






Direct Cannulation of a Calvarial Diploic Vein for Embolization of a Symptomatic Intraosseous Arteriovenous Fistula: A Case Report

두개관 판사이정맥의 직접 관삽입술을 통한 유증상 골내 동정맥류의 색전술: 증례 보고

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Diploic arteriovenous fistulas (AVFs) or intraosseous dural AVFs are rare arteriovenous shunts. A diploic AVF is formed between a meningeal artery and an intraosseous diploic vein or the transosseous emissary vein, and the nidus is located exclusively within the bone. Currently, endovascular embolization with a transvenous approach is considered the treatment of choice for most dural AVFs. However, in the absence of an accessible venous channel, an alternate treatment approach should be considered. Herein, we report a case of a diploic AVF that was treated using embolization with transosseous direct cannulation.

Index terms Arteriovenous Fistula; Embolization, Therapeutic; Cerebrovascular Disorders

INTRODUCTION

Dural arteriovenous fistulas (AVFs) are abnormal connections between the dural arteries and venous sinuses. They may cause hypertension in the cerebral venous system, leading to local congestive edema and hemorrhage (1). Current management strategies for dural AVFs include endovascular embolization, radiosurgery, and open surgery. Among these, endovascular transvenous and/or transarterial embolization is consid-

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ered the treatment of choice for most dural AVFs, with the development of various embolic materials, including coils, N-butyl cyanoacrylate, and Onyx (2).

Diploic AVFs or intraosseous dural AVFs are rare. In diploic AVFs, a fistula is formed between the meningeal arteries and intraosseous diploic veins (DVs) or the transosseous emissary veins, and the nidus is located exclusively within the bone. Usually, they drain into the dural sinuses or jugular veins, but rarely into isolated segments of the DV and cortical veins without antegrade sinus drainage (3). The transvenous approach for embolization via the jugular vein is considered the most effective approach when the AVF eventually drains into the jugular vein (2). However, alternative approaches should be considered in the absence of an accessible venous channel. In this report, we describe the direct access of the calvarial DV for the treatment of a diploic AVF located along the left frontal bone.

CASE REPORT

An 81-year-old male with a medical history of hypertension visited the emergency room with sudden dysarthria for 5 minutes. He had no history of head trauma or cerebrovascular disease. Neurological examination showed no abnormal findings, except for dysarthria.

On brain CT, a small amount (about 2.1 cm × 1.0 cm × 1.9 cm in size) of intracerebral hemorrhage (ICH) in the left frontal lobe was detected. Brain CT angiography revealed prominent cortical veins surrounding the ICH with corkscrew appearance. Brain MRI revealed an abnormally dilated venous sac within the left frontal bone. The lesion manifested as an intraosseous signal-intensity void. In addition, multiple corkscrew flow voids surrounding the left frontal lobe were observed on the MR images (Fig. 1A). Digital subtraction angiography revealed a diploic AVF located along the entire length of the anterior temporal DV, from the pterion to the parasagittal area of the left calvarium. This fistula was fed by the intraosseous branches of the left superficial temporal artery and middle meningeal artery, and a markedly dilated anterior temporal DV drained reversely through the emissary of the frontal bone into the frontal cortical veins (Fig. 1B). An internal carotid angiogram showed significant congestion of the cortical veins due to increased venous pressure as a result of the AVF reflux.

The patient decided to proceed with endovascular embolization of the diploic AVF. Although the transvenous or transarterial route through the femoral vessels is a traditional approach for embolization of dural AVFs, we decided to perform direct cannulation of the DV. Because the antegrade venous flow to the jugular vein was not observed on angiography, the retrograde approach to the sinus draining the diploic AVF via the ipsilateral internal jugular vein was technically impossible. Furthermore, transarterial embolization was also technically difficult, because arterial feeders of this AVF were composed of numerous small feeding arteries without dominant main feeders.

Embolization of the diploic AVF was performed using a mobile C-arm in the operating room under general anesthesia. Right common femoral artery access was obtained, and the left external carotid angiography was performed before, during, and after embolization to visualize the anatomy of the fistula. The intraosseous diploic channel containing the draining vein of the fistula was visualized using fluoroscopy.

Under the CT-based navigation system, the left frontal craniotomy was performed to ac-

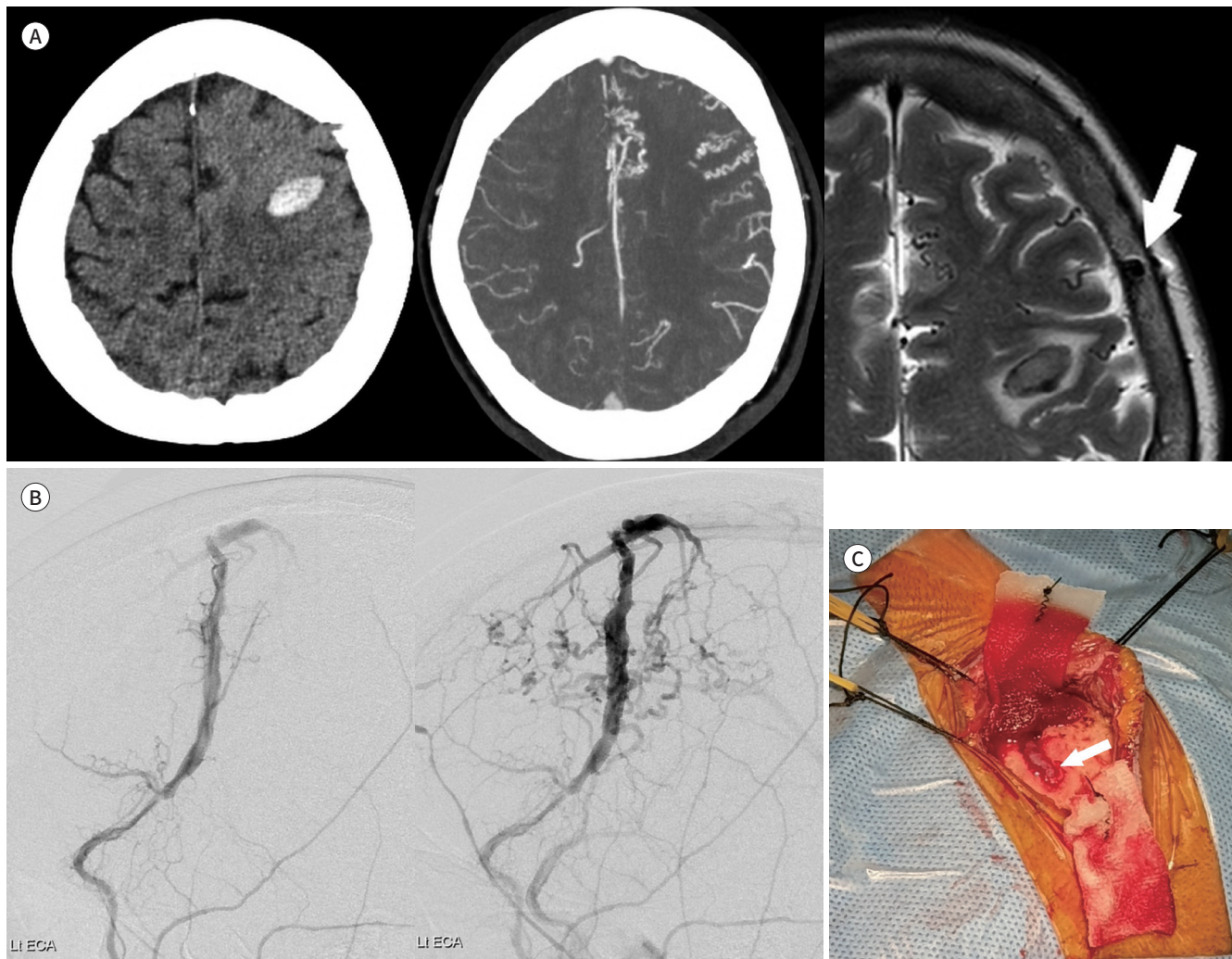
Fig. 1. Transcranial embolization of a diploic AVF along the frontal bone, presented as intracranial hemorrhage, in an 81-year-old male with sudden dysarthria.

A. Brain CT demonstrates a small amount of ICH at the left frontal lobe (right image). Brain CT angiography reveals prominent cortical veins surrounding the ICH with a corkscrew appearance (left image). T2-weighted MRI shows an abnormal dilated diploic venous sac (arrow, right image) in the left frontal bone.

B. Left external carotid angiography shows a diploic AVF fed by the left superficial temporal artery and middle meningeal artery and drained through the long segmental left anterior temporal DV into the frontal cortical veins (early phase: left image, late phase: right image).

C. An intraoperative photograph following outer cortex decortication shows the arterialized DV (arrow).

AVF = arteriovenous fistula, DV = diploic vein, ICH = intracerebral hemorrhage



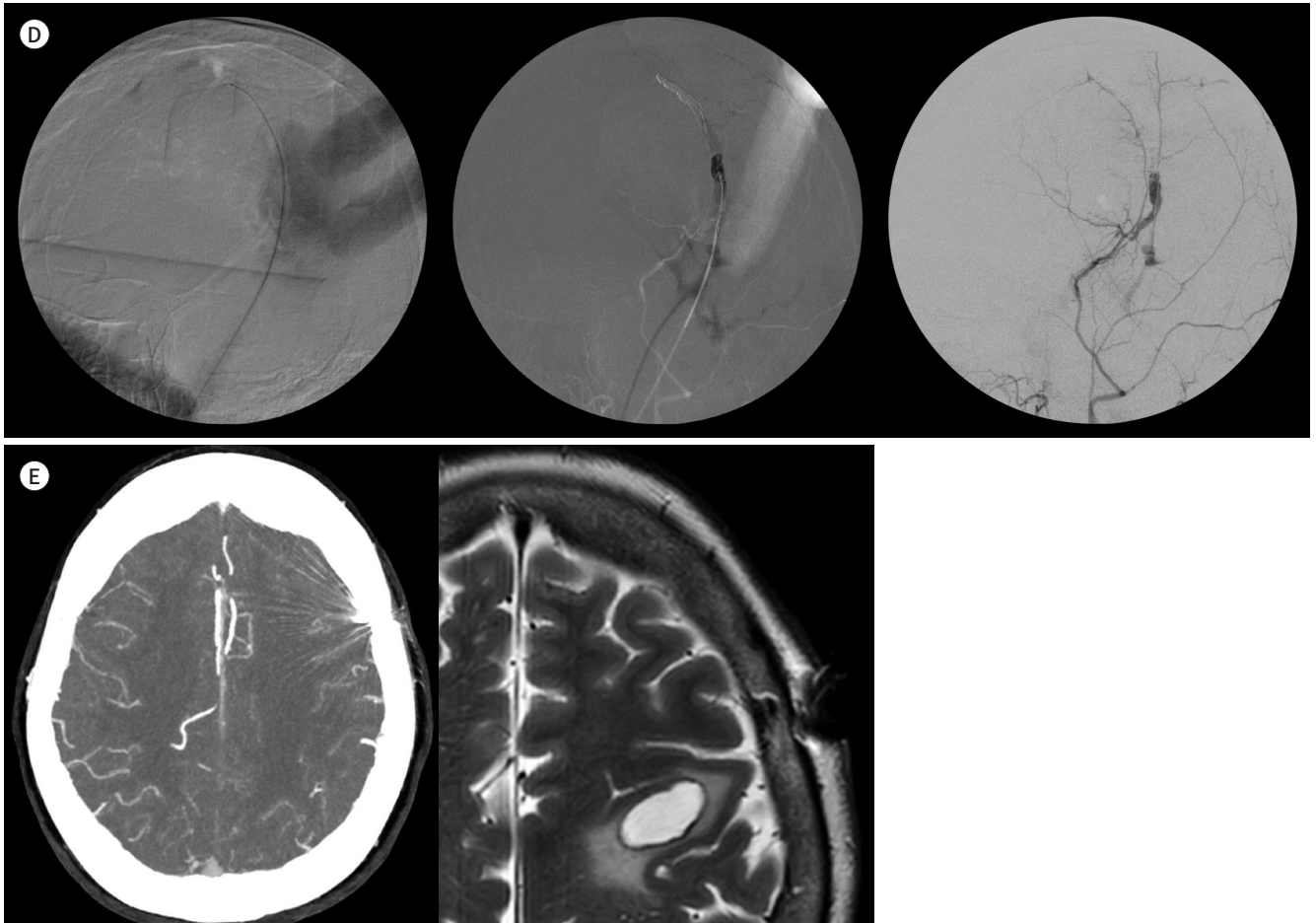
cess the dilated DV in the frontal bone. A high-speed drill with a diamond burr was used to decorticate the outer cortex of the skull over the diploic draining vein. Once the red, arterialized blood of the draining vein could be visualized through the thinned out cortical bone (Fig. 1C), a 5-Fr dilator was inserted into the exposed DV using the micropuncture technique. Venography performed through the dilator demonstrated the course of the draining vein and confirmed the appropriate access. An Excelsior 1018 Microcatheter (Stryker, Kalamazoo, MI, USA) was inserted at the midpoint of the diploic AVF in the retrograde direction (Fig. 1D, right image). To fix and stabilize the microcatheter, we tied the distal portion of microcatheter using polypropylene suture. The distal segment of the anterior temporal DV was embolized us-

Fig. 1. Transcranial embolization of a diploic AVF along the frontal bone, presented as intracranial hemorrhage, in an 81-year-old male with sudden dysarthria.

D. A microcatheter is inserted at the midpoint of the diploic AVF in a retrograde direction (left image). The distal segment of the anterior temporal DV is embolized using detachable coils (middle image). Immediately after embolization, angiography shows obliteration of the cortical venous reflux and stagnation of the contrast media at the proximal segment of the anterior temporal DV (right image).

E. Follow-up CT angiography and T2-weighted MRI, 3 months after embolization, show complete resolution of the dilated cortical veins at the left frontal regions.

AVF = arteriovenous fistula, DV = diploic vein



ing seven tornado detachable coils (5 mm × 2 mm and 6 mm × 2 mm, Cook Medical, Bloomington, IN, USA) (Fig. 1D, middle image).

Immediately after embolization, angiography showed obliteration of the cortical venous reflux and stagnation of contrast media at the proximal segment of the anterior temporal DV (Fig. 1D, left image). Follow-up CT angiography and MR scan 3 months after embolization showed complete resolution of the dilated cortical veins in the left frontal regions (Fig. 1E). Six months after embolization, the patient was symptom-free with no reported neurologic deficits.

This study was approved by the Institutional Review Board of our institution and the requirement for informed consent was waived (IRB No. KHNMC 2021-08-053).

DISCUSSION

DVs are valveless channels that travel between the inner and outer tables of the calvaria in the diploic space. These veins anastomose with each other and with an extensive network of microscopic venous channels. The DVs are divided into four major channels: frontal, anterior temporal, posterior temporal, and occipital DV. They communicate with the dural sinuses and pericranial veins and may become collateral routes when normal cerebral pathways are compromised (4).

AVFs involving DVs are rare and are referred to as diploic or intraosseous AVFs. Although the etiology remains unclear, several previously reported cases suggest that traumatic injuries, anatomical variations, and vascular pathologies (superior sagittal sinus thrombosis) can contribute to an environment suitable for the development of diploic AVFs (5). Rivera-Lara et al. (3) proposed a classification of diploic AVFs with three potential venous outflow patterns: dural sinus drainage only, extracranial drainage only, or dural sinus and extracranial drainage. Although anterior temporal DV usually drains through the sphenopalatine sinus or middle meningeal vein, the present case showed diploic AVF refluxed into the frontal cortical veins, resulting in serious venous congestion in the left frontal area.

Regarding the treatment of diploic AVFs, there have been several reports describing intraosseous AVFs (3, 5-7). From a literature review of the PubMed database using the search terms “diploic AVF” and “intraosseous AVF, 36 cases of diploic AVFs have previously been reported. Twenty-three of the 36 cases were treated by endovascular treatment via the transvenous route. Nine cases were treated by endovascular treatment via transarterial route. Two cases were treated by surgical resection, and two diploic AVFs were treated by direct cannulation via transosseous or transfacial approach. There were no reported complications relating to the treatment of diploic AVFs, and there was no recurrence of the diploic AVF during 4-day to 4-year follow-up.

Traditionally, endovascular embolization with a transvenous approach is considered the treatment of choice for most dural AVFs. In the transvenous approach, it is very important to define and access the abnormally dilated venous pouch with a microcatheter and to determine the venous connection with the abnormally dilated venous pouch. However, in the absence of an accessible venous channel, an alternative treatment approach should be considered. In our case, the retrograde approach to the sinus draining the diploic AVF via the ipsilateral internal jugular vein was not technically possible. Transarterial embolization of the diploic AVF could be applied as a palliative option for symptomatic relief when a diploic AVF is not technically accessible with transvenous catheterization. Because the main feeders of the diploic AVFs not only supply the diploic AVF, but also supply the facial structures and peripheral cranial nerves, transarterial embolization of the meningeal arteries could increase the risk of neurological injury, and most of the fistulas recur after transarterial embolization (3, 5-7). Furthermore, in our case, embolization of the feeding artery was difficult because the diploic AVF was fed by numerous small feeding arteries without dominant main feeders.

Direct cannulation of a calvarial DV via the transcranial approach was first reported for the embolization of dural AVF located along the sphenoid ridge by White et al. in 2020 (8). We

could not find other cases of embolization of diploic AVF via a transcranial direct approach from a literature review of the PubMed database. In our case, direct cannulation of a calvarial DV via the transcranial approach was successful for the embolization of the diploic AVF. It facilitated the precise access to the AVF through a small scalp incision and direct visualization of the draining DV. Under the CT-based navigation system, frontal craniotomy was easily performed to access the dilated DV in the frontal bone.

When there are multiple fistula niduses along the AVF, it is important to target the area that is the main cause of the clinical problem. Because there were multiple small arterial feeders along the entire length of the anterior temporal DV, we punctured the midpoint of the diploic AVF and focused the embolization on the distal segment of the anterior temporal DV directly connecting with the emissary vein. Immediately after embolization, angiography showed obliteration of the cortical venous reflux despite a small remnant of diploic AVF. As this plan was significantly effective, angiography after embolization showed complete obliteration of the cortical venous reflux despite a small remnant of diploic AVF.

Another tip of this procedure was that because there is no commercialized microcatheter of short length that can deliver coils, it was important to fix the microcatheter well in the operating room.

In conclusion, in our case, diploic AVF along the frontal bone was successfully treated with endovascular embolization via a transcranial approach with direct cannulation of the calvarial DV. Despite our limited experience, we can conclude that direct cannulation of the diploic AVF for embolization can be a safe and useful adjunct in diploic AVF therapy when percutaneous transarterial or transvenous approaches are not possible.

Author Contributions

Conceptualization, R.C., S.H.S., K.H.C.; data curation, J.J.I., R.C.; formal analysis, J.J.I., R.C., K.H.C.; investigation, J.J.I., R.C., K.H.C.; methodology, R.C., S.H.S., K.H.C.; project administration, R.C.; supervision, R.C.; writing—original draft, J.J.I.; and writing—review & editing, R.C.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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두개관 판사이정맥의 직접 관삽입술을 통한 유증상 골내 동정맥류의 색전술: 증례 보고

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판사이 동정맥류(diploic arteriovenous fistula) 혹은 골내 동정맥류(intraosseous arteriovenous fistulas)는 동정맥 단락의 한 형태로 드물게 발생한다. 판사이 동정맥류에서 누공은 뇌막동맥(meningeal artery)과 골내 판사이 정맥(intraosseous diploic vein) 혹은 이끝정맥(emissary vein) 사이에 형성되고, 누공의 핵은 골내에만 위치한다. 현재, 경막 동정맥류에 대한 대표적 치료 방법은 혈관내 색전술로 대부분이 대퇴동맥/대퇴정맥을 통하여 접근한다. 하지만 혈관내 색전술 시 접근 가능한 통로가 없는 경우에는 대체할 수 있는 다른 접근법을 고려해야 한다. 우리는 이번 증례에서 두개골내 판사이정맥을 직접 천자하여 혈관내 색전술로 치료된 판사이 동정맥류 증례를 보고하고자 한다.

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