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Case Report

Mixed Pial-Dural Arteriovenous Malformation in the Anterior Cranial Fossa Mimicking Dural Arteriovenous Fistula

Abstract

Mixed pial-dural arteriovenous malformation (MpdAVM) and dural arteriovenous fistula (dAVF) are rare entities in the anterior cranial fossa (ACF). As dural-pial vascular anastomosis can exist near the cribriform plate, MpdAVM with a small nidus, which cannot be apparently identified, can be logically indistinguishable from dAVF in ACF. A 71-year-old man was referred for evaluation of possible intracranial vascular malformation. Cerebral angiography revealed an arteriovenous shunt in the ACF. The lesion was fed by the bilateral ethmoidal arteries and right orbitofrontal artery, draining through the bilateral cortical veins to the superior sagittal sinus. As a nidus was not detected, dAVF was suspected. Venous interruption was planned with direct surgery. Intraoperatively, an arterial aggregation was observed in the right frontal lobe. The arterial aggregation seemed to be connected to the interrupted drainer in the right ACF. The arterial aggregation was removed and pathologically diagnosed as arteriovenous malformation. Postoperatively, intracerebral hemorrhage was confirmed, and postoperative cerebral angiography confirmed the resolved arteriovenous shunt. The intracranial hemorrhage was possibly due to the timing gap between drainer interruption and removal of the nidus. MpdAVM with a small nidus in the ACF can mimic dAVF. Clinicians must be aware that an unremoved nidus of MpdAVM may postoperatively result in fatal intracranial hemorrhage.

Keywords: *Anterior cranial fossa, cerebral angiography, dural arteriovenous fistula, mixed pial-dural arteriovenous malformation, surgery*

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Introduction

Mixed pial-dural arteriovenous malformation (MpdAVM), which has vascular supply from the pial artery and the dural artery,[1] has been reported as a relatively rare entity in the anterior cranial fossa (ACF).[2] Dural arteriovenous fistula (dAVF) in the ACF occupies 5.8% of the intracranial dAVFs[3,4] and is also considered rare.[3] MpdAVM in the ACF can be fed by the ethmoidal artery, the orbitofrontal artery, and the branches from the internal carotid artery,[2] while dAVF in the ACF is usually supplied by the anterior ethmoidal artery draining into the superior sagittal sinus (SSS).[5] As vascular anastomosis between the ethmoidal arteries and the dural branches of the anterior cerebral artery exists in the region of the cribriform plate,[6] it is thus logically possible that MpdAVM with a small nidus, which is not apparently identified on radiological examinations, can mimic dAVF in the ACF. However, to the best of our knowledge, a case of MpdAVM in the ACF mimicking dAVF has not been described possibly due to its rarity. Here, we present a relatively rare case of MdpAVM in the ACF mimicking dAVF.

Case Report

A 71-year-old male patient was referred to our department of neurosurgery by a cardiologist for hypertension and angina. He had a family history of cerebral infarction and requested magnetic resonance imaging (MRI). Cerebral infarction or hemorrhage was not observed, but the SSS was demonstrated on magnetic resonance angiography. Cerebral angiography was performed to rule out any vascular malformation. The right internal carotid angiography showed an arteriovenous shunt supplied by the ethmoidal artery and the orbitofrontal artery. The arteriovenous shunt in the ACF was draining to the SSS [Figure 1a and b]. The left internal

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carotid angiography revealed a shunting point in the right ACF [Figure 1c and d]. The dilated drainers and feeding arteries were also confirmed on heavy T2-weighted MRIs. However, we were unable to detect any vascular lesion in the parenchyma, with which we could suspect the existence of a nidus [Figure 1e-h]. As the arteriovenous shunt in the ACF seemed not to be accompanied by an apparent nidus, we considered that the lesion in the ACF could be dAVF fed by the bilateral ethmoidal arteries, draining to the SSS through bilateral cortical veins. Vascular supply of dAVF from the orbitofrontal artery seemed atypical, but we considered that atypical feeding from the orbitofrontal artery could be due to vascular anastomosis between the ethmoidal artery and the orbitofrontal artery.[6] As the dAVF with variceal venous dilatation in the ACF was reported with a high incidence of bleeding events, surgical intervention was planned.

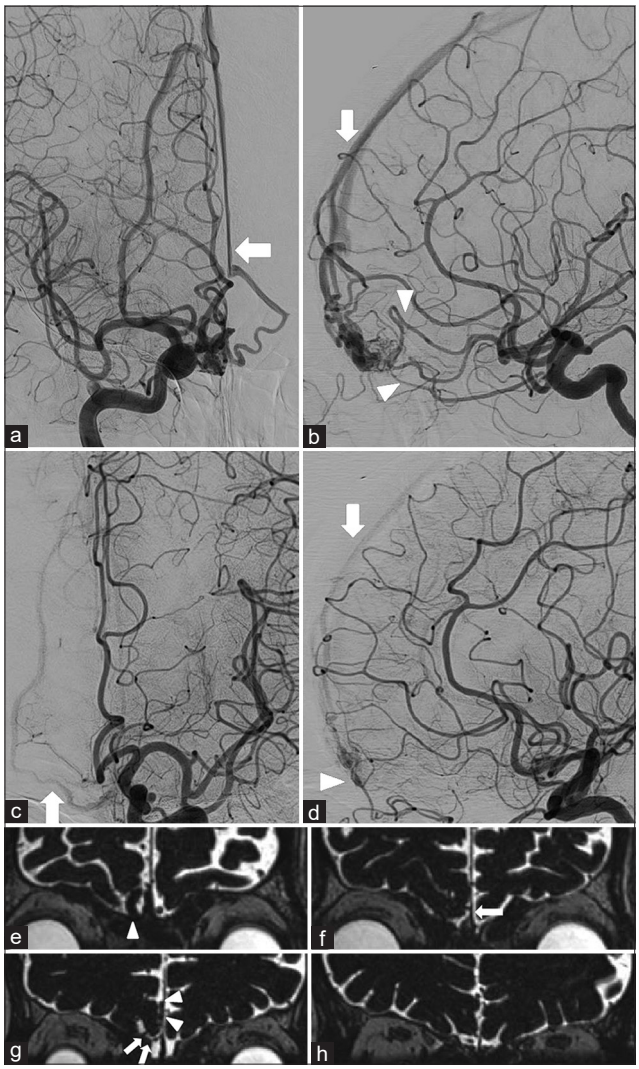

Figure 1: (a and b) The right internal carotid angiography showing the arteriovenous shunt (white arrow). The lesion is fed by the right ethmoidal artery and the orbitofrontal artery (white arrow heads). (a: Anterior–posterior projection, b: lateral projection). (c and d) The left internal carotid angiography revealing the arteriovenous shunt (white arrow). The vascular supply from the left ethmoidal artery was also confirmed (white arrowhead). (e-h) Heavy T2-weighted magnetic resonance images. The dilated drainers (arrowheads) and feeding arteries (arrows) were confirmed in the arachnoid spaces (e: At the level of the foramen cecum, f-h: Three different slices of the cribriform plate). No apparent vascular lesion was observed in the parenchyma

Operation

The patient was sedated under general anesthesia and set in the supine position. The head was straight and slightly elevated. Bifrontal craniotomy was made following bicoronal skin incision. First, we observed the right ACF. A cortical vein, which was related to a drainer of the arteriovenous shunt on the cerebral angiography, was recognized as red. The cortical vein was detached from the right frontal lobe. We followed the cortical vein to the SSS and found the variceal drainer [Figure 2a]. The variceal drainer in the right ACF was coagulated and cut. The red cortical vein changed then as usual vein appearance. Following the cut of the drainer in the right ACF, we observed an arterial aggregation in the frontal lobe [Figure 2b]. The arterial aggregation seemed to be connected to the major drainer. Since no other lesions except the possible dAVF were not preoperatively observed on the cerebral angiography, we suspected the arterial aggregation to be a vascular malformation. Thus, the arterial aggregation was removed approximately 1 h after drainer interruption. Following this procedure, we observed the left ACF. We entered the interhemispheric space dissecting the arachnoid, and we detected the drainer in the left ACF. The drainer was then coagulated and cut.

Postoperative course

Postoperative computed tomography (CT) image showed intracranial hemorrhage in the right frontal lobe. The hemorrhage in the right ACF was located in the region of the arterial aggregation and intraoperatively recognized [Figure 3a]. Postoperative cerebral angiography showed that the arteriovenous shunt in the ACF had resolved [Figure 3b-e]. The patient was treated with severe blood pressure control under 120 mmHg. The intracranial hemorrhage did not remarkably increase and surgical

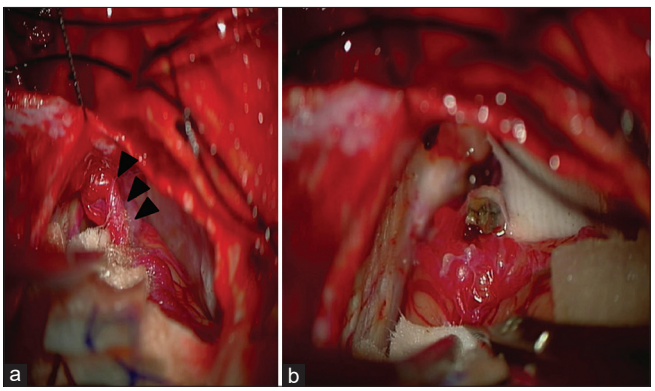

Figure 2: (a) The variceal drainer in the right anterior cranial fossa was detected (black arrowheads). (b) The arterial aggregation was observed (black arrow)

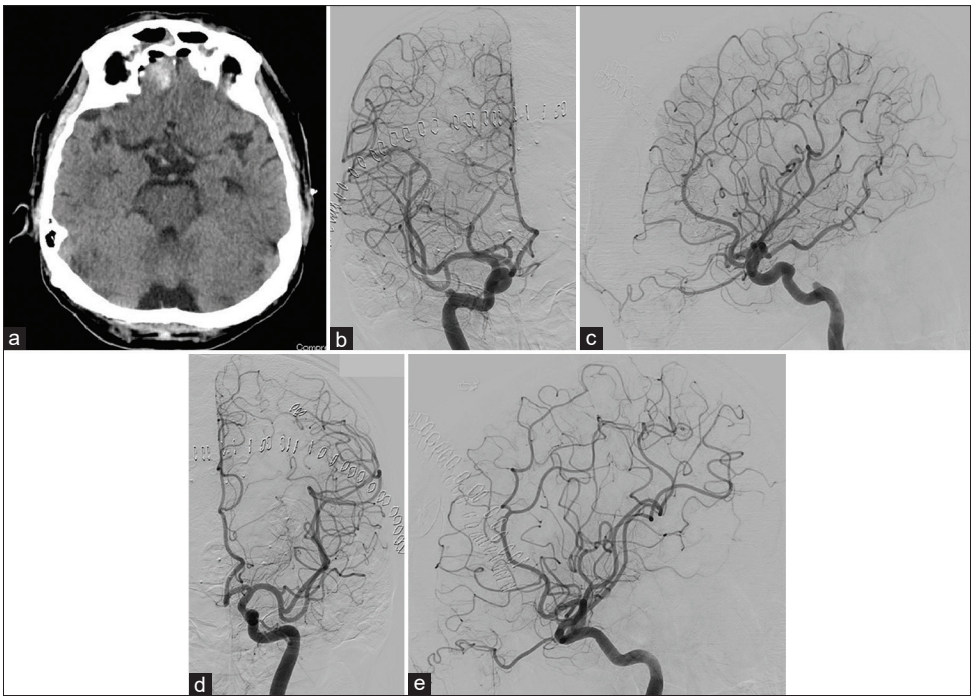

Figure 3: (a) Postoperative computed tomography demonstrating the intracranial hemorrhage in the right frontal lobe where the nidus was located. (b-e) Postoperative internal carotid angiography revealing disappearance of the arteriovenous shunt. (b and d: Arterial phase of anterior-posterior projection, c and e: Arterial phase of lateral projection)

removal of intracranial hematoma was not needed. Two months postoperatively, the patient experienced drowsiness and disorientation. A CT image revealed symptomatic hydrocephalus for which ventriculoperitoneal shut surgery was performed. After rehabilitation therapy, the patient was discharged from the hospital on postoperative month 8. The patient is now monitored in the outpatient clinic and is independent in activities of daily living at the follow-up of 1 year and 6 months after the discharge.

The pathological evaluation of the arterial aggregation in the right ACF was AVM [Figure 4].

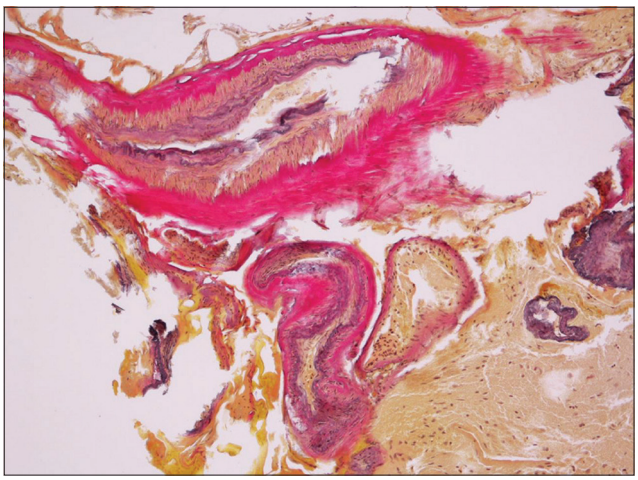

Figure 4: Elastica van Gieson staining of the specimen (original magnification ×100). The arterial and venous structures were confirmed in the cerebral parenchyma. These findings corresponded to arteriovenous malformation

Discussion

Here, we report a case of MpdAVM preoperatively considered as dAVF. The preoperative erroneous diagnosis was possibly because the nidus of MpdAVM was not apparently identified. The MpdAVM in our case, which was preoperatively considered as dAVF, was fed by the bilateral ethmoid artery and right orbitofrontal artery, draining bilaterally to the SSS. Pial supply from the orbitofrontal artery to the vascular malformation was not typical for pure dAVF. Jimbo *et al*. proposed that another vascular malformation except dAVF should be considered if the lesion has a pial artery supply.[2] However, dAVF can be fed by pial arteries.[7] Besides, vascular anastomosis between the ethmoidal arteries and the branches of the internal carotid artery can exist near the region of the cribriform plate.[6,8] Thus, we could not completely exclude that the vascular malformation was dAVF with vascular anastomosis between the dural feeding artery and the normal pial arteries. The bilateral drainage pattern seemed not to be typical as dAVF but can rarely exist.[5,9] Concerning the indication of surgery of dAVF, drainer interruption can be recommended for the cases accompanied by cortical venous drainage and variceal venous dilatation due to the high incidence of intracranial hemorrhagic events (62%–91%).[9,10] Direct surgery of drainer interruption has been often performed to interrupt the drainer of dAVF in the ACF, and endovascular treatment for dAVF seems to remain controversial with visual field defect or visual loss due to occlusion of the central retinal artery.[1,11,12]

The number of reports on MpdAVM in the ACF is still limited to date.[2,13-17] MpdAVM in the ACF can be fed by the ethmoidal artery, the orbitofrontal artery, and the branches from the internal carotid artery.[2] MpdAVM in the ACF is categorized into two groups according to the location of the nidus: parenchymal nidus or dural nidus.[2] Nidus removal should be necessary for the parenchymal nidus type, while drainer interruption can be enough for the dural nidus type.[2,12] In our case, only venous drainer interruption was planned because we firstly considered that the lesion could be dAVF. Venous drainer interruption had been adequate in case MpdAVM could have been dural nidus type. However, the nidus of MpdAVM intraoperatively confirmed as arterial aggregation was located in the brain parenchyma; thus, removal of the parenchymal nidus was necessary.

Postoperatively, the intracranial hemorrhage in the right frontal lobe was identified in the location where the nidus of MpdAVM existed. Intraoperative hemorrhage close to the nidus of MpdAVM occurred as the drainer of MpdAVM was cut prior to removal of the nidus. In that case, if we had not removed the nidus of MpdAVM, fatal hemorrhagic complication in the nidus could have occurred. Preoperative erroneous diagnosis of a lesion which can be revealed as arteriovenous malformation based on pathological evaluation is not so rare,[18] but, to the best of our knowledge, any report has not been previously described on a case of MpdAVM in the ACF firstly considered as dAVM. This is possibly due to the rarity of those two entities.

As seen in our case, MpdAVM with a small nidus in the ACF can be misdiagnosed for dAVF due to vascular anastomosis between the ethmoidal arteries and the branches from the internal carotid artery. Selective three-dimensional rotational angiography through the branches of the anterior cerebral artery can be also an option in case we suspect the possibility of MpdAVM with a small nidus in the ACF mimicking dAVF. Thus, we can avoid an erroneous surgical strategy which can otherwise result in fatal intracranial hemorrhage.

Conclusion

MpdAVM with small nidus in the ACF can mimic dAVF. Failure to be aware of this possibility may lead to sole drainer interruption without removal of the nidus of MpdAVM resulting in postoperative fatal intracranial hemorrhage.

Ethical standard

We obtained informed consent from the patient and his family.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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