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...
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

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.
thrombosis of a cortical vein may have played an important role. Therefore, it was considered that cortical venous hypertension due to sinus thrombosis. 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 recurred after total resection of the original lesions had been verified, and they have begun to cast doubt on the theory of congenital AVMs as congenital phenomenon. The present case also is significant in that it calls into question the theory that a pial AVM is exclusively congenital in nature.

Ravens examined anastomoses in the vascular bed of the human cerebrum. According to his findings, arteriovenous anastomoses were present in both the cerebral cortex and subcortical 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 patent somehow became patent when cerebral ischemia occurred.

Discussion

Newton and Cronqvist classify intracranial AVMs into 3 types on the basis of their arterial supply: pure pial, mixed pial and dural, and pure dural. Mixed pial and dural malformations receive their blood supply not only from cerebral or cerebellar arteries but also from meningeal vessels. Therefore, the vascular malformation presented here was termed a dural-pial type. It has been emphasized that development of dural AVMs correlates positively with venous hypertension due to sinus thrombosis. Usually, dural AVMs develop adjacent to a thrombosed sinus. The present case was unique in that the dural component of the AVM was not located at the site of a sinus thrombosis. Although the SSS and the TS were recanalized, as shown in Figure 3C, retrograde cortical venous drainage was found. This suggests that thrombosis of the SSS and the TS probably propagated into the adjacent cortical veins, that the connections between the cortical veins and the sinuses had been obliterated, and that increased cortical venous pressure persisted. Therefore, it was considered that cortical venous hypertension due to thrombosis of a cortical vein may have played an important role in the development of the dural AVM in this case.

Kerber and Newton suggest that minute arteriovenous shunts, which are normally present within the dura mater, might enlarge in the presence of increased venous pressure and lead to angiographically significant arteriovenous shunting. Furthermore, Lawton et al speculate that an increased cortical venous pressure induces angiogenic activity either directly or indirectly by decreasing cerebral perfusion and decreasing ischemia and that dural AVM formation may be the result of aberrant angiogenesis.

On the other hand, pial AVMs are generally considered congenital vascular malformations. Indeed, there are very few studies (only 3 relevant reports could be found) in which the presence of a pial AVM is ruled out in initial angiography but is later discovered to have developed. However, the present case is the first reported in which a pial AVM 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.

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References

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