

## Pial Arteriovenous Fistula Caused by Trauma: A Case Report

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### Abstract

We report an extremely rare case of pial arteriovenous fistula (AVF) caused by trauma. A 61-year-old man suffered from brain contusion by a traffic accident. He was neurologically normal on admission. However, his headache gradually worsened, and partial seizures occurred thereafter. He presented with general tonic seizure 7 days after the head injury. Magnetic resonance imaging demonstrated the exacerbation of brain edema and an abnormal vein near the contusion. Subsequent angiography showed a pial AVF, which was considered to be responsible for the brain edema. After treatment of the AVF by direct surgery, the brain edema was ameliorated. We should take into consideration the formation of vascular disease in cases with unexpected worsening of edema after brain injury.

Key words: pial arteriovenous fistula, trauma, edema

### Introduction

Intracranial pial arteriovenous fistulas (AVFs) are rare vascular lesions of the brain, accounting for about 1.6% of all brain vascular malformations.<sup>1)</sup> Most of the pial AVFs are considered to be congenital, and manifest congestive heart failure or intracerebral hemorrhage in newborn infancy.<sup>2)</sup> In adults, pial AVFs are considered to occur in association with brain infarction, infection, or trauma;<sup>3)</sup> however, they have not been well documented. We here report a very rare case of pial AVF resulting from trauma, which was considered to be responsible for brain edema and was successfully treated surgically. To the best of our knowledge, this is the first report that clearly demonstrated the development of a pial AVF after head injury.

### Case Report

A 61-year-old man was brought to our hospital because of a traffic accident. He suffered from head injury on the right temporal area. On admission, there was no problem on the neurological examination. Brain computed tomography (CT) scan showed traumatic subarachnoid hemorrhage and contusion on the right temporal lobe. Magnetic resonance imaging (MRI) exhibited brain edema in the right temporal lobe, and magnetic resonance (MR)

angiography was normal (Fig. 1). He was treated with the conservative therapy initially.

The patient presented with general tonic seizure 7 days after the onset. MRI revealed that the brain edema at the right temporal lobe worsened, and that an abnormal vein around the contusion of the temporal lobe appeared (Fig. 2). Subsequent angiograms demonstrated a pial AVF, fed by the branch of the middle cerebral artery and draining into the vein of Labbé (Fig. 3). On the capillary phase, the sylvian vein was also visualized by the blood flow back from the vein of Labbé, including its high venous pressure. Several hours later, he was still drowsy possibly due to the seizure and the brain edema. To ameliorate the brain edema and to prevent intracranial hemorrhage, the patient underwent an emergent operation.

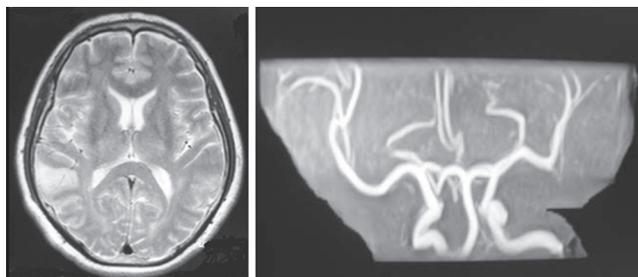


Fig. 1 Magnetic resonance (MR) imaging (*left*) and MR angiography (*right*) obtained on admission. There was mild brain edema; however, no abnormal vessels were observed.

After right temporal craniectomy, we observed that the vein of Labbé turned red. We dissected the sulcus in which the feeding artery passed. The artery was clipped just proximal to the fistula, and the fistula was coagulated (Fig. 4). Thereafter, the red vein turned blue. The bone flap was removed for the severe brain edema.

Postoperative course was uneventful. Angiograms after the operation exhibited the disappearance of the arteriovenous shunt. Follow-up CT scans demonstrated the reduction of the edema on the right temporal lobe. After cranioplasty, the patient was discharged.

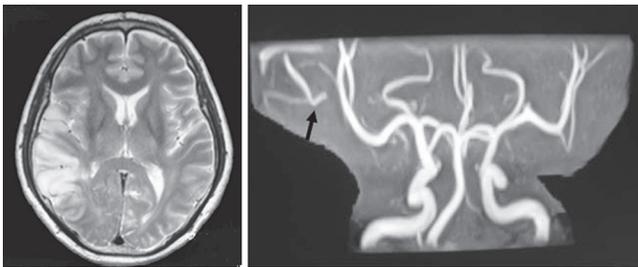


Fig. 2 Magnetic resonance (MR) imaging (*left*) and MR angiography (*right*) obtained 7 days after the onset. Brain edema worsened, and an abnormal vessel appeared in the right cerebrum (*arrow*).

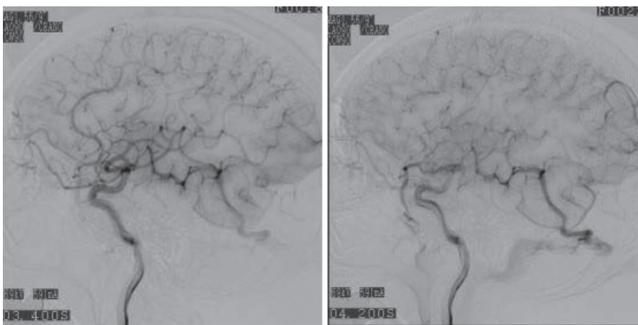


Fig. 3 Angiography on the arterial phase (*left*) and on the capillary phase (*right*). A pial arteriovenous fistula was fed by the branch of the right middle cerebral artery, and draining to the vein of Labbé. On the capillary phase, the sylvian vein was visible by the back flow from the vein of Labbé.



Fig. 4 Intraoperative photographs. The feeding artery was identified (*left*), and was clipped near the fistula (*middle*). The fistula was coagulated (*right*).

## Discussion

Since 1970, intracranial pial AVFs have been reported in less than 100 cases,<sup>4)</sup> and their natural history remains unknown. Nelson et al. reported eight patients with symptomatic pial AVF managed conservatively, and among them, five died due to cerebral bleeding.<sup>5)</sup> Intracranial dural AVFs are also well known to follow an aggressive course by venous hypertension, and the curative therapy is necessary in symptomatic cases.<sup>6)</sup> In pial AVFs, the high pressure of cortical vein disturbs the venous drainage, which may cause brain edema and intracranial hemorrhage. Therefore, curative treatment is also considered to be necessary for symptomatic pial AVFs.

The optimal treatment of pial AVFs is disconnection of direct shunting, and complete resection is usually not needed.<sup>7)</sup> Oya et al. insisted that a direct operation by microsurgery is recommended if the shunt point is easy for surgical approach, and that endovascular treatment is recommended in deep located lesions.<sup>5)</sup> Detailed analysis of cerebral angiography is important to detect the exact location of a feeding artery and a shunting point of an AVF, and is useful in deciding which treatment is better.

After trauma, dural AVFs are known to develop between the middle meningeal artery (MMA) and the diploic veins located around the skull fractures, and medial defects of the MMA are responsible for their formation.<sup>8,9)</sup> In the current case, a similar mechanism seemed to be responsible for the formation of the pial AVF between the middle cerebral artery and the surrounding veins.

Of course, the aggravation of brain edema often occurs after the brain injury, and therefore, we seldom evaluate the vascular abnormalities. Nevertheless, in cases with unexpected worsening of brain edema, we should undergo vascular studies to investigate the presence of AVFs.

## Conclusion

We report a very rare case of a pial AVF by trauma manifesting severe brain edema. It is necessary to take into consideration the formation of vascular disease in cases with unexpected severe edema after brain injury.

## Conflicts of Interest Disclosure

None.

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