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De novo Dural Arteriovenous Fistula at the Drainer Site after Embolization of Brain Arteriovenous Malformation

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Abstract

We report a rare case of de novo ectopic dural arteriovenous fistula (dAVF) that developed late after the endovascular embolization of a brain arteriovenous malformation (AVM). A 25-year-old man with severe chronic headache was diagnosed with an unruptured right frontal AVM located on the medial side of the Sylvian fissure. The AVM was completely occluded using transarterial liquid embolization. However, 12 months after the embolization, the patient developed right pulsatile tinnitus. Angiography revealed a de novo dAVF at the convexity, distant from the original AVM nidus. The dAVF was supplied by multiple dural arteries and a small pial contribution and drained into a cortical vein previously used as the AVM's drainage route. The fistula was successfully treated with additional embolization and radiosurgery. It is well known that large, superficial AVMs tend to involve meningeal arterial supply. However, this case demonstrated delayed ectopic dAVF formation along a draining vein far from the nidus. Although the exact etiology remains unclear, drastic hemodynamic changes in cortical veins previously used as shunt draining route may induce unexpected angiogenesis, leading to the formation of such an unusual de novo dAVF. Even after successful AVM occlusion, long-term follow-up imaging is important, and the possibility of dAVF formation would offer stronger clinical guidance.

Keywords
- ► angiogenesis
- ► brain arteriovenous malformation
- ► de novo dAVF
- ► dural arteriovenous fistula
- ► embolization

Introduction

Dural arteriovenous fistulas (dAVFs) commonly develop after sinus thrombosis, surgery, trauma, and infection of adjacent air sinuses.[1–3] In some cases, dural feeders contribute to the supply of brain arteriovenous malformation (AVM) following AVM treatment.[4–7] Additionally, the dAVFs have been reported to form in typical locations as a complication of endovascular embolization.[8] However, the development of a dAVF after AVM embolization, sharing the same drainage route but occurring at a separate site from the original AVM, is rare. Here, we describe a case of de novo ectopic dAVF that developed 1 year after a successful AVM embolization, in the absence of any apparent causal events or risk factors.

Case Presentation

A 25-year-old man with a severe chronic headache was diagnosed with a right frontal AVM via magnetic resonance angiography (MRA) at a local hospital. Digital subtraction angiography (DSA) revealed a right frontal AVM located on the medial side of the Sylvian fissure (►**Fig. 1**). The AVM was

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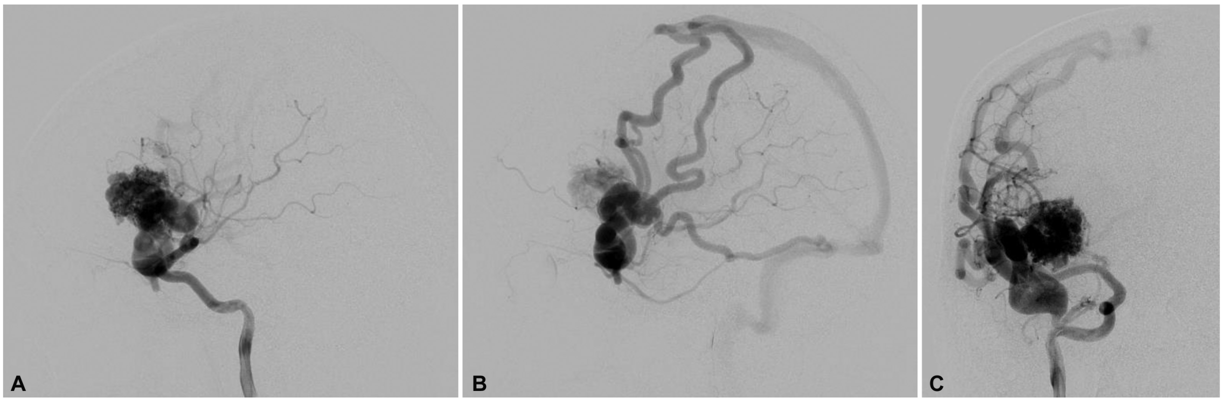

Fig. 1 Preoperative angiogram of the right internal carotid artery ([A, B] lateral views [A: early phase, B: late phase], [C] anteroposterior view) showing a high-flow AVM in the medial frontal lobe. The AVM is fed by the middle cerebral artery and drains into the superior sagittal sinus and transverse sinus via superficial cortical veins. AVM, arteriovenous malformation.

supplied by several feeders from the right middle cerebral artery (MCA) and drained into the superior sagittal sinus (SSS) via two large cortical veins, into the sigmoid sinus via the basal vein of Labbe, and into the straight sinus via the basal vein of Rosenthal. Since this AVM was classified as that of Spetzler–Martin Grade III and was a potential cause of the patient's severe headache, we decided to pursue radical treatment, including the endovascular embolization.

Under general anesthesia, a 6-Fr guiding catheter was advanced into the right internal carotid artery via a transfemoral approach. A Marathon microcatheter (Medtronic, Irvine, California, United States) and a Magic microcatheter (Balt, Montmorency, France) were selectively navigated into the feeders of the right MCA. Two fistulous feeders were embolized using 25% diluted n-butyl cyanoacrylate (NBCA). Next, a Scepter balloon catheter (4×7 mm; Terumo, Tokyo, Japan) was advanced into the main feeder, and Onyx 18 (Medtronic, Irvine, California, United States) was injected into the nidus under flow control with balloon inflation, resulting in complete occlusion of the nidus. The final angiogram showed no arteriovenous shunting and marked contrast stagnation in cortical drainers (►**Fig. 2**). Follow-up

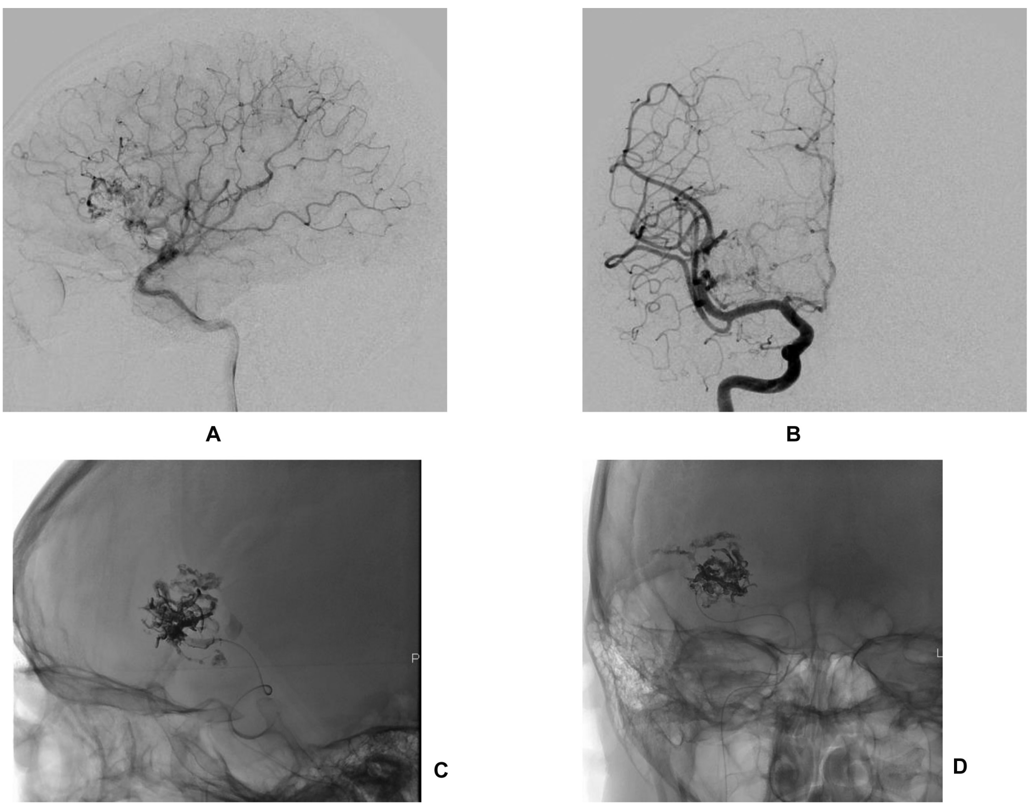

Fig. 2 Postoperative angiogram of the right internal carotid artery ([A] lateral view, [B] A-P view) demonstrating complete occlusion of the AVM with no abnormal shunts. Skull X-ray images ([C] lateral view, [D] A-P view) show the cast of liquid embolic materials filling the nidus. Notably, no embolic materials migrated into any draining veins. A-P, anteroposterior; AVM, arteriovenous malformation.

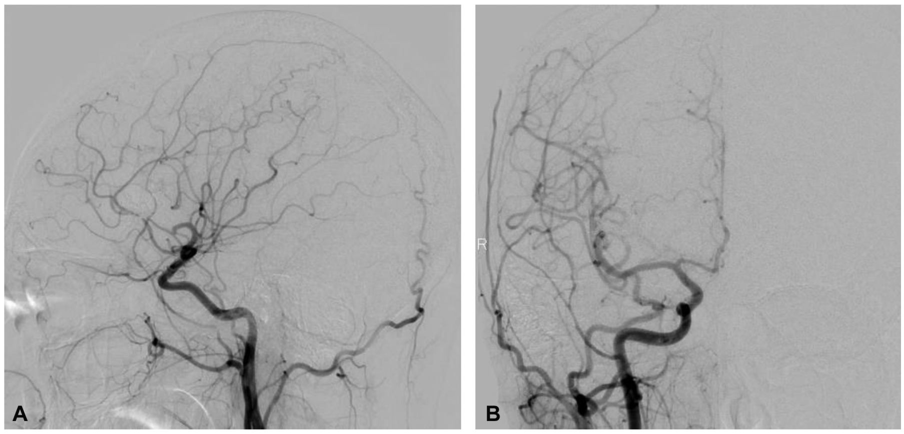

Fig. 3 Follow-up angiogram of the right common carotid artery ([A] lateral view, [B] A-P view) taken 7 days postoperatively, confirming no recanalization of the AVM. Importantly, no shunts from meningeal feeders originating from the external carotid artery are visible. A-P, anteroposterior; AVM, arteriovenous malformation.

angiography on postoperative day 7 confirmed no recurrence (►**Fig. 3**). No abnormal vascular networks supplied by either the external or internal carotid systems were observed, but the vein of Trolard was less opacified.

The postoperative course was uneventful. However, abnormal hypervascularity was observed in the distal portion of the right dural artery on magnetic resonance imaging performed 7 months later (►**Supplementary Fig. S1**, available in the online version). The patient developed right-sided pulsatile tinnitus 12 months after embolization. MRA revealed an abnormally dilated external carotid system and an arterialized cortical vein in the right Rolandic area. DSA showed no recurrence of AVM, but identified an abnormal arteriovenous shunt located at a cortical vein (vein of Trolard) (►**Fig. 4**). This de novo dAVF was fed by the right middle meningeal artery, middle deep temporal artery (MDTA), superficial temporal artery (STA), and left STA. Additionally, fine pial feeders from the branches of the right MCA contributed to the shunt, which did not correspond with the AVM feeders. The location of the dAVF was entirely

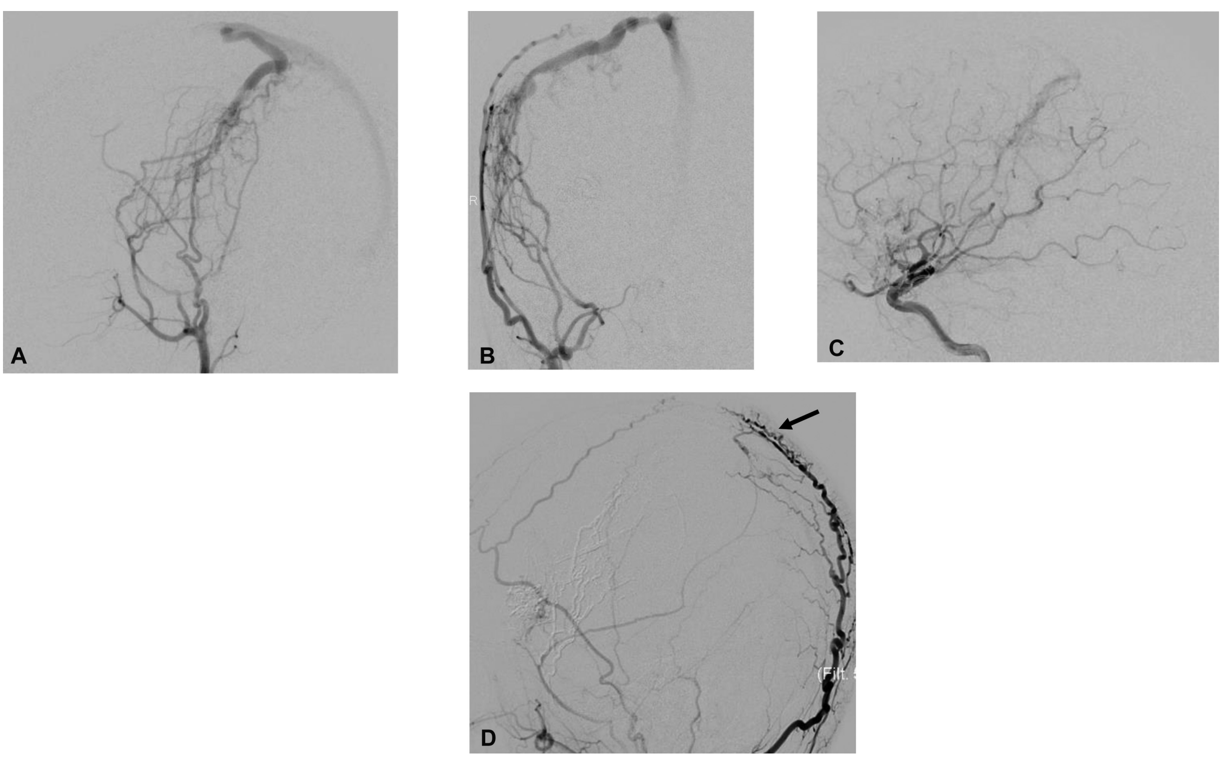

Fig. 4 Right external carotid artery angiogram taken 12 months after AVM embolization revealing a de novo dural arteriovenous shunt at the right parietal convexity ([A] lateral view, [B] A-P view) fed by branches of the middle meningeal artery. The right internal carotid angiogram (C) shows a small contribution from dural branches of the middle cerebral artery to this lesion, with no recurrence of the AVM. Additionally, another “daughter” dAVF (arrow), fed by the occipital artery, is identified at the right parietal apex (D). A-P, anteroposterior; AVM, arteriovenous malformation; dAVF, dural arteriovenous fistula.

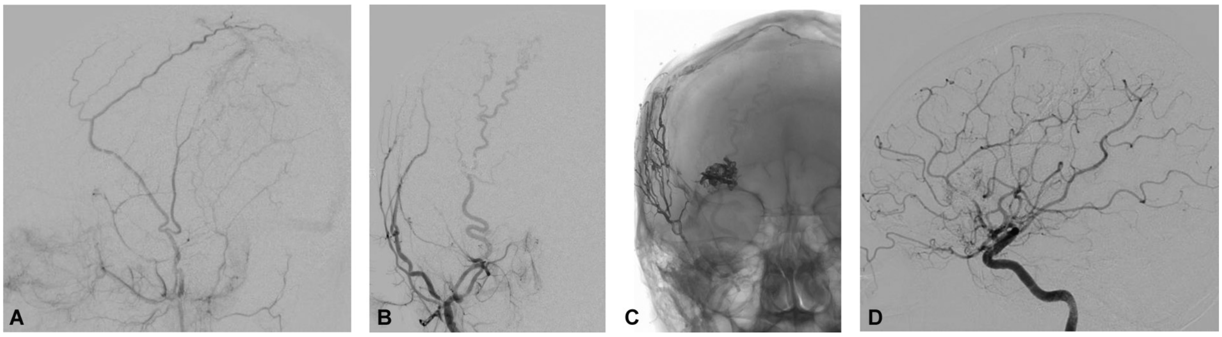

Fig. 5 Postoperative angiogram of the external carotid artery ([A] lateral view, [B] A-P view) showing almost complete occlusion of the main dAVF. The cast of embolic materials injected into the dAVF is visible on the postoperative X-ray image (C), located far from the AVM nidus. A right internal carotid angiogram taken 3 months later (D) confirms the absence of abnormal shunts at both the AVM and dAVF sites. A-P, anteroposterior; AVM, arteriovenous malformation; dAVF, dural arteriovenous fistula.

different from that of the treated AVM; however, its drainage route was one of the same cortical veins flowing into the SSS that had been used as an AVM drainage pathway and had experienced extreme flow reduction following AVM embolization. Another dAVF, fed by the right occipital artery, was also identified at the parietal apex, draining into SSS via emissary veins (►**Fig. 4**). Transarterial embolization was performed for the dAVF at the right convexity. Bilateral STA feeders were embolized with NBCA, followed by embolization of the right MDTA feeders using Onyx, which penetrated into the draining vein. Immediately after the procedure, almost all shunts disappeared, along with the patient's tinnitus (►**Fig. 5**). Three months after the second embolization, follow-up angiography showed no recurrence of AVM or convexity dAVF, except for the small residual dAVF at the SSS (►**Fig. 5**). Additional stereotactic radiosurgery was performed to treat this remaining lesion.

Discussion

Superficial, large, and high-flow AVMs often receive meningeal blood supply from the external carotid artery (ECA). These meningeal feeders can develop in response to AVM growth or following incomplete surgical intervention.[4] The reported prevalence of meningeal blood supply in AVMs varies, with recent studies estimating rates 6.6 and 7% in recent reports.[5,6] Moreover, Söderman et al reported the development of dural feeders following AVM embolization.[7] In their study, 32 patients with no initial transdural blood supply underwent at least one embolization, and 8 (25%) developed de novo transdural blood supply within 12 months postembolization. Paramasivam et al studied 16 patients with pial AVFs treated via endovascular embolization and found that 4 developed de novo dAVFs (not present before treatment) during the treatment or follow-up period.[8] They concluded that acute hemodynamic changes postembolization could trigger de novo dAVF formation. Their theoretical framework suggests that sinus thrombosis, venous hypertension, or intrasinus hypertension plays a role in dAVF pathogenesis, as supported by both clinical observations and experimental studies in rat models.[9–11] In this model, the slow blood flow and thrombosis in the venous channels following embolization may cause localized endothelial cell ischemia and segmental venous hypertension, leading to the release of angiogenetic factors.[12–14] Buell et al suggested three potential pathophysiological mechanisms for embolization-induced angiogenesis in AVMs: (1) hypoxia-mediated angiogenesis, (2) inflammatory-mediated angiogenesis, and (3) hemodynamic-mediated angiogenesis.[15] After embolization, hypoxia within the obliterated portion of the nidus may upregulate hypoxia-inducible factor-1a, vascular endothelial growth factor (VEGF), and fetal liver kinase-1 (FLK-1), all of which stimulate angiogenesis. Additionally, inflammatory reactions within the AVM vessel walls may further promote angiogenesis. Increased blood flow and shear stress in non-occluded portions of the AVM postembolization may also contribute to neovascularization by upregulating VEGF and FLK-1. Buell et al hypothesized that embolization-induced angiogenesis likely results from a combination of these mechanisms. In our case, the inflammatory effects of embolic materials are unlikely to have played a significant role, as the nidus was located medially and did not attach to the dura, and liquid agents did not migrate into the cortical veins. Furthermore, the hypothesis that occult meningeal arterial supply was "unmasked" following embolization does not apply, as the de novo feeders did not contribute to AVM reperfusion.

Although selective ECA angiography images were not recorded, our patient had no meningeal feeders supplying the AVM from the ECA on the initial angiogram. A follow-up angiogram on postoperative day 7 demonstrated persistent complete occlusion of the AVM without pial or dural feeders, and cortical draining veins appeared normalized without evidence of sinus thrombosis. Despite this, a de novo dAVF developed over the following 12 months, using the distal part of the same drainage route as the treated AVM—a drainage pathway that had initially regressed after embolization (►**Fig. 6**). One possible mechanism for this phenomenon is that drastic hemodynamic change in cortical veins previously used as a shunt draining route and flow stagnation-induced vascular remodeling triggered unexpected angiogenesis.

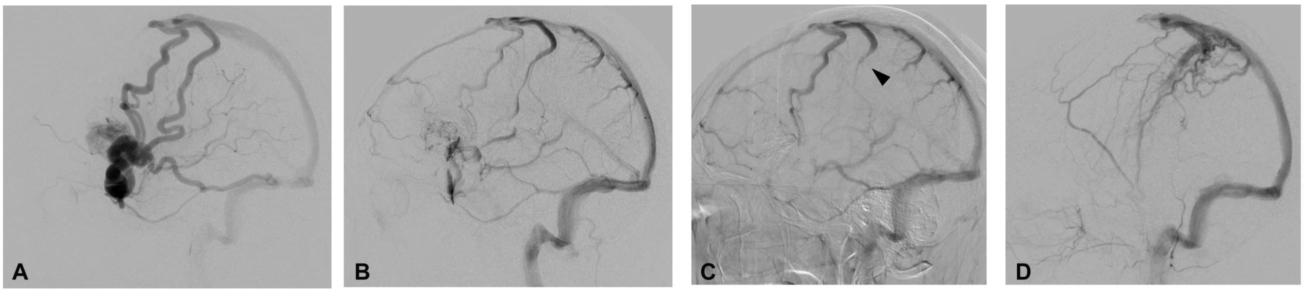

Fig. 6 Angiographic changes in the cortical vein, previously the drainage route of the AVM, and subsequent development of a de novo dAVF. (A) Preoperative angiogram showing multiple superficial cortical drainage routes of the frontal AVM. (B) Postoperative angiogram demonstrating marked flow reduction and stagnation in the draining veins. (C) Follow-up angiogram 7 days postembolization showing normalized cortical veins, with reduced opacification of the vein of Trolard (arrowhead). (D) Angiogram taken 12 months later revealing a de novo dAVF at the expanded vein of Trolard. AVM, arteriovenous malformation; dAVF, dural arteriovenous fistula.

However, it remains unclear whether venous flow stagnation alone, without thrombosis, is sufficient to promote such localized, excessive induction of meningeal feeders. This unique pathological course—characterized by delayed ectopic angiogenesis and the re-establishment of an arteriovenous shunt in a temporarily normalized cortical vein—may provide insight into the underlying etiology of dAVF formation.

Conclusion

We report a case of de novo ectopic dAVF formation following endovascular embolization of an AVM. In this case, the dAVFs developed along the AVM's former drainage route, using the same venous pathway. This suggests that angiogenesis at the AVM drainer, triggered by hemodynamic changes after embolization, may play a role in dAVF development. As such, de novo dAVFs can mature even after successful AVM embolization, highlighting the importance of careful long-term follow-up and management to detect and address unexpected vascular changes.

Authors' Contributions

N.M., R.K., and F.A. treated the patient, designed and conceptualized the study, and analyzed the data, T.I. drafted the manuscript for intellectual content, S.M. revised the manuscript, and T.W. read and approved the final version.

Ethical Approval

This study waived approval of the Institutional Review Board (IRB), and IRB approval for case reports is not required at our institution. We obtained individual written informed consents for the procedure and publication.

Conflict of Interest

S.M. reported consulting fees from NIPRO and payment or honoraria from Kaneka Medics, Japan Medtronic, Daiichi-Sankyo. S.M. had advisory role of NIPRO Co. and received lecture fees from Kaneka Medics, Japan Medtronic, Daiichi-Sankyo. All authors who are members of the Japan Neurosurgical Society (JNS) have registered self-reported COI disclosure statement forms through the website for JNS members.

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