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Technical Note

A Case of Curative Onyx Embolization for Tentorial dAVF via Low-flow Feeders with Temporary Balloon Occlusion of High-flow Feeders

Ryosuke Ogura,[1,2] Hitoshi Hasegawa,[1] Shunsuke Kumagai,[2] Haruhiko Takahashi,[1,2] Hidemoto Fujiwara,[2] Junichi Yoshimura,[1,2] and Yukihiko Fujii[1]

Objective: For curative Onyx embolization of dural arteriovenous fistulas (dAVF) with multiple feeders, it is essential to select the optimal target artery as well as to control the blood flow at the fistula point. We report a case of tentorial dAVF (TdAVF) treated by Onyx embolization under flow control using balloon catheters.
Case Presentation: A 66-year-old male was admitted to our hospital for treatment of TdAVF detected incidentally by MRI, which revealed a dilated and tortuous vein around the cerebellum. Cerebral angiography demonstrated a TdAVF, fed mainly by bilateral middle meningeal arteries (MMA) and bilateral occipital arteries (OA), with the fistula point at the torcular and venous drainage to the two superior vermian veins (SVVs). Onyx 18 was injected from the low-flow feeder of the MMA under flow control by occluding the high-flow feeder of the OA using balloon catheters, obliterating the arteriovenous shunt.
Conclusion: In treatment of TdAVF involving low- and high-flow feeders, Onyx embolization via the low-flow feeder with temporary balloon occlusion of other high-flow feeders is a useful method. This technique makes it easier for Onyx to penetrate the fistula point.

Keywords ▶ tentorial dural arteriovenous fistula, torcular, Onyx, balloon catheter, DeFrictor

Introduction

Although the incidence of tentorial dual arteriovenous fistula (TdAVF) is low, it requires positive therapeutic intervention because it exhibits aggressive features frequently with hemorrhagic onset.[1)] As TdAVF is often a non-sinus fistula that directly drains retrogradely into a cortical vein not through sinuses, it is rarely treated by transvenous embolization (TVE) and more frequently by a combination of transarterial embolization (TAE) using n-butyl-2-cyanoacrylate (NBCA) or Onyx, direct surgery and radiation therapy. In this report, we present a case of TdAVF with multiple feeders that was treated by curative Onyx embolization with temporary occlusion of high-flow feeders, which were not the target vessels, using balloon catheters.

[1]*Department of Neurosurgery, Brain Research Institute, University of Niigata, Niigata, Niigata, Japan*
[2]*Department of Neurosurgery, Nagano Red Cross Hospital, Nagano, Nagano, Japan*

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Corresponding author: Ryosuke Ogura. Department of Neurosurgery, Brain Research Institute, University of Niigata, 1-757 Asahimachi, Chuo-ku, Niigata, Niigata 951-8585, Japan
Email: oguryou@bri.niigata-u.ac.jp

Case Presentation

The patient was a 66-year-old male with a history of multiple myeloma (stage II). The patient was referred to us as he was suspected to have a dural arteriovenous fistula (dAVF) by head MRI performed for intracranial examination during maintenance therapy for multiple myeloma at the hematology department of our hospital.

On the initial physical examination, he was conscious, and no obvious neurological symptoms, including headache and tinnitus, were noted.

Concerning imaging examinations, head MRI revealed no clear abnormality in the brain parenchyma, but a large

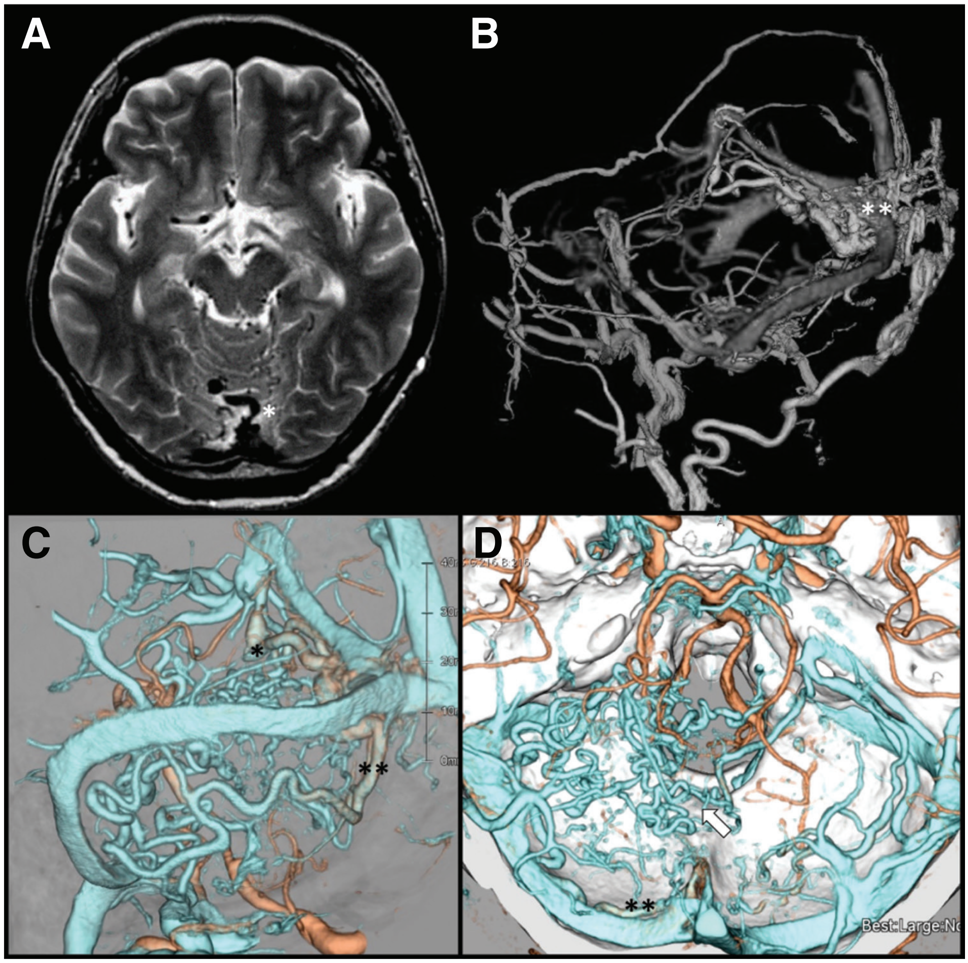

Fig. 1 T2-weighted image (**A**) showed markedly enlarged cerebellar veins and two SVVs (white asterisk). Fused 3D-DSA (**B**) showed a tentorial arteriovenous fistula, fed mainly by the bilateral MMA and bilateral OAs with the fistula point at the torcular (double white asterisks), draining into two SVVs. Fused 3D CTA and venography (**C** and **D**) showed the venous drainage into the two SVVs, the inferior vermian vein (double black asterisks), and markedly dilated cerebellar veins (white arrow). MMA: middle meningeal arteries; OAs: occipital arteries; SVVs: superior vermian veins

number of low voids were observed on the superior surface of the cerebellum, and the superior vermian veins (SVVs) were dilated (**Fig. 1A**). On MRA, peripheral transosseaus branches of the bilateral occipital arteries (OA) were developed, and dilated SVVs and the straight sinus were delineated. By cerebral angiography, the lesion was diagnosed as straight sinus-type TdAVF draining primarily through two irregularly dilated SVVs with a venous pouch into the straight sinus, having a fistula point at the cerebellar tentorium to the left of the confluence, and fed by the posterior convexity branches (PCB) of the bilateral middle meningeal arteries, bilateral OAs, right posterior meningeal artery (PMA), a dural branch of the left superior cerebellar artery (SCA), and the left tentorial artery (**Figs. 1B** and **2A–F**). In addition to the SVVs, there were two other drainage routes: from the inferior vermian vein via the lateral mesencephalic vein to the basal vein of Rosenthal (BVR), and from a cortical vein on the superior surface of the cerebellum via the right petrosal vein to the right superior petrosal sinus (**Fig. 1C** and **1D**). The patient was asymptomatic, but as reflux of superficial veins of the brain and dilatation of draining veins were observed, the risk of hemorrhage was judged to be high, and treatment was considered necessary.

As the lesion was a non-sinus fistula, we planned to perform TAE using Onyx via the bilateral MMA PCBs. To facilitate penetration of Onyx into the fistula point, the bilateral OAs were temporally occluded with balloons. Under general anesthesia, 6Fr long sheaths were placed in the bilateral femoral arteries, and the patient was systemically heparinized. A 6Fr Road Masters 90 cm (Goodman, Aichi, Japan) was navigated to each of the bilateral external

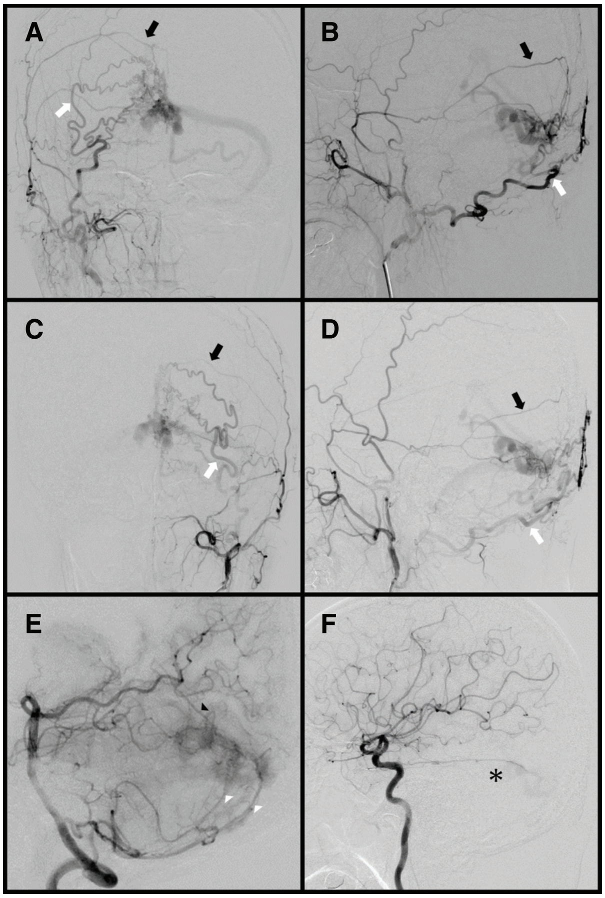

Fig. 2 DSA anteroposterior (**A**) and lateral (**B**) projections of the right ECA, and anteroposterior (**C**) and lateral projections (**D**) of the left ECA showed the TdAVF supplied by the bilateral MMA (black arrow) and the bilateral OAs (white arrow). DSA lateral projection of the right VA (**E**) and lateral projection of the left internal carotid artery (**F**) showed the feeders from the right PMA (black arrowhead), the tentorial branch of SCA (white arrowhead), and the left tentorial artery (asterisk). ECA: external carotid artery; MMA: middle meningeal arteries; OAs: occipital arteries; PMA: posterior meningeal artery; SCA: superior cerebellar artery; TdAVF: tentorial dural arteriovenous fistula; VA: vertebral artery

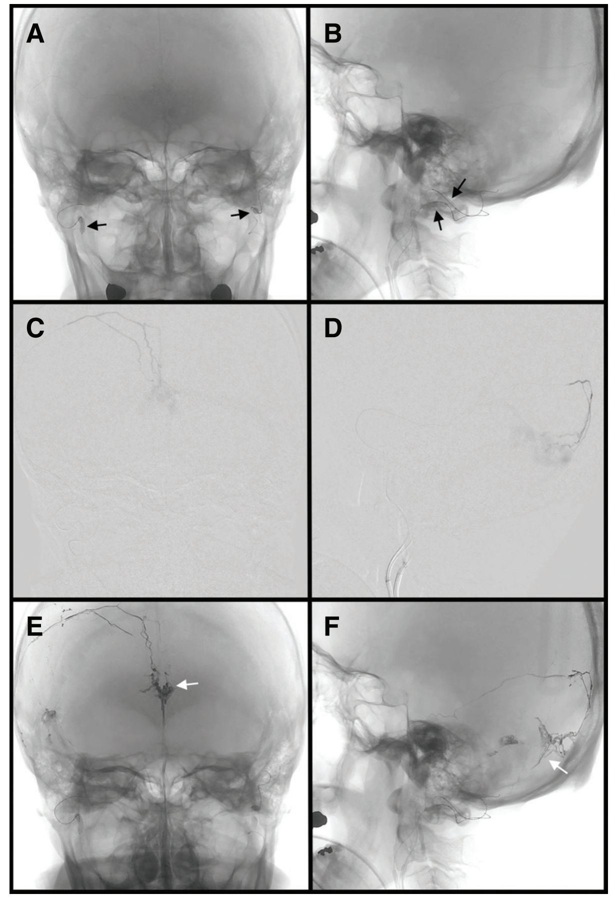

Fig. 3 The anteroposterior (**A**) and lateral view (**B**) of craniography showing the balloon catheters that were inflated at the bilateral OAs (black arrow). Anteroposterior (**C**) and lateral (**D**) views of the right MMA angiography showed the fistula point and the draining veins. Anteroposterior (**E**) and lateral (**F**) views of craniography demonstrating that Onyx did not penetrate the venous pouch through the fistula point and proceeded to the extradural drainer (white arrow). MMA: middle meningeal artery; OAs: occipital arteries

carotid arteries and a Scepter C 4 × 10 mm (Terumo, Tokyo, Japan) was navigated to each of the bilateral OAs using ASAHI CHIKAI14 (Asahi Intecc, Aichi, Japan), and the OAs were occluded by inflating them (**Fig. 3A** and **3B**). A DeFrictor Nano Catheter (Medico's Hirata) was inserted, guided by ASAHI CHIKAI10 and ASAHI CHIKAI008 (Asahi Intecc), and placed at the distal end of the right MMA PCB (**Fig. 3C** and **3D**). TAE was initiated by the plug and push technique using Onyx18 (Covidien, Minnesota, USA). Onyx penetrated to the area considered to be near the fistula point around the confluence but did not reach the SVV beyond the fistula point. It also reached a point near the right transverse sinus-sigmoid sinus, suggesting drainage into the venous sinuses, but angiography confirmed that the drainers were dilated veins in the vicinity rather than the trunk of the venous sinuses (**Fig. 3E** and **3F**). Embolization from the right side was finished after injecting 2.03 mL of Onyx. The DeFrictor was able to be withdrawn without problem. In expectation of more complete embolization of the fistula point, the DeFrictor was navigated to the left MMA using ASAHI CHIKAI10 and advanced to the periphery of the left MMA PCB (**Fig. 4A** and **4B**). Onyx18 was also injected by the plug and push technique. Although time was needed to form a plug, Onyx was delivered to the draining veins beyond the fistula point. When the SVV, the affected vein, was filled with Onyx to an extent, a clear arteriovenous shunt disappeared. Embolization was finished after injecting 2.83 mL of Onyx from the left side (**Fig. 4C** and **4D**). A total of 4.86 mL of Onyx was used. After removing the DeFrictor, releasing balloon

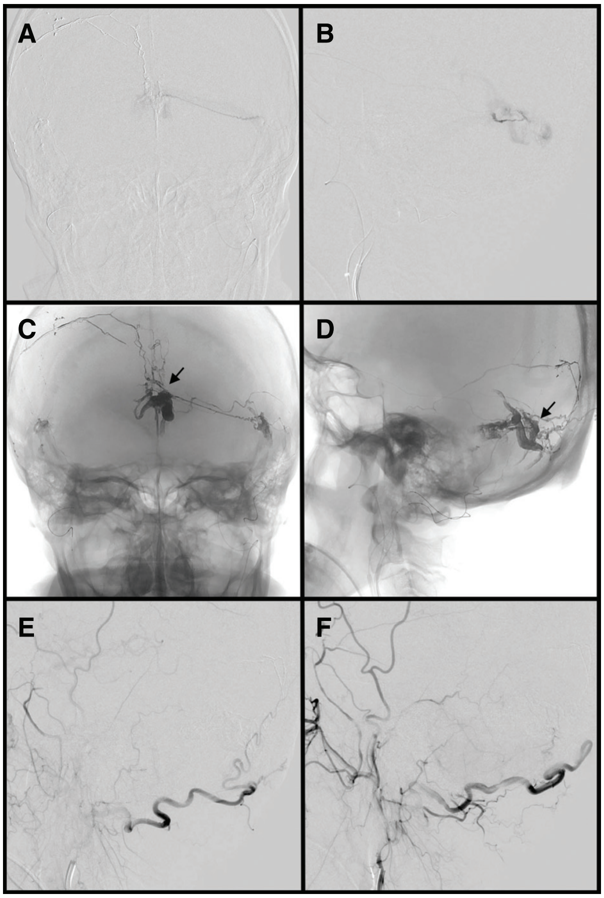

Fig. 4 Anteroposterior (**A**) and lateral (**B**) views of the left MMA angiography showed the fistula point and the draining veins. Anteroposterior (**C**) and lateral (**D**) views of craniography illustrated that Onyx penetrated the venous pouch through the fistula point (black arrow) and proceeded to the extradural drainer. Lateral projections of the right ECA (**E**) and the left ECA (**F**) angiography demonstrated the disappearance of the TdAVF after balloon deflation. ECA: external carotid artery; MMA: middle meningeal artery; TdAVF: tentorial dural arteriovenous fistula

occlusion of the bilateral OAs, and removing the balloons, confirmatory imaging was performed. No clear arteriovenous shunt was noted by angiography of the bilateral external and internal carotid arteries and the right vertebral artery (VA), and the condition was regarded as neuroangiographic cure (**Fig. 4E** and **4F**). Moreover, no drainage of Onyx into the venous sinuses was noted. There were no clear changes in vital signs during the procedure. After surgery, no new neurological symptoms developed, no clear infarction was newly demonstrated by head MRI, and the patient followed an uneventful course and was discharged from our department on the 6th hospital day. At 3 months after treatment, no neurological symptoms had developed and no recurrence was noted by head MRI/MRA.

Discussion

The flow pattern of TdAVF varies among patients, reflecting the tentorial sinuses intrinsically receiving an influx of venous blood from both above and below. Regarding the venous sinuses and draining veins (infratentorial and supratentorial) associated with the shunt site (midline, paramedian, and lateral) from the viewpoint of surgical approach, Lawton et al. classified TdAVF into six types (type I: galenic, type II: straight sinus, type III: torcular, type IV: tentorial sinus, type V: superior petrosal sinus, and type VI: incisura).[2] The lesion reported here had multiple feeders, and as they all concentrated on the left side of the confluence, this site was considered the fistula point. However, as the drainage route was primarily infratentorial, the lesion was diagnosed as the straight sinus type according to the classification of Lawton et al. In addition, the lesion had multiple drainers, including the routes from the inferior vermian vein via the left lateral mesencephalic vein to the BVR and from a cortical vein on the cerebellar surface via the right petrosal vein to the right SPS. In addition, it drained via the two SVVs into the straight sinus, and this dispersion of venous flow is considered to partly explain the absence of symptoms. However, as asymptomatic TdAVF often exhibits aggressive behavior, we decided to proactively intervene therapeutically.

For curative treatment of TdAVF, which is often a non-sinus fistula, a combination of TAE, direct surgery, and radiation is performed rather than TVE. Direct surgery is unlikely to be selected as the first choice because of the high invasiveness, and radiation therapy requires 1–2 years until effects appear. Expectations for TAE as a curative treatment for TdAVF are increasing because of the improvements in diagnostic imaging, which have made understanding complicated vascular anatomy and hemodynamics possible, and the development of devices.[3] As for embolic agents, if NBCA is injected via the MMA with a slow flow, the treatment is likely to end in proximal ligation. Moreover, as skill is needed for concentration control, curative treatment by TAE alone is reported to be difficult.[4] Onyx, on the other hand, is a non-adhesive depositing-type liquid embolic agent that can be injected intermittently over a long period, and the embolized area is easy to control. Favorable results have been reported by TAE using Onyx for TdAVF.[5,6] For curative treatment by TAE, it is necessary to deliver Onyx to the affected vein, including the fistula point. For this purpose, the appropriate selection of the target vessel and injection of Onyx with the pressure gradient in mind are important. The MMA is often selected as the target vessel because it has no dangerous anastomoses in the convexity, runs linearly over a relatively long distance with only mild meandering, and is easy to approach within the vicinity of the fistula point.[7)] In the present case, based on intracranial angiographic findings, the bilateral OAs and bilateral MMAs were considered candidates for target vessels. The OA was thicker than the MMA and was a high-flow feeder, but we selected the MMA PCB as the target vessel because embolization via the OA would have involved risks such as "occipital skin necrosis," "incomplete embolization due to inability to navigate the microcatheter to the vicinity of the fistula point because of the marked tortuousness of the peripheral region," "difficulty in removing the catheter due to marked tortuousness," and "nerve damage due to unintended influx of the embolic agent via dangerous anastomoses." However, as the periphery of the MMA PCB is thin, it was considered difficult to navigate the catheter to the vicinity of the fistula point. Therefore, we used a DeFrictor (external tip diameter: 1.3 Fr), which has the smallest external tip diameter among the microcatheters compatible with DMSO. To navigate the DeFrictor as close as possible to the fistula point, a 0.010-inch guidewire was used in the proximal MMA and was replaced with a 0.008-inch guidewire when the DeFrictor reached the MMA PCB.

TAE using balloon catheters for arteriovenous malformation (AVM) or AVF is a common and well-reported procedure. Goto et al. proposed temporary occlusion of the proximal side of the main feeder and other feeders with balloon catheters as a method for nidus flow control in TAE using NBCA for AVM.[8)] Shi et al. reported that flow control by placing a balloon catheter on the proximal side during Onyx embolization of AVF facilitated plug formation and adjustment of the injection area.[9)] Kotsugi et al. reported a case of dAVF in the superior sagittal sinus involving multiple branches of the bilateral external carotid arteries in which complete embolization was achieved by injecting NBCA through a microcatheter guided to the vicinity of the shunt point via the MMA while occluding the origins of the bilateral external carotid arteries using 6Fr balloon guiding catheters.[10)] In our patient, as vessels other than those of the external carotid artery system, such as the SCA, PMA, and tentorial artery, were involved, and because the bilateral MMAs, which were the target feeders, were markedly thin, we performed TAE using highly penetrable Onyx in consideration of the possibility of NBCA embolization ending in proximal occlusion due to the inability to navigate the microcatheter to the vicinity of the shunt point. By selective balloon occlusion of the OAs without occluding the external carotid artery, angiography through the ipsilateral or contralateral guiding catheter was possible during Onyx embolization when appropriate, and the state of embolization and whether the venous sinuses were occluded was able to be confirmed as necessary. However, it is important to perform embolization in consideration of the possibility that the blood flow at the fistula point is not completely obstructed because occlusion with a balloon catheter is made at a relatively proximal point, resulting in persistence of the blood flow through feeders other than the OAs and OA-VA anastomosis.

In our patient, complete occlusion was achieved by TAE via the bilateral MMAs, but two sessions were necessary, possibly because the flow control was insufficient, as mentioned above. In addition, the MMA ran in a relatively straight course but was thin, and despite the selection of a low-profile microcatheter, it was unable to be navigated to the fistula point and Onyx embolization was performed from a relatively distal site. Furthermore, when Onyx embolization via the right MMA had progressed to an extent, left external carotid artery angiography was performed during balloon occlusion of the bilateral OAs, and the fistula and affected veins were able to be examined via the left MMA. We therefore changed the plan to embolization via the left MMA without adhering to embolization via the right MMA alone. This was considered to have necessitated the two sessions.

Complications of Onyx embolization include angialgia due to dimethyl sulfoxide (DMSO), which is the solvent of the ethylene vinyl alcohol (EVOH) copolymer, a long treatment time due to the necessity of gradual injection by the plug and push technique, and associated increase in exposure, risk of difficulty in catheter withdrawal associated with extension of the reflux area, and the unintended entry of Onyx, which is less adhesive than NBCA, into dangerous anastomoses, and migration to the venous side after its injection into a high-flow fistula.[11)] Moreover, if there are multiple feeders, the persistence of a high-flow feeder makes control of the flow of Onyx difficult, leading to the risk of its migration into unintended vessels, such as the normal venous sinuses, and difficulty of sufficiently delivering Onyx to the target site using the pressure gradient. In our patient, the control of Onyx was facilitated by temporarily blocking the blood flow of the bilateral OAs, which were well-developed feeders, using balloons, resulting in prevention of its diffusion to the normal veins and sufficient penetration of Onyx to the affected vein beyond the fistula point. Furthermore, as occlusion was temporary,

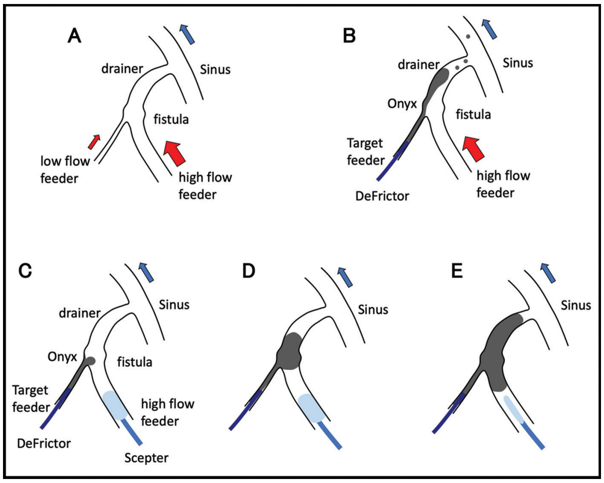

Fig. 5 The scheme for a dAVF with low- and high-flow feeders (**A**). Onyx embolization via a low-flow feeder without balloon occlusion (**B**). Onyx cast is carried away or fragmented by blood flow from the high-flow feeder. Onyx embolization via a low-flow feeder with balloon occlusion (**C**–**E**). Onyx penetrated the drainer through the fistula point under flow control by occluding the high-flow feeder using a balloon (**D**). Obliteration was achieved after balloon deflation (**E**). dAVF: dural arteriovenous fistula

the risk associated with OA embolization was considered to have been avoided, increasing the safety of the procedure (**Fig. 5**). However, it is still necessary to carefully follow-up the patient to evaluate the long-term effects of embolization.

Conclusion

As the use of Onyx for dAVF is now covered by health insurance, opportunities to perform Onyx embolization for TdAVF are expected to increase. To safely execute curative Onyx embolization, it is important to formulate a treatment strategy based on sufficient preoperative evaluation of the vascular anatomy and blood flow. In TdAVF with multiple feeders, if the target vessel is thin, effective Onyx embolization is made possible by temporary balloon occlusion of other high-flow main feeders.

This paper was written after receiving informed consent from the patient in person while observing the "Guidelines concerning the protection of patients' privacy in publication of medical papers, including case reports and presentations at scientific conferences."

Disclosure Statement

The first author and coauthors have no conflicts of interest.

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