Objective: We report a case of superior sagittal sinus (SSS) dural arteriovenous fistula (dAVF) treated with transarterial Onyx injection under flow control using two balloon guiding catheters and a dual-lumen balloon microcatheter.

Case Presentation: A 54-year-old man previously diagnosed with SSS dAVF with cortical venous reflux at 4 years prior, who did not request treatment as he was asymptomatic. During follow-up, he developed right temporal lobe subcortical hemorrhage and showed an occlusive change in venous drainage. We performed transarterial Onyx injection with assistance using multiple balloons placed in the bilateral external carotid and right middle meningeal arteries. Using this technique, we were able to sufficiently fill the proximal part of the venous drainage with Onyx and achieved complete obliteration. No treatment-related complication was observed, and follow-up angiography performed at 8 months after the treatment showed no recurrence.

Conclusion: Multiple balloon-assisted transarterial Onyx embolization is useful for adequate fillings of shunt and the proximal part of the venous drainage.

Keywords: dural arteriovenous fistula, superior sagittal sinus, transarterial embolization, balloon-assisted embolization, Onyx

Introduction

Dural arteriovenous fistula (dAVF) is a relatively rare disease that accounts for 10%–15% of intracranial vascular malformations, with an incidence of 0.16–0.51/100000 people.1,2) dAVF involving the superior sagittal sinus (SSS) reportedly accounts for 2.3%–12% of intracranial dAVFs.1,3) As some dAVFs of the SSS directly reflux into the cortical veins without draining into the normal sinus, while others are drained into the normal sinus, transvenous embolization (TVE) is often difficult. Thus, there are a number of reports on separation of the cortical veins by direct surgery.4) The main endovascular treatment for this lesion has primarily been transarterial embolization (TAE) using a liquid embolic material n-butyl-2-cyanoacrylate (NBCA) although there are recent reports of TAE using Onyx.5,6) To achieve complete cure by transarterial glue embolization, the glue should reach into the venous drainage penetrating the shunt. During glue injection, it is important to control the blood flow from feeders other than the vessel into which glue is injected. Furthermore, to avoid premature polymerization of the glue because of blood flow from other feeding arteries or its fragmentation into the venous drainage, controlled injection is important, and the use of balloon-assisted techniques for this purpose has been reported.5,7–9) Herein, we present a case of successful treatment of dAVF involving the SSS using Onyx embolization with flow control using two balloon guiding catheters and a dual-lumen balloon microcatheter.
Case Presentation

The patient was a 54-year-old man with a history of hypertension and cholecystectomy. In 1992, he noted a floating sensation and MRI of the head performed at a local hospital suggested vascular anomalies. However, he was not followed up or treated thereafter. In October 2012, he consulted a local physician primarily with a floating sensation and headache. Head MRI suggested vascular abnormalities again, and he was referred to our department and admitted for close examinations. DSA of the common carotid arteries revealed a shunting point adjacent to the SSS fed by the bilateral middle meningeal arteries (MMAs), and he was diagnosed with dA VF involving the SSS that directly refluxed into the cortical veins of the right cerebral hemisphere (Fig. 1A and 1B). In the venous phase, anterograde blood flow was observed in the SSS. As the lesion had been present for more than 10 years, and as it was asymptomatic, the patient did not wish further treatment and selected outpatient follow-up. However, his visits gradually became irregular thereafter, and the patient decided to discontinue oral medication for hypertension. On April 14, 2016, the patient visited the emergency department of our hospital because of intense headache, and was admitted immediately as abnormalities were noted on head CT.

Neurologic findings on admission: The patient showed normal consciousness, but evidence of hemiparesis of the left upper and lower extremities with a manual muscle testing (MMT) of 4/5.

Neuroradiologic findings: CT of the head showed a subcortical hemorrhage in the right temporal lobe (Fig. 1C). Similar to previous imaging findings, DSA performed to identify the source of bleeding confirmed a dAVF, in contact with the SSS, fed by the bilateral MMAs, and accompanied by cortical venous reflux into the right cerebral hemisphere. Compared with previous angiograms (Fig. 1A), one of the cortical venous drainages was obliterated, and a varix in contact with the hematoma was enlarged (Fig. 2C).

Clinical course after admission: The subcortical hemorrhage was treated conservatively in consideration of the hematoma volume. Rehabilitation was initiated early, and as paralysis improved rapidly, the patient was discharged home on the 7th hospital day with unassisted ambulation. Angiographic findings suggested that one of the cortical venous refluxes was thrombosed, and that subcortical hemorrhage was induced by an increased venous pressure. As the patient became symptomatic, we explained the necessity for aggressive treatment of the dAVF. As the dAVF was unrelated to the sinus, it was considered difficult to treat by TVE, and TAE using a liquid embolic material was planned. We selected Onyx as the embolic material, as it could be injected in a sufficient volume over a relatively long period.

Endovascular treatment: As the dAVF was fed by the bilateral MMAs and showed a relatively rapid flow, proximal flow control was implemented by placing balloon guiding catheters in the bilateral external carotid arteries. Furthermore, we decided to place a Scepter C 4 mm × 10 mm microcatheter (MicroVention-Terumo, Tustin, CA, USA) into the posterior branch of the right MMA, through which the embolic material was injected to promote early plug formation. After the induction of general anesthesia, two 7 French (Fr) 80 cm Shuttle sheaths (Cook Medical,
Bloomington, IN, USA) were placed in the bilateral common carotid arteries via the bilateral femoral arteries. After administration of 750 mg fosphenytoin and 500 mg prednisolone, heparin was administered intravenously at 4000 units, followed continuous infusion at 1000 units/hour. Two 7 Fr Optimo guiding catheters (Tokai Medical Products, Aichi, Japan) were placed in the bilateral common carotid arteries. Right common carotid artery angiography (Fig. 2A–2C) and left external carotid artery angiography (Fig. 2D–2F) were then performed, and the dAVF was confirmed to have bilateral MMAs as feeding arteries and the cortical venous reflux as suggested by the previous imaging studies. Simultaneous injection of contrast media was also performed through two 7 Fr Optimo catheters advanced to points immediately after the bifurcations of the bilateral external carotid arteries and the occipital arteries (OAs). A Waters’ view in which the bilateral MMAs and the shunt point were visualized was selected as the working angle (Fig. 3A), and a Scepter was placed in the right MMA posterior branch near the shunt using an Asahi Chikai 14 (Asahi Intec, Aichi, Japan) (Fig. 3A, arrow). The position of the catheter tip was checked by microinjection from the Scepter (Fig. 3B), and DSA was performed under inflation of the Scepter balloon (Fig. 3C). The contrast medium stagnated distally to the catheter because of the Scepter balloon, and the blood flow control of the vessel through which Onyx would be injected was confirmed as adequate. However, laminar flows of blood entering from other MMA branches were clearly observed in the venous drainage. When the two balloons of the Optimo (Fig. 3D, arrowhead) were inflated in addition to the Scepter balloon (Fig. 3D, arrow), the contrast medium stagnated in the venous drainage under fluoroscopy, indicating adequate control of blood flow into the shunt. Onyx was then injected with all balloons of the two Optimo guiding catheters, and the Scepter microcatheter was inflated. An amount of Onyx 34 (0.4 mL) nearly equal to the dead space of the Scepter (0.44 mL) was injected, followed by injection of Onyx 18 by the plug and push technique. While Onyx showed slight reflux in the direction of the balloons, it immediately began to advance anterogradely, and the shunt and the proximal part of the venous drainage were completely occluded (Fig. 3E) by injecting a volume sufficient to permeate these areas (2.26 mL). The catheters were removed after deflating the Scepter balloons without being trapped. After the two balloons of the Optimo guiding catheters were deflated, bilateral common carotid artery angiography (Fig. 4) was performed, and occlusion of the shunt with a patent SSS was confirmed and the procedure was ended.

Postoperative course: No new neurologic deficits appeared after the procedure. The patient was discharged to home at

Fig. 2 Serial right common carotid angiograms (anterior–posterior view [A] lateral view of arterial phase [B] and late arterial phase [C]) and left external carotid angiograms (anterior–posterior view of early arterial phase [D] and late arterial phase [E] lateral view [F]) show a dural arteriovenous fistula adjacent to the superior sagittal sinus associated with the cortical venous reflux.
Fig. 3  A caudal view of DSA (A) obtained by simultaneous bilateral external carotid artery injections shows the dural arteriovenous fistula directly draining into the cortical vein. Note the Scepter microcatheter (arrows) advanced into the right middle meningeal artery. A selective injection from Scepter microcatheter (B) shows the fistulous point and draining vein. Another injection (C) after inflation of Scepter balloon shows apparent stasis of contrast media around the tip of the catheter; however, residual flow from other meningeal feeders results in rapid washout of the contrast media in the draining vein. A non-subtracted image (D) obtained just after starting the injection of Onyx. Note inflated Scepter balloon (arrow) in the middle meningeal artery and bilateral Optimo balloons (arrow heads) placed at the external carotid arteries. A non-subtracted image (E) obtained at the end of the Onyx injection shows minimal reflux toward the tip of the microcatheter.

Fig. 4  Lateral view of right common carotid angiograms (arterial phase [A] venous phase [B]) shows complete obliteration of the dural arteriovenous fistula with the patent superior sagittal sinus. An anterior view of left common angiogram (C) also shows no residual filling around the superior sagittal sinus.
leads to the recurrence of dA VF.10) On the other hand, as complete occlusion of the proximal side of venous drainage was difficult because of its adhesiveness to the catheter; incompletely occluded, and 41% with a hemorrhagic presentation.3) Therefore, dAVF of the SSS is considered to require aggressive treatment including craniotomy, endovascular treatment, or radiation therapy alone or in combination.3,4) Direct surgery of dAVFs involving separation and interruption of the venous drainage is also widely used, and a high obliteration rate has been reported.5) Endovascular treatment for non-sinus type dAVFs is primarily treated by TAE using a liquid embolic material, and there have recently been some reports of TAE using Onyx.5,8,9) In the above-mentioned series of Gross et al.,10 the complete obliteration rate of dAVF of the SSS by TAE improved from 60% to 82% after the introduction of Onyx, while the percentage of patients who required additional direct surgery decreased from 14% to 8%.

NBCA is often used as a liquid embolic material for TAE although it is not suitable for injection over a long time because of its adhesiveness to the catheter; incomplete occlusion of the proximal side of venous drainage leads to the recurrence of dAVF.10) On the other hand, as Onyx does not adhere to the catheter, it can be slowly injected until sufficient occlusion of the venous drainage is achieved. Indeed, there are increasing reports of its therapeutic use of Onyx because of this benefit.5) When performing curative TAE using a liquid embolic material, adequate flow control from other feeders and vessels joining near the shunt is important for prevention of premature polymerization of the embolic material before it sufficiently fills the venous drainage. For this purpose, dual-lumen balloon microcatheter was reported to be useful in Onyx embolization for dAVF.5,8,9) The advantages of this catheter in Onyx embolization include the short time required for plug formation, leading to reduced fluoroscopic times, and the simplicity of the procedure, as a second catheter for flow control of the feeder vessels is not required.5,7–9) It is important to note that a dual-lumen balloon microcatheter has a larger outer diameter and is more rigid than the Marathon catheter, which is often used in Onyx embolization. In our patient, we used a Scepter C (2.1 Fr) with a crossing profile even smaller than that of the Scepter XC (2.6 Fr) although it was clearly more difficult to navigate to distal areas to approach the shunting point compared with the Marathon catheter (outer diameter of the tip: 1.5 Fr). Unlike intracranial vessels, the tortuous MMA is difficult to straighten with a microguidewire, and complications have been reported, including dissection and rupture of the vessel.5,7–9) Furthermore, if a Scepter catheter is not guided to a distal position, and Onyx embolization is performed from proximally, then a relatively long distance of the proximal side is embolized compared with embolization using a Marathon catheter placed near the shunt, which may cause unintended migration of embolic material via dangerous anastomoses. Since our patient had MMAs of a sufficient diameter, we could advance the Scepter relatively easily to a point immediately prior the shunt. Nevertheless, forcing a Scepter through a smaller vessel should be avoided.

While a higher occlusion rate was reported with TAE using Onyx compared with TAE using NBCA, there are some potential recurrence issues at long-term follow-up. Ambekar et al.11) reported that 14.3% of the patients exhibited angiographic recurrences at a mean follow-up period of 14 months after receiving prior curative TAE using Onyx and were judged angiographically to have been “cured”; recurrences were also frequently observed within 1 year after treatment. Insufficient penetration of the embolic material to the shunt and venous drainage, and failure to achieve complete occlusion of the venous drainage, were suggested as factors responsible for this recurrence. We considered that the use of balloon guiding catheters in the combination with previously reported procedure of TAE using a dual-lumen balloon microcatheter has the following advantages. A balloon guiding catheter (Optimo) placed in the external carotid artery controls the blood flow of the bilateral MMAs. Then, when Onyx begins to advance from the shunt into the venous drainage, this prevents fragmentation of Onyx into the venous drainage, allowing Onyx to fill layer by layer in the venous drainage. As shown in Fig. 3C, when Onyx began to advance into the venous drainage, the control of unintended fragmentation with a Scepter balloon alone was considered difficult because of the blood flow from other feeding arteries. Indeed, Onyx can be injected by the...
standard method, that is, repeatedly injecting it with short pauses when it enters the venous drainage without the flow control of the external carotid artery although more consistent injection was achieved using our method. In the second half of the TAE procedure with balloon guiding catheters, blood flow from the bilateral MMAs was controlled by the Optimo balloons placed in the external carotid artery, allowing Onyx to be more readily refluxed into their feeding arteries. Although Onyx is considered to reflux easily into feeding arteries, however, when considering the high recurrence rate reported by Ambekar et al., re-bleeding was observed in 3/32 patients with a history of bleeding, and bleeding was observed in 1/53 patients without history of bleeding. Based on these reports and others, the standard guidelines published by Lee et al. in 2015 also reported that the annual bleeding rate of asymptomatic lesions with cortical venous reflux (1.4%–1.5%) was lower than that after hemorrhagic presentation (7.4%–7.6%), supporting the difficulty of predicting the natural history of dAVF based on the finding of cortical venous reflux alone. The obstructing change in the venous drainage observed in our patient is considered a risk factor despite the absence of symptoms. With more detailed evaluation in the future, this case is expected to promote further understanding of the natural history of dAVF.

### Conclusion

We reported a case of dAVF of the SSS with a hemorrhagic presentation successfully treated by transarterial Onyx embolization with blood flow control using multiple balloon assist. Multiple balloon-assisted Onyx injection is useful technique because the injected Onyx can be accurately controlled by flow control from an early stage in TAE.

### Disclosure Statement

Neither the first author nor any of the coauthors have any conflicts of interest regarding this paper.

### References


