

A case of dural arteriovenous fistula draining to the diploic vein presenting with intracerebral hemorrhage

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The authors report an unusual case of a dural arteriovenous fistula (dAVF) draining only to the diploic vein and causing intracerebral hemorrhage. A 62-year-old woman presented with disturbance of consciousness and left hemiparesis. Brain CT scanning on admission showed a right frontal subcortical hemorrhage. Digital subtraction angiography revealed an arteriovenous shunt located in the region around the pterion, which connected the frontal branch of the right middle meningeal artery with the anterior temporal diploic vein and drained into cortical veins in a retrograde manner through the falcine vein. The dAVF was successfully obliterated by percutaneous transarterial embolization with *N*-butyl-2-cyanoacrylate. The mechanism of retrograde cortical venous reflux causing intracerebral hemorrhage is discussed.

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IN the classic form of dural arteriovenous fistula (dAVF), the fistula is situated within the dura mater, making connections between meningeal arteries and venous sinuses, meningeal veins, or cortical veins. A dAVF draining to diploic veins (DVs) is relatively rare and occurs predominantly in the infratentorium due to a high incidence of transosseous emissary veins in the infratentorial bony structures, such as the clivus, petrous bone, and foramen magnum.^{2,3} By contrast, a dAVF involving DVs located in the supratentorial cranium is extremely rare.^{1,2,7,12} A dAVF with this type of venous drainage caused intracerebral hemorrhage in our case. Here, we report an extremely rare case of dAVF draining to the DV of the skull around the pterion causing intracerebral hemorrhage, and we discuss this mechanism.

Case Report

A 62-year-old woman was admitted to our hospital because of disturbance of consciousness and left hemiparesis with acute onset. The patient did not have a history of head trauma, surgery, or other diseases such as intracranial infection or hypertension. Her body temperature and blood pressure were within the reference range. On initial neurological examination, her consciousness was somnolent with dysarthria and left hemiparesis. Blood analysis

findings, including blood cell counts and coagulation cascades, were within the reference range. Brain CT scanning revealed an abnormal high-density area, indicating right frontoparietal subcortical hemorrhage (Fig. 1). Digital subtraction angiography (DSA) of brain vessels demonstrated an arteriovenous shunt around the pterion. This fistula was supplied by the anterior branch of the middle meningeal artery (MMA) and drained into the anterior temporal diploic vein (ATDV) (Fig. 2A and B). The DV reached the parietal convexity near the superior sagittal sinus (SSS) on the right side and drained into cortical veins through the falcine vein (Fig. 2C and D). The middle meningeal vein was not visualized. The shunt had no connection to the internal carotid artery. The SSS was patent with no stenosis. The DV was detected as a signal void of the vessel in the diploic layer of the parietal bone on T2-weighted MRI (Fig. 2E). These imaging findings indicated dAVF involving the ATDV. Intracerebral hemorrhage seemed to result from retrograde cortical venous reflux of the draining pathway. Transarterial embolization of the fistula was planned to prevent rebleeding.

Under local anesthesia, a 6-Fr Envoy guiding catheter (Cordis) was placed in the external carotid artery and a microcatheter (Excelsior SL-10, Boston Scientific) was navigated to the anterior branch of the MMA. Superse-

ABBREVIATIONS ATDV = anterior temporal DV; AVF = arteriovenous fistula; dAVF = dural AVF; DSA = digital subtraction angiography; DV = diploic vein; MMA = middle meningeal artery; NBCA = *N*-butyl-2-cyanoacrylate; SSS = superior sagittal sinus.

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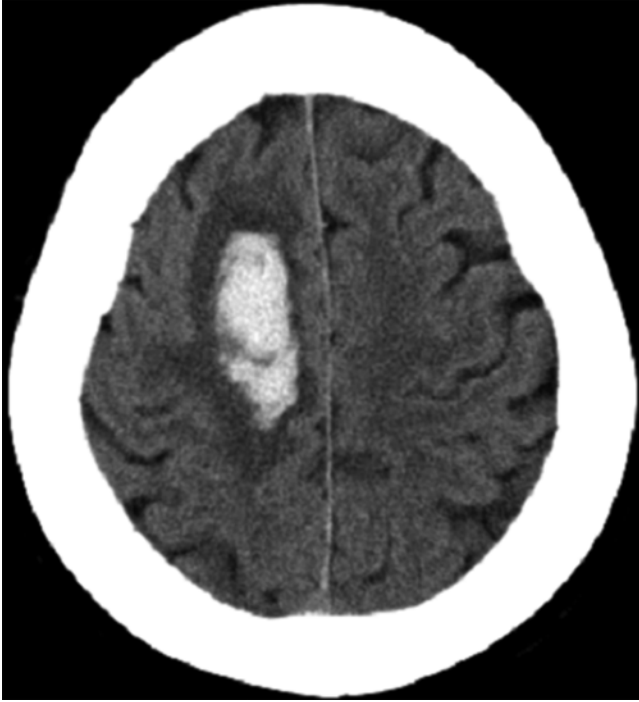


FIG. 1. Preoperative brain axial CT scan showing the right frontoparietal subcortical hemorrhage.

lective angiograms of the MMA clearly demonstrated multiple arterial channels of the MMA connecting to the dilating ATDV (Fig. 3A), eventually draining into cortical veins through the falcine vein. A diluted mixture of 25% *N*-butyl-2-cyanoacrylate (NBCA; Cordis Microvascular, Inc.) was injected into the MMA. A small amount of NBCA glue reached into the DV penetrating to the shunt (Fig. 3B). A right external carotid angiogram demonstrated a complete obliteration of the AVF (Fig. 3C).

Postoperative CT scanning revealed a small high-density area indicating a cast of NBCA glue in the diploic space of the skull on the right anterolateral side of the middle cranial fossa (Fig. 3D). The intracerebral hematoma was removed by endoscopic surgery 1 week after the endovascular procedure (Fig. 3E). Follow-up DSA 3 months after transarterial embolization demonstrated no recurrence of the AVF. The patient became ambulatory with the aid of a cane at the time of discharge. MR angiography showed no recurrence of the AVF 1 year after the treatment.

Discussion

We report a case of intracerebral hemorrhage caused by an idiopathic dAVF draining to the ATDV. A dAVF usually develops within the dura mater, making a connection between meningeal arteries and venous sinuses, dural veins, or cortical veins. A dAVF draining to only diploic

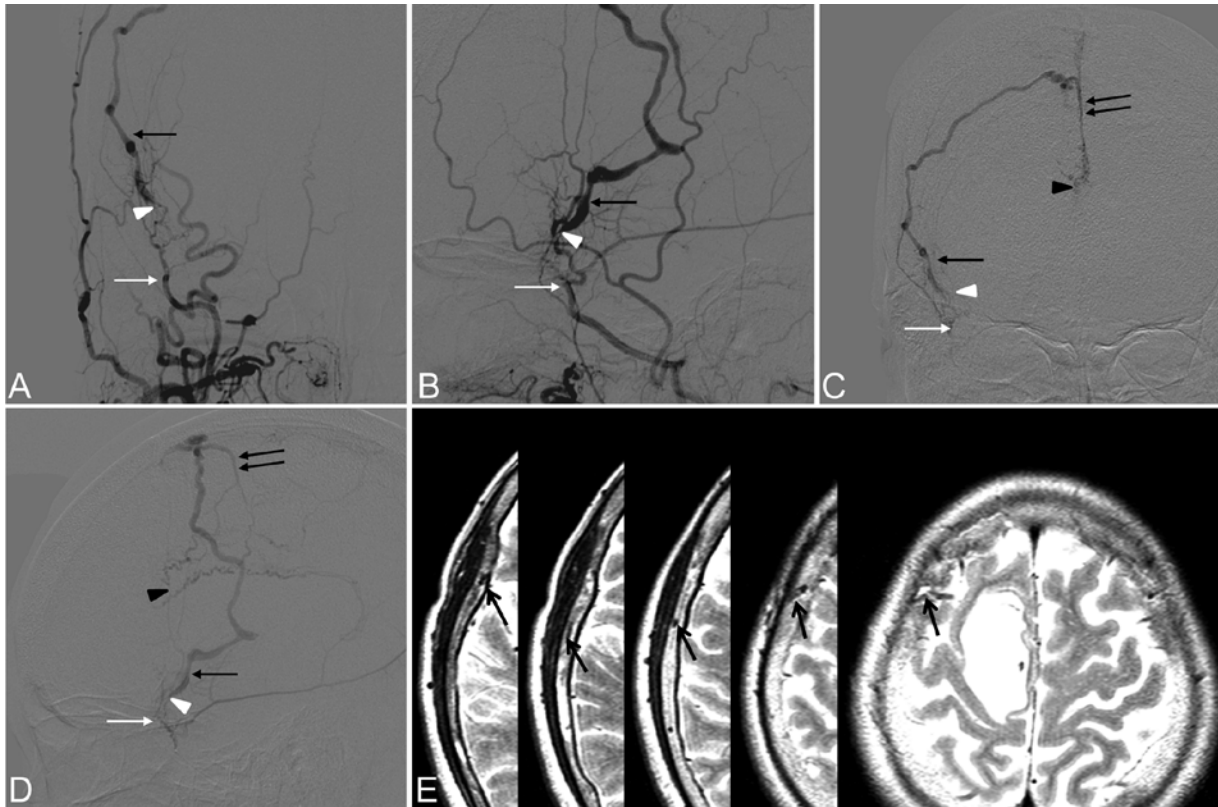


FIG. 2. A–D: Anteroposterior view (A) and lateral view (B) of the right external carotid angiograms before endovascular surgery, and anteroposterior view (C) and lateral view (D) of the superselective angiograms of the MMA, demonstrating an arteriovenous shunt (white arrowheads). The arteriovenous shunt is fed by the anterior branch of the MMA (white arrows) and connected with the ATDV (black arrow) around the pterion, draining into cortical veins (black arrowheads) through the falcine vein (black double arrows). E: Axial T2-weighted MR images, demonstrating a signal intensity void (arrows) of the vessel in the diploe of the parietal bone as the ATDV.

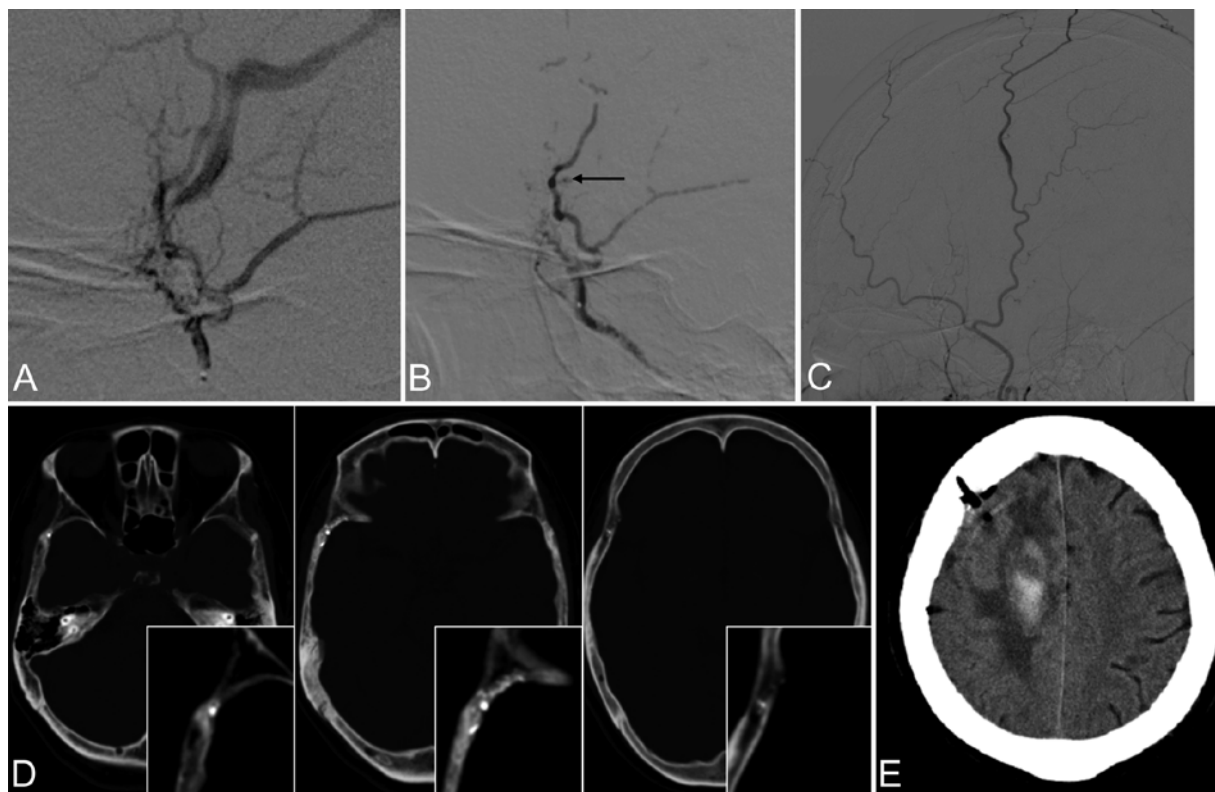


FIG. 3. **A:** Superselective angiogram of the right MMA obtained just before NBCA injection, showing multiple arterial channels flowing in the fistula from the MMA. **B:** Intraoperative angiogram obtained just after NBCA injection, demonstrating a cast of glue occluding the fistula including feeding arteries and the diploic vein (*black arrow*). **C:** Postoperative lateral view of the right external carotid angiogram, revealing a complete obliteration of dAVF. **D:** Postembolization axial CT scans showing a high-density area indicating a cast of NBCA glue in the diploic space of the skull on the right anterolateral side of the middle fossa. **Insets** are enlarged views of the lesion area. **E:** Brain CT scans obtained after endoscopic surgery, showing complete removal of the cerebral hematoma.

veins is extremely rare and is also referred to as a diploic AVF, intraosseous AVF, or intraosseous dAVF.¹⁻³

The frontal DV and ATDV form an anterior diploic venous system converging in the pterional area, which connects the SSS with the sphenoparietal sinus. The posterior diploic venous system converging in the asterion is formed by the posterior temporal DV and occipital DVs, and connects the posterior SSS with the transverse and sigmoid sinuses.⁵ DVs communicate with the dural sinuses and pachymeningeal veins and pericranial veins via emissary veins.⁴ Under normal venous draining conditions, DVs are

usually invisible on DSA studies. In pathological states such as sinus thrombosis,⁶ trauma,⁷ and meningiomas,¹⁰ however, DVs can be dilated by opening diploic channels.⁵ The present case indicates that dAVFs can be a pathological condition causing a dilation of DVs.

Dural AVFs draining to diploic veins rarely cause intracerebral hemorrhage since DVs have no direct connection with cortical veins under physiological conditions. To our knowledge, only 20 cases of dAVF draining to DVs have previously been reported. Of these 20 cases, 4 cases were dAVFs in the supratentorium^{1,2,7,12} (Table 1), and

TABLE 1. Literature review of supratentorial dAVF draining to the diploic vein

Authors & Year	Age (yrs), Sex	Symptoms	Predisposing Factor	Feeder	Site of Shunt	Drainer	Treatment
Benndorf & Lehmann, 2004	59, F	Subgaleal hematoma	No	MMA, MCA	Parietal	DV	TVE
Burger et al., 2005	28, F	Headache, tinnitus	Pregnancy	STA, MMA	Parietal	DV, SCV	Surgery, TVE
Ishii et al., 1976	65, M	Incidental	Trauma	MMA	Parietal	DV	Not operated
Shim et al., 2011	46, F	Headache, tinnitus	Yelling	MMA	Frontal	DV, MMV	TAE
Present study	62, F	Intracerebral hemorrhage	No	MMA	Frontal	DV	TAE

MCA = middle cerebral artery; MMV = middle meningeal vein; SCV = subcutaneous vein; STA = superficial temporal artery; TAE = transarterial embolization; TVE = transvenous embolization.

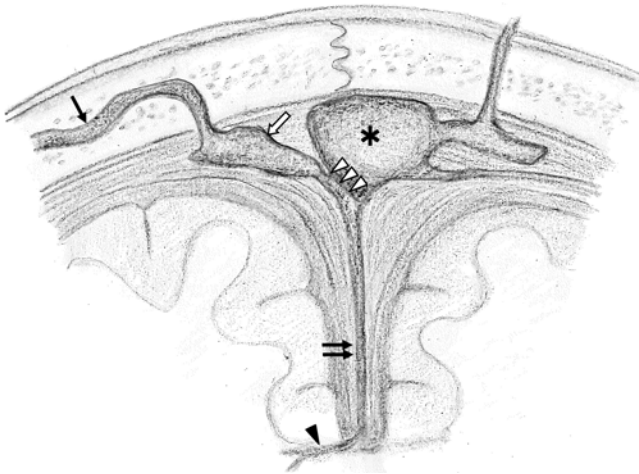


FIG. 4. Schematic illustration of a hypothetical draining pathway to cortical veins in the present dAVF. The enlarged ATDV (black arrow) might flow into the dural venous sinuses via emissary veins without connections with the SSS (asterisk). The falcine vein (black double arrows) once might have communicated with both the venous lacuna (white arrow) and the SSS. In the process of the maturation of the arteriovenous shunt, the occlusive change of drainage site might occur at the channel between the venous lacuna and the SSS (white arrowheads). The venous lacuna then might be isolated from the SSS, and consequently the shunt flow might be directed from the venous lacuna toward cortical veins (black arrowhead) via the falcine vein. Copyright Rie Yako. Published with permission.

there were no case reports in which the dAVF drained only to the diploic veins, causing intracerebral hemorrhage.

In the present case, we propose a possible mechanism for intracerebral hemorrhage as follows. The enlarged DV might have connections not with the SSS itself but with the venous lacuna via emissary veins. The falcine vein might have communicated with both the venous lacuna and the SSS originally. In the process of the maturation of the arteriovenous shunt, the occlusive change of drainage site might occur at the channel between the venous lacuna and the SSS. The venous lacuna might be isolated from the SSS, and consequently the shunt flow might be directed from the venous lacuna to the cortical veins via the falcine vein (Fig. 4). Eventually, these venous drainage changes involving the occlusive process might result in intracerebral hemorrhage. In dAVFs, the occlusive process of the draining system frequently occurs during the development of the shunt by unknown mechanisms. In the sinus type of dAVF, the sinus may become occluded due to possible mechanisms including thrombogenesis associated with activated coagulopathy and hemodynamic hypertrophy of the sinus wall.^{8,9,11} In our case with the nonsinus type, the occlusive change may occur through similar mechanisms.

Conclusions

Dural AVFs involving DVs causing intracerebral hemorrhage are extremely rare. The putative occlusive change of the venous draining system might be related to the underlying mechanism for cerebral hemorrhage.

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Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Yako. Acquisition of data: Yako. Analysis and interpretation of data: Yako. Drafting the article: Yako. Critically revising the article: Yako, Masuo, Nakao. Reviewed submitted version of manuscript: Yako, Masuo, Nakao. Approved the final version of the manuscript on behalf of all authors: Yako. Administrative/technical/material support: Yako, Kubo, Nishimura, Nakao.

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