Dural-Pial Arteriovenous Malformation After Sinus Thrombosis

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Background—We report an unusual case of acquired dural-pial arteriovenous malformation (AVM) following sinus thrombosis.

Case Description—Initial angiography performed in a 39-year-old man showed thrombosis of the superior sagittal sinus (SSS) and the right transverse sinus (TS) but no vascular malformations. Follow-up angiography 29 months later revealed recanalization of the SSS and the TS, retrograde cortical venous drainage which suggested that thrombosis of the sinuses probably propagated into the adjacent parietal cortical veins, and development of a dural-pial AVM at or near the site of thrombi in more than one cortical vein. Complete surgical excision of the lesion was accomplished without neurological deterioration.

Conclusions—The present case suggests the possibility that the pial AVM is not only a congenital condition but also may develop as an acquired lesion. (Stroke. 1998;29:1721-1724.)

Key Words: angiogenesis ■ cerebral arteriovenous malformations ■ sinus thrombosis

Dural arteriovenous malformations (AVMs) appear to be acquired rather than congenital lesions, and it is well known that they often develop at the site of a sinus thrombosis.1,2,3,4 On the other hand, pial AVMs are generally considered to be congenital malformations. We report a case of acquired dural-pial AVM following superior sagittal and transverse sinus thrombosis and discuss the causes both of the unusual location of the dural AVM and of the development of the pial lesion in this case.

Case Report

The patient was a 39-year-old man who, on July 27, 1994, experienced headache and vomiting, followed 2 days later by weakness in the upper part of his left arm. He was hospitalized 4 days later, and neurological examinations indicated left hemiparesis.

CT scan revealed a low-density area with a mass effect in the right frontal lobe (Figure 1), and on enhanced CT an empty delta sign was observed in the superior sagittal sinus (SSS). MRI showed sinus thrombosis of the SSS and the right transverse sinus (TS). However, MRI revealed no abnormal findings suggestive of a vascular malformation. Because no image of the anterior half of the SSS or the right TS could be detected in angiograms of the right common carotid artery, a diagnosis of sinus thrombosis of the SSS and the right TS was made (Figure 2A and 2B). Nevertheless, no dural or pial AVMs were found (Figure 2C and 2D).

Immediately after admission, anticoagulant therapy was initiated. Both the headache and the left hemiparesis improved gradually and since, there were no clearly abnormal findings on neurological examination other than a slight exaggeration of the deep tendon reflex on the left side, the patient was discharged. Although MRI performed on September 27, 1996, revealed recanalization of the SSS and the right TS, at the same time a large number of flow void signals

Figure 1. Axial CT scan without contrast enhancement showing a low-density area with mass effect in the right frontal lobe.
suggestive of a vascular malformation made their appearance in the right parietal lobe. On December 19 of the same year, right selective internal carotid angiography demonstrated a pial AVM in the right parietal region, supplied by branches from the middle cerebral artery and draining into the cortical veins (Figure 3A and 3B). It was also noted that the SSS was patent but there was a reflux of blood into the cortical veins, and the venous routes between the cortical veins and the SSS were occluded (Figure 3C). Meanwhile, right selective external carotid angiography revealed a dural AVM supplied by the superficial temporal artery and the middle meningeal arteries and also draining into the cortical veins (Figure 3D).

**Operative and Postoperative Findings**

After a negative result was obtained in a provocation test conducted on March 24, 1996, before direct surgery, the feeder of the pial AVM was selectively occluded with use of a liquid coil (Target Therapeutics), and a parieto-occipital craniotomy was performed. The superficial temporal artery was found to pierce the cranium, becoming a feeding artery. When the dura mater was opened, several draining veins that were red veins were observed on the surface of the cerebrum. These draining veins on the cerebral surface formed vascular connections together with the dural AVM fed by the middle meningeal arteries at 2 sites (Figure 4). A section of the dura

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**Figure 2.** A and B, Right common carotid angiograms showing 15° oblique anteroposterior (A) and lateral (B) views of the venous phase. Occlusion of the superior sagittal sinus and the right transverse sinus is demonstrated. C and D, Right common carotid angiograms of the lateral (C) and anteroposterior (D) views of the arterial phase, demonstrating no vascular malformations.
measuring 4×6 cm was resected together with a dural AVM and the connections of the cortical vein. Many of the vessels seen on histological examination were dilated and appeared to be dysplastic, which confirmed the presence of an AVM in the dura.

In addition, the pial AVM, whose feeding arteries were 3 branches of the middle cerebral artery, was seen surrounding the same superficial draining veins; we removed these structures in their entirety. The structures removed were found histologically to be pial AVM tissue. The resulting defect of the dura was repaired with the epicranial aponeurosis beneath the skin flap with watertight suturing.

Postoperative selective internal and external carotid angiography revealed no vascular malformation. The postoperative course was uneventful, and there was no deterioration in the patient’s neurological condition.

Figure 3. A and B, Selective right internal carotid angiograms of the lateral (A) and anteroposterior (B) views of the arterial phase, demonstrating a pial AVM supplied by branches from the middle cerebral artery and draining to the cortical vein. C, Right internal carotid angiogram of the lateral view of the venous phase, demonstrating the patency of the superior sagittal sinus. D, Selective right external carotid angiograms of the lateral view of the arterial phase, demonstrating a pial AVM fed by the superficial temporal and middle meningeal arteries and draining to the cortical vein.
Role in the development of the dural AVM in this case. Thrombosis of a cortical vein may have played an important role in the development of the dural AVM in this case. Increased cortical venous pressure persisted. Therefore, it is thought to have newly formed after a sinus thrombosis. Lyons et al. reported that angiogenic growth factors such as basic fibroblast growth factor appeared in the cerebral tissue surrounding an area of cerebral ischemia. In our case, the pial AVM occurred adjacent to the posterior part of the ischemic region (Figure 1), an area in which obstruction of the connections between the cortical veins and the sinus was present and a reduction of cerebral perfusion due to the increased cortical venous pressure persisted. Thus, the possibility that the increased expression of angiogenic factors and the resulting neovascularization played a part in the development of an acquired pial AVM was suggested.

Kader et al. recently described pediatric cases in which pial AVMs occurred after total resection of the original lesions had been verified, and they have begun to cast doubt on the theory of cerebral AVMs as congenital anomalies. The present case also is significant in that it calls into question the theory that a pial AVM is an exclusively congenital phenomenon.

Ravens examined anastomoses in the vascular bed of the human brain. According to his findings, arteriovenous anastomoses were present in both the cerebral cortex and white matter in the normal human brain.

We suggest the possibility that the pial AVM in the present case developed because closed arteriovenous fistulas that were originally present somehow became patent when cerebral ischemia occurred.

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**References**

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