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Case Report

A Case of Intraosseous Dural Arteriovenous Fistulas Involving Diploic Vein Treated with Transarterial Onyx Embolization

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Intracranial dural arteriovenous fistulas (DAVFs) are abnormal arteriovenous connections that lie within the dura. Intraosseous DAVFs involving diploic venous system are extremely rare. A 46-year-old woman presented with headache and right pulsatile tinnitus for three weeks. The tinnitus started after yelling. Digital subtraction angiography revealed DAVF within the basal portion of right parietal bone along the middle meningeal artery (MMA) groove. The fistula was fed by frontal branch of right MMA and drained into right transverse sigmoid sinus junction through dilated middle meningeal vein. The intraosseous DAVF involving diploic vein was successfully obliterated with Onyx embolization via transarterial route.

Key Words : Dural arteriovenous fistulas · Tinnitus · Onyx · Diploic vein · Transarterial embolization.

INTRODUCTION

Intracranial dural arteriovenous fistulas (DAVFs) are abnormal arteriovenous connections that lie within the dura[14]. It accounts for 10-15% of all intracranial arteriovenous malformations[1].

Classic DAVFs usually involve the dura adjacent to the venous sinuses[4]. However, DAVFs can occur at any site within the dura maters. There have been many cases of DAVFs with extrasinusal locations which include skull base, tentorium, and intraorbital area[5,8,10-13,16]. But, DAVFs involving diploic veins (DVs) are extremely rare[2]. The authors report a case of spontaneous intraosseous DAVF involving DV which was successfully treated with Onyx embolization via transarterial route.

CASE REPORT

History and examination

This 46-year-old woman without medical history presented with headache and right pulsatile tinnitus for three weeks. This pulsatile tinnitus started after yelling. There was no history of head trauma or other precipitating event. The intensity of pulsatile tinnitus was decreased by manual compression of right carotid artery. Neurological examination was normal, but bruit was audible by auscultation on the right temporal area.

Image findings

Brain MRI did not show any brain parenchymal lesions, but TOF MRA images demonstrated tangled vessels on right temporo-parietal area. Bone window of brain CT showed prominent intraosseous diploic space on the basal portion of right parietal bone with small defect on inner table of the skull (Fig. 1A , B). Digital subtraction angiography revealed DAVF within the basal portion of right parietal bone along the middle meningeal artery (MMA). Dilated fistulous venous pouch within diploic space of the right parietal bone was measured 29×9 mm on 3D rotational angiogram. The DAVF was fed by frontal branch of right MMA. It was drained into right transverse sigmoid sinus junction through the middle meningeal vein (MMV) without retrograde cortical venous reflux (Fig. 1C, D, E). Internal carotid arterial system did not contribute to the fistulous flow.

Treatment

Under general endotracheal anesthesia, a 5 French Envoy guiding catheter (Cordis, Miami, FL, USA) was placed proximal to the origin of right internal maxillary artery via a transfemoral arterial route. After intravenous bolus injection of 3,000 unit of heparin, a microcatheter of Echelon-10 (ev3 Neurovascular, Irvine, CA, USA) was introduced and placed to the

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frontal branch of MMA at the just proximal portion of DAVF under roadmap guidance. Microcatheter angiograms clearly demonstrated multiple fine arterial channels of MMA connecting to dilated venous pouch, drained into right transverse sigmoid sinus junction through enlarged MMV (Fig. 2A, B).

Onyx 18 (ev3 Neurovascular, Irvine, CA, USA) injection was uneventful. Two milliliter of Onyx injection resulted in complete obliteration of the DAVF including multiple fine arterial channels from the MMA (Fig. 2C, D, E).

Postoperative course

Her preoperative symptoms resolved completely after embolization. Postoperative CT showed Onyx cast within the right parietal bone and middle fossa dura along the MMA, which confirmed the intraosseous location of DAVF (Fig. 3). Her postoperative course was uneventful. She was discharged 3 days postoperatively.

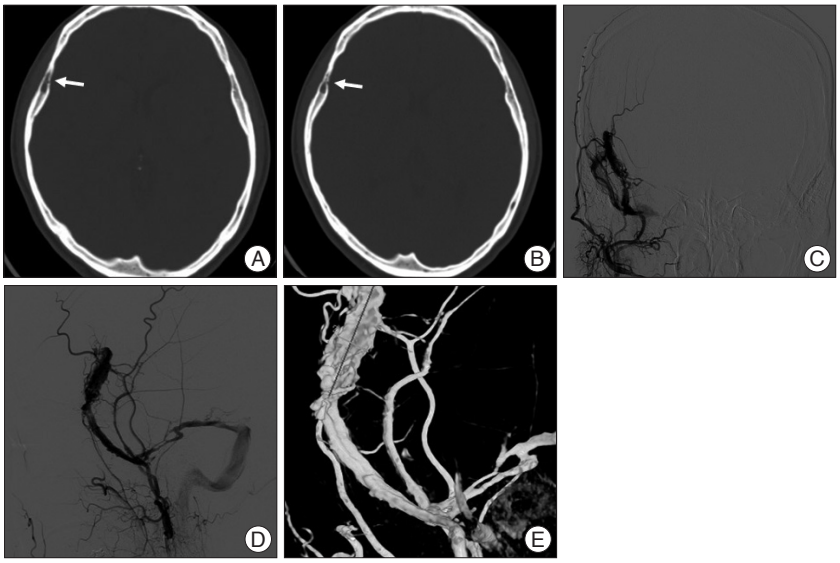

Fig. 1. Preoperative images showing intraosseous dural arteriovenous fistula in a 46-year-old woman. Bone window of brain CT images (A and B) revealing prominent diploic space on the right parietal bone and small defect on inner table along the middle meningeal artery (MMA) groove (white arrows). Right external carotid angiograms, anteroposterior view (C) and lateral view (D), demonstrate a dilated fistulous venous pouch within the basal portion of right parietal bone, which is fed by frontal branch of the right MMA, drains into transverse sigmoid sinus junction through dilated middle meningeal vein. The intraosseous fistulous venous pouch is measured 29×9 mm on 3D rotational angiogram (E).

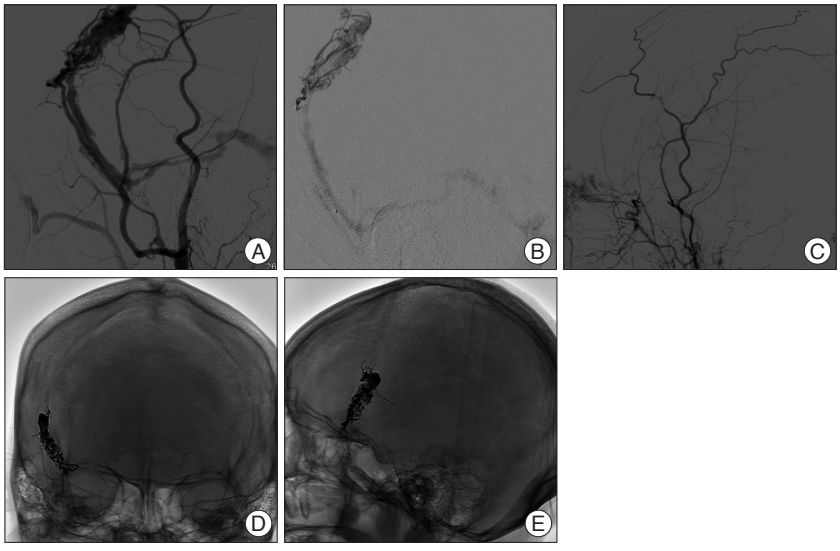

Fig. 2. Intraoperative angiograms and skull radiographs after Onyx embolization. Magnified view (A) and superselective angiogram (B) just before Onyx injection show enlarged frontal branch of middle meningeal artery (MMA) and middle meningeal vein as well as the intraosseous dural arteriovenous fistula (DAVF). Multiple fine arterial recruitments from right MMA to fistulous venous pouch are clearly seen. Postoperative lateral view of right external carotid angiogram (C) demonstrates complete obliteration of intraosseous DAVF. Skull radiographs after Onyx embolization (D and E) show Onyx cast on the right temporoparietal area.

DISCUSSION

DVs are located between two cortical bones and are lined by a single endothelial layer without vascular valves. In normal conditions, the DVs can be seen as slightly delayed more than the rest of cerebral veins on cerebral angiography[6]. DVs communicate with the dural sinus, emissary veins, pericranial veins, meninges, and skull base. This anatomical relationship plays a role in pathological situations, such as subgaleal hematoma, sinus thrombosis and DAVFs. Under such pathologic circumstances, the normal venous draining pathway can be compromised, and the DVs may show increased flow on cerebral angiography[2].

Classic DAVFs are situated within the dura adjacent to the venous sinus[4]. However, DAVFs can occur wherever the veins follow adjacent the dura if there is a fistulous connection between the artery and vein. Malik et al.[12] reported 2 cases of DAVFs with vascular nidus that was situated within the bone, and they used the term "intraosseous DAVF". In intraosseous DAVFs, these extrasinusal fistulous connections can be developed between meningeal arteries and the DVs or emissary veins. Common extrasinusal locations are the skull base, tentorium and intraorbital area via emissary veins[5,8,10-13,16]. However, the DAVFs involving DV are extremely rare and only few cases have been reported[1,2,7,17].

The known causes of DAVFs are related to the head injury,

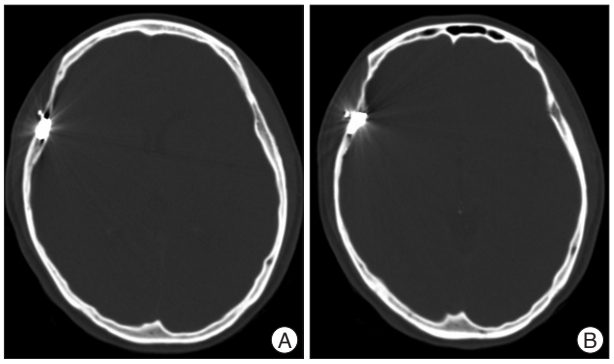

Fig. 3. Postoperative bone window of brain CT scans (A and B) showing complete obliteration of dural arteriovenous fistula (DAVF) via transarterial Onyx embolization. The Onyx cast is demonstrated within the diploic space on right parietal bone, which confirms intraosseous DAVF.

cranial surgery, or venous sinus thrombosis[19]. But, in this case she had no history of head trauma, or venous sinus thrombosis. Her tinnitus symptom was developed immediately after yelling. It is unclear how the intraosseous DAVF involving DV was formed spontaneously after yelling. One possible explanation is that any fistulous connection between MMA and DV might have been developed after yelling because there was no tinnitus symptom until that event. Her brain CT showed a small defect on inner table of the right parietal bone along the MMA groove as well as neighboring prominent diploic space. Therefore, it is probable that the MMA and DV were being in contact with each other. The longstanding contact of the lateral wall of the MMA with sharp bony spicule on the MMA groove defect could have made some erosion of the MMA wall before the occurrence of fistula. At the moment of yelling, an abrupt increase in blood pressure and intracranial pressure might have resulted in simultaneous ruptures of erosive portion of the MMA and neighboring thin walled DV. Fistulous communication between artery and vein can occur, which further recruits fine meningeal branches to grow intraosseous DAVF.

Intraosseous DAVFs can be treated by surgical resection or endovascular treatment. Surgical resection of DAVFs achieved curative results in several reports, but it still remained the risk of surgery[11,12]. Historically, transvenous approach has been considered more ideal to occlude the fistula completely than transarterial embolization, which requires multiple feeder catheterization. However, the transvenous approach requires retrograde catheterization, which carries a high risk of sacrifice of the involved venous structures. This approach can be associated with severe complications, such as vessel perforation, venous infarction and hemorrhage[9,18]. Recently, some authors reported high rates of complete obliteration of DAVFs via transarterial approach using Onyx[3,15,19]. Introduction of Onyx embolic material make it possible to infiltrate the fistulas and their multiple feeders as a retrograde fashion due to good penetration capability of Onyx. Penetration of Onyx through the fistula can be controlled effectively by plug and push technique. In this case, complete obliteration of intraosseous DAVF involving DV was achieved with single arterial injection of Onyx without any complications.

CONCLUSION

Intraosseous DAVF is a rare disease entity. The authors report a case of intraosseous DAVF involving DV which was treated with transarterial injection of Onyx embolic material without any complications. Transarterial Onyx embolization seems to be a safe and effective treatment modality for DAVF involving DV.

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