Dural-pial Arteriovenous Malformation with Unusual Venous Drainage

—Case Report—

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Abstract

A 41-year-old female presented with an uncommon mixed dural-pial arteriovenous malformation (AVM) with unusual venous drainage. Angiography at the initial operation for massive intracerebral hematoma in the parietal lobe showed no vascular malformations. Follow-up angiography 3 years later revealed a mixed dural-pial AVM at the previous surgical site. Unexpectedly, the venous drainage from the AVM was not into the patent superior sagittal sinus, but was retrograde into the contralateral cortical veins in the occipital lobe. No skull fracture or sinus thrombosis was demonstrated. The AVM was resected successfully without neurological deterioration. The pial portion of the lesion may have been a congenital anomaly, and the dural portion acquired. The development of a dural AVM at an earlier surgical site is uncommon, and may indicate the possible pathogenesis of acquired dural AVMs unrelated to sinus occlusion or accidental trauma. Furthermore, there are various possible patterns of venous drainage in vascular malformations.

Key words: cerebral arteriovenous malformation, dural arteriovenous malformation, cerebral vein, postoperative complication

Introduction

Arteriovenous malformations (AVMs) may be classified as pure pial, mixed pial and dural, or pure dural types.^{1,2)} Pial AVMs are generally thought to be developmental malformations,⁸⁾ whereas dural AVMs may be either congenital or acquired. Acquired dural AVMs are well known to usually develop secondary to sinus thrombosis or trauma.^{3,5,6)}

We present an uncommon case of a mixed dural-pial AVM with unusual, contralateral venous drainage occurring at the craniotomy site, with no associated sinus thrombosis or accidental trauma.

Case Report

A 41-year-old female was first admitted to Kitasato University Hospital on December 8, 1990. Four

hours before admission, she had suffered sudden onset of severe headache and vomiting.

Neurological examination on admission revealed symptoms of transtentorial herniation accompanied by a semicomatous state, anisocoria (right > left), and decerebrate posture in the left extremities. Computed tomography (CT) showed a massive intracerebral hematoma in the right parietal lobe (Fig. 1). Emergency right common carotid angiography showed no dural and pial AVMs (Fig. 2). Laboratory studies showed no evidence of risk factors for hemorrhage.

Emergency right parieto-occipital craniectomy was undertaken to remove the hematoma on the day of admission. No dural or pial AVM-like lesions were demonstrated. At the end of the operation, the dura was not closed, and a homologous lyophilized dura mater (Lyodura; B. Braun Melsungen AG, Melsungen, Germany) was placed over the dural and bone defect. The muscular, subcutaneous, and cutaneous layers were closed in the usual fashion.

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Fig. 1 Precontrast CT scan at the first admission, showing an intracerebral hematoma in the right parietal lobe.

Fig. 3 Postcontrast CT scan on September 29, 1993, demonstrating marked linear enhancement in the left parieto-occipital lobe.

Fig. 2 Preoperative right common carotid angiograms, lateral view of the arterial (*upper*) and venous (*lower*) phases, showing no dural or pial AVMs.

She became alert 14 days after the operation. On January 4, 1991, cranioplasty using a resinous material (methyl methacrylate; Codman, Blackpool, U.K.)

was performed without complications. Neurological examination at discharge on January 21, 1991 showed slight left hemiparesis, left lower quadrant hemianopsia, and slight right parietal lobe symptoms. On September 29, 1993, she suffered a grand mal seizure. Postcontrast CT showed an unexpected abnormal enhanced area in the left occipital lobe (Fig. 3).

Neurological examination on October 22, 1993 found left lower quadrant hemianopsia and the slight right parietal lobe symptoms that had been present previously. Right selective internal carotid angiography demonstrated a small pial AVM in the right parietal lobe supplied by branches from the middle cerebral artery, and draining into the cortical vein (Fig. 4). Right selective external carotid angiography showed a dural AVM supplied by the superficial temporal and middle meningeal arteries, and draining into the cortical vein, which also drained the pial AVM (Fig. 5). Unexpectedly, the venous drainage from the dural-pial AVM was not into the superior sagittal sinus, but was retrograde into the contralateral cortical veins in the occipital lobe (Figs. 4 and 5). This retrograde flow into the contralateral veins was compatible with the abnormal enhancement on the CT scans. Left internal carotid angiography demonstrated the patency of the superior sagittal sinus (Fig. 6).

On December 7, 1993, a second operation was undertaken. The previous skin incision was opened and the bone flap was removed. The superficial temporal artery was transected at the posterior rim of the zyg-

Fig. 4 Selective right internal carotid angiograms at the second admission, lateral (*left column*) and anteroposterior (*right column*) views of the arterial (*upper row*) and venous (*lower row*) phases, showing a pial AVM supplied by branches from the middle cerebral artery, and draining into the cortical vein. Note the retrograde venous drainage into the contralateral cortical veins.

Fig. 5 Selective right external carotid angiograms at the second admission, lateral (*left*) and anteroposterior (*right*) views of the arterial phase, demonstrating a dural AVM fed by the superficial temporal and the middle meningeal arteries, and draining into the cortical vein.

Fig. 6 Left internal carotid angiograms at the second admission, lateral (*left*) and anteroposterior (*right*) views of the venous phase, showing the patency of the superior sagittal sinus. No septum in the parietal or occipital superior sagittal sinus is seen.

matic arch. The dura was markedly thickened and contained abnormal vessels. There was dense adhesion between the dura and the cerebral cortex, and a rich vascular network including the draining vein between the dura and the surface of the cortex. The entire dura over the craniotomy was resected. A cluster of abnormal vessels in the parietal cerebral cortex supplied by small branches from the middle cerebral artery was removed. Histological examination of the specimens of the dura and the parietal cerebral cortex showed a dural-pial AVM.

Postoperatively, her neurological condition did not deteriorate. Postoperative selective internal and external carotid angiography 18 days after the operation showed no vascular malformations.

Discussion

Our patient had none of the risk factors for intracerebral hemorrhage, such as systemic hypertension, clotting disorders, neoplasms, arteritis, or venous thrombosis. We concluded that the pial vascular malformation was the most likely cause of the intracerebral hematoma. Since pial AVM is a congenital anomaly,⁸⁾ we must consider why the initial angiograms in this patient failed to show the pial vascular malformation. Previous cases have demonstrated

that a pial AVM not revealed by the initial angiography can be visualized during the follow-up period.^{16,17)} This was probably due to compression of the AVMs by the hematoma or to incomplete thrombosis with later recanalization.^{13,17,19)}

In our patient, initial common carotid angiograms and the initial surgery showed no dural vascular malformations. Superselective external carotid angiography is necessary to exclude completely the possibility that a dural AVM was present at the time of the initial operation. Therefore, the present case showed at least marked enlargement of a dural AVM at the site of the previous operation.

Dural AVMs may be congenital or acquired. Acquired dural AVMs usually develop in association with sinus thrombosis or accidental trauma.^{3,5,6,11)} Our case is a unique example of a delayed postoperative dural AVM which developed at the site of earlier surgery and was unrelated to these well-known causes. Only two cases of a dural AVM occurring at the site of a previous craniotomy have been reported.¹¹⁾ The blood vessels and microvascular shunts in the scalp at the surgical site may progressively develop fistulous communication with increasing recruitment of the dural vasculature. These microvascular arteriovenous channels occur normally and are not symptomatic or readily visualized until they become larger, with a much greater blood supply.^{11,14)} The greater arterial supply provided by the blood vessels of the scalp and muscle blood vessels then lead to the development of a dural AVM.¹¹⁾

The congenital pial AVM may also have been involved in the development of the dural malformation. Anastomotic pathways between the dural meningeal and cortical leptomeningeal arteries ("rete-mirabile anastomoses") occur as collateral anastomoses in patients with cerebrovascular occlusions.^{10,18)} Similar collateral contributions from dural meningeal arteries occur with parenchymal AVMs which were initially supplied only by pial arteries.¹²⁾ Russell and Berenstein¹⁵⁾ reported a cerebral AVM supplied by the middle cerebral artery in which normal branches of the middle cerebral artery were filled from the middle meningeal artery. Dahl and Kline⁴⁾ and Miyachi *et al.*⁹⁾ found that the contributions of the meningeal arteries to mixed pial and dural AVMs were more common in adults. These results suggest that the presence of a congenital pial AVM may be involved in the development of the dural vascular malformation in our case.

Parasagittal pial and dural AVMs drain into the superior sagittal sinus except in cases of sinus thrombosis. In our patient, the superior sagittal sinus was patent, but was not involved in the venous drainage.

The pathophysiology of the anomalous and unusual venous drainage to the opposite side in the present AVM is uncertain. The main venous drainage of the original malformation involving both the ipsilateral parasagittal cortical veins and the superior sagittal sinus may have been ruptured at the time of the initial hemorrhage. This would interrupt the route between the parasagittal cortical veins on the AVM side and the superior sagittal sinus, and the new venous drainage system to the opposite side was probably produced when the pial malformation reopened and the dural AVM developed. Another possibility is a septum in the parietal or occipital superior sagittal sinus. The right channel could then have been thrombosed and obstructed the outflow of the original AVM. However, neither the angiograms at the time of the initial operation nor the follow-up angiograms 3 years later revealed a septum in the superior sagittal sinus. Although the exact mechanisms of formation of the venous drainage pathway were not understood, retrograde venous drainage is significantly correlated with a risk of hemorrhage.^{1,2,7)} Therefore, surgical treatment was undertaken to prevent intracranial hemorrhage in our patient.

This unusual case shows that acquired dural AVMs have various etiologies, which may be related to surgical trauma or to pre-existing pial malformations. Vascular malformations may also present with a variety of venous drainage patterns.

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