Traumatic Pseudoaneurysm of the Middle Meningeal Artery with an Arteriovenous Fistula on a Non-Fractured Site

JUNG HO KO, YOUNG-JOON KIM

Department of Neurosurgery, Dankook University College of Medicine; Cheonan, Korea

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Summary

We describe a rare case of a combined traumatic pseudoaneurysm and arteriovenous fistula (AVF) of the middle meningeal artery (MMA) on a non-fractured site. A 24-year-old man was admitted to our hospital with head trauma. He underwent a craniotomy and removal of an epidural hematoma on the right side. Twenty-five days later, he complained of pulsatile tinnitus on the left non-fractured side. Angiography revealed a markedly dilated proximal MMA with flow shunting to the pterygoid plexus. We performed proximal occlusion on the proximal MMA for the traumatic pseudoaneurysm and the AVF of the MMA using coils. Although immediate angiography showed retrograde contrast filling from the collateral vessels into the distal part of the pseudoaneurysm, follow-up angiography revealed that the lesion had successfully disappeared.

Introduction

Traumatic pseudoaneurysm and arteriovenous fistula (AVF) of the middle meningeal artery (MMA) are rare complications of head trauma. Furthermore, most of these lesions are related to skull fractures, and lesions on non-fractured sites are very rare. Many lesions are missed if there are no clinical manifestations and are only discovered incidentally on neuroimaging. We describe a case of a combined traumatic pseudoaneurysm and AVF of the MMA on a non-fractured site.

Case Report

A 24-year-old man was admitted to our hospital with head and multiple traumas after a motor vehicle accident. The patient was unconscious on admission, and brain computed tomography (CT) showed an epidural hematoma (EDH) combined with a skull fracture in the right temporoparietal area. Urgent craniotomy with removal of the hematoma was performed, and the hematoma was completely removed (Figure 1). After the operation, the patient's neurological status improved and was uneventful. However, around the 25th day post-operation he complained of gradually increasing pulsatile tinnitus on the opposite side of the operation site. Brain magnetic resonance angiography (MRA) revealed an abnormal vascular lesion on the left external carotid artery (ECA). A left external carotid angiogram showed a markedly dilated proximal MMA with flow shunting to the pterygoid plexus (Figure 2). We considered an endovascular embolization for the traumatic MMA pseudoaneurysm with AVF using coils and a liquid embolic agent such as n-butyl-2-cyanoacrylate (NBCA).

We planned the procedure in the following order: first, the coil embolization would be performed to reduce the shunting flow. Second, after sufficiently reducing the flow into the AVF and MMA pseudoaneurysm, an NBCA embolization was considered for the remnant lesion. The procedure was conducted with the patient awake. The right femoral artery was accessed using a 6-French sheath and the left ECA was catheterized with a 6-French Fas Guide guiding

Figure 1 A,B) Brain computed tomography (CT) showed an epidural hematoma (EDH) combined with a skull fracture in the right temporoparietal area. C) Urgent craniotomy with removal of the hematoma was performed. The hematoma was completely removed.

Figure 2 A) Brain magnetic resonance angiography (MRA) revealed an abnormal vascular lesion on the left external carotid artery (ECA) (white arrow). B) A left external carotid angiogram showed the markedly dilated proximal MMA (arrowhead) with flow shunting to the pterygoid plexus (black arrow).

catheter (Boston Scientific/Target Therapeutic, Fremont, CA, USA). An Excelsior SL10 microcatheter (Boston Scientific/Target Therapeutic, Fremont, CA, USA) was then introduced into the MMA pseudoaneurysm beyond the suspected fistulous portion, which could have been located around the foramen spinosum. Platinum coils were deployed to reduce the flow of blood into the shunt. After deployment of the fifth

Figure 3 A) After the eighth and final coil was deployed, the immediate post-operation angiogram showed complete occlusion of the fistula. B,C) However, there was scant retrograde contrast filling into the distal part of the pseudoaneurysm from the collateral meningeal arteries (arrowhead).

Figure 4 A follow-up angiogram was obtained in the third month post-embolization. A complete disappearance of the pseudoaneurysm was revealed until the late venous phase of the ECA angiogram.

coil, a digital subtraction angiogram showed complete occlusion of the fistula. However, blood flow into the distal portion of the pseudoaneurysm was patent. Although we tried to advance the microcatheter to the distal portion through the coil mesh, it was difficult to move the microcatheter forward. We were compelled to perform proximal occlusion on the proximal MMA. After the eighth and final coil was de-

ployed, an immediate post-operation angiogram showed complete occlusion of the fistula but scant retrograde contrast filling into the distal part of the pseudoaneurysm from the collateral meningeal arteries (Figure 3). The pulsatile tinnitus disappeared immediately during the procedure. The patient's post-procedural neurological status was unchanged, and his subsequent clinical course was uneventful. To assess poten-

tial recurrence of the pseudoaneurysm due to collateral flow, a follow-up angiogram was obtained in the third month post-embolization. A complete disappearance of the pseudoaneurysm was revealed until the late venous phase of the ECA angiogram (Figure 4). The patient did not complain of any further neurological or physical symptoms.

Discussion

Traumatic AVF and pseudoaneurysm of the MMA are rare and usually followed by blunt or penetrating head trauma¹⁻³. MMA injury occurs in 4% of head trauma patients and AVF of the MMA in 1.8%⁴. Dural AVF of the MMA is the second most common traumatic dural AVF following traumatic carotid cavernous sinus fistula^{2,5}. Pseudoaneurysms account for less than 1% of all intracranial aneurysms and are mostly related to a history of head trauma³. The majority of cases affect the internal carotid artery, and pseudoaneurysms of the MMA are very rare¹. Furthermore, a combination of pseudoaneurysm and AVF is extremely rare, and only 5% of cases of pseudoaneurysm of the artery have been reported^{5,6}. To our knowledge, a combined case on the non-fractured side has never been reported. Injury to the MMA from cranial fractures might cause epidural hematomas, traumatic aneurysms, or arteriovenous fistulae^{4,7,8}. Skull fracture is mostly associated with AVF, forming a shunt with the middle meningeal and diploic veins. About 70-90% of cases of traumatic MMA pseudoaneurysms are also associated with fracture across the MMA^{1,5}. Pseudoaneurysms and AVF seem to have the same pathogenesis. Tearing of the arterial wall by skull fracture or by separation of the dura mater and bone has been suggested⁶. A rare case of traumatic middle meningeal AVF on the side of the head opposite the injured side has been reported⁹⁻¹¹. With respect to the pathogenesis, these authors suggested that the deformation in the skull by the impact of the injury at the onset may have caused dura mater-bone separation or distortion of the dura, and this may have indirectly caused a collapse of the dural vessels. However, our case may have been caused from a direct connection between the MMA to the middle meningeal vein (MMV) in the extradural portion. It was not a true dural AVF. We consider that avulsion of the MMA between the vessels and the foramen spinosum could have

been a potential etiology. The clinical symptoms of AVF depend on the velocity of blood flow, fistula size, and shunt volume, and can occur from a few minutes to several weeks after trauma². The symptoms may include headache, dizziness, disorder of consciousness, and pulsatile tinnitus. Pulsatile tinnitus is rare but can be the first symptom of life-threatening disorders¹². Our patient complained of pulsatile tinnitus, which was seen as a clue to the possible detection of a lesion. Pseudoaneurysms of the MMA have the potential to rupture, resulting in neurological deterioration from intracranial hemorrhage post-trauma in the third to thirtieth day interval^{1,6,8}. When bleeding, they lead to a high mortality rate. The most frequent presentation of traumatic pseudoaneurysms is acute or delayed epidural hematoma, but they may also uncommonly lead to subdural, subarachnoid, or even intracerebral hemorrhage^{1,3,7}. Nonetheless, the natural history of traumatic aneurysms is not well known, and spontaneous resolutions have even been reported¹³.

Usually, traumatic pseudoaneurysms of the MMA demonstrate specific characteristics^{1,3,5,7}: they have an irregular wall, are located peripherally and do not have a true neck, with delayed filling and emptying. Optimal treatment entails trapping the aneurysm and parent vessels by endovascular means or direct surgical removal. In the present case, the pseudoaneurysm had unusual findings, such as a proximal location and fusiform appearance. Tsutsumi et al.⁶ reported a similar case, which developed at the proximal MMA beyond the foramen spinosum and was a fusiform. The pseudoaneurysm in their case was secured by endovascular trapping using coils. In the present case, after proximal occlusion was done, immediate angiography showed retrograde contrast filling from the collateral vessels into the distal part of the pseudoaneurysm, which had a risk of recurrence or bleeding (Figure 3). Although trapping could not be performed, follow-up angiography showed that the aneurysm had successfully disappeared. However, if feasible, we consider trapping to be the best treatment for pseudoaneurysms when endovascular treatment is considered.

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Young-Joon Kim, MD
 Department of Neurosurgery
 Dankook University College of Medicine
 359 Manhyangro, Dongnam-gu, Cheonan
 Choongnam 330-714, Republic of Korea
 Tel.: 041-550-3980
 Fax: 041-552-6870
 E-mail : kimyj@dankook.ac.kr