intracranial angiography illustrated an AVM on the tentorium, drained by dural vessels. A subsequent myelogram and spinal arteriogram illustrated a spinal AVM. Both lesions were treated conservatively. Di Chiro and Wener4,5 observed three patients with concurrent intracranial and intraspinal AVM’s, but reported no details as to location, treatment, or follow-up review. Krayenbühl, et al.19 and Yasargil22 observed a case of concurrent cerebellar and spinal malformation. Wyburn-Mason,20,21 who first called attention to the co-existence of multiple arteriovenous lesions, makes no mention of bleeding from an intracranial as well as a spinal lesion in one individual.

As in the case of intracranial AVM’s it has been generally established that total surgical excision is a treatment of choice.2,10,12-14 Kunc and Bret11 thought the fear of postoperative neurological deficits was exaggerated, and that most poor results in surgery were due to delay, with progression of the lesion due to dilation of collateral vessels and deterioration of the intrinsic circulation to the cord. Krayenbühl, et al.19 and Yasargil22 who have an extensive experience with these lesions believe that a patient who survives the initial hemorrhage without suffering a transverse myelitis, or death, is always under constant threat of both, if untreated.

Bailey and Sperl9 believed that the spinal cord circulation was not dependent upon the vessels feeding the malformation. This is an important factor in the consideration of surgical management. A final feeding artery can never nourish tissue but can always steal from a branch that is nourishing tissue.18

As far as we know this is the only reported case of spontaneous hemorrhage from first an intracranial AVM and then a spinal AVM, followed by removal of each in turn. We would again stress the advisability of myelography in the investigation of spontaneous SAH in patients with no demonstrable intracranial source.1,8,19

References


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