Two Cases of Arteriovenous Fistula of the Scalp in Which the Pressure Cooker Technique Was Useful

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Objective: We could cure two cases of arteriovenous fistula (AVF) of the scalp by the pressure cooker technique (PCT).

Case Presentations: Case 1 showed scalp AVF with a direct arteriovenous (AV) shunt between the left posterior auricular artery (PAA) and the left superficial temporal vein (STV), which was also fed by a branch of the left occipital artery (OA). Case 2 showed scalp AVF with a direct AV shunt between the left occipital vein (OV) connected to the left STV and the left OA and left PAA as the collateral feeders. The shunts could be completely occluded by forming a plug using coils and low-concentration n-butyl-2-cyanoacrylat (NBCA) and injecting Onyx with pressure through a non-detachable microcatheter (MC) by the PCT via the left PAA in Case 1 and via the left OA in Case 2. The MC could be removed in both patients.

Conclusion: This technique may be useful if it is applied only to the external carotid artery system, in which adhesion of the MC is manageable, and Onyx injection finishes within a several minutes.

Keywords ▶ scalp arteriovenous fistula, pressure cooker technique Onyx, n-butyl-2-cyanoacrylat

Introduction

Scalp arteriovenous fistula (AVF) is a relatively rare disorder that has an extensive and complicated network in the external carotid artery system and exhibits clinical symptoms such as tinnitus, local pain, headache, hemorrhage, and scalp necrosis. Various treatments have been reported, and craniotomy, endovascular treatment, or their combination is often performed, but skin grafting is necessary after resection if the lesion involves a wide area. The pressure cooker technique (PCT), reported by Chapot et al., is a technique to enhance the embolization rate of the lesion by forming a plug to prevent reflux using coils and a small amount of n-butyl-2-cyanoacrylat (NBCA) through a microcatheter (MC) inserted separately on the proximal side of a detachable MC for injection of Onyx under pressure cooker through the distal detachable MC. In the two cases of scalp AVF reported here, which presented with tinnitus, we could completely embolize the shunt area by the PCT using a non-detachable MC for pressure injection of Onyx.

Case Presentations

Case 1
The patient was a 75-year-old male with histories of cataract and sinusitis but no trauma. Tinnitus of the left ear appeared 3 years before, and it was gradually exacerbated from 1 year before. Sleep disorders occurred 1 month before, and oral bortizolam was prescribed. He underwent imaging examinations at a local hospital and was referred to our department.

On admission, no neurologic abnormality was noted. Pulsatile vascular murmur was heard posteriorly to the left auricle.

From findings on cerebral angiography, scalp AVF with a direct arteriovenous (AV) shunt between the left posterior auricular artery (PAA) and left superficial temporal vein (STV) and influx from a branch the left occipital artery (OA) as a feeder was diagnosed (Fig. 1).
Endovascular treatment was initiated under general anesthesia. After the placement of an 8 Fr sheath in the right femoral artery and a 6 Fr sheath in the left femoral artery, systemic heparinization was started by intravenous injection of 5000 units. An 8 Fr FUBUKI (Asahi Intec Co., Ltd, Aichi, Japan) was placed at the origin of the left external carotid artery as a guiding catheter, and a 4.2 Fr FUBUKI was placed at the origin of the left PAA as an intermediate catheter. On the venous side, a 6 Fr Optimo (Tokai Medical Products, Aichi, Japan) was placed at the origin of the left STV as a guiding catheter, and a 3.6 Fr JB2 catheter (Gadeilius Medical K.K., Tokyo, Japan) was placed near the shunt point as an intermediate catheter.

First, a Marathon (eV3 Covidien, Irvine, CA, USA) for Onyx injection was inserted to a site closest to the shunt point through the left PAA. Next, an Excelsior SL-10 (Stryker, Kalamazoo, MI, USA) was inserted through the parent catheter 8 Fr FUBUKI for coil embolization and placed about 1 cm before the Marathon. An Axium 7 × 30 cm (eV3 Covidien) was placed on the venous side immediately after the shunt to prevent draining of Onyx into the lung through the left STV using an Excelsior SL-10, and coil embolization was performed using Axium 3 mm × 10 cm and ED coils 2 mm × 6 cm (Kaneka Medix Corp., Osaka, Japan) through the Excelsior SL-10 on the arterial side (Fig. 2A). Then, a plug was formed by injecting 0.05 mL of 25% NBCA through the Excelsior SL-10, at the proximal portion of the Marathon (Fig. 2B). The Excelsior SL-10 on the arterial side was removed, and 1.5 mL of Onyx34 (Micro Therapeutics, Inc., Irvine, CA, USA) was injected through the remaining Marathon. The venous side was occluded from the left PAA across the shunt, then, Onyx refluxed from the shunt area to the feeding arteries of the left OA and left PAA (Fig. 2C–2D), and the AV shunt was completely occluded (Fig. 3). The injection took about 5 minutes, but the Marathon could be removed without resistance. The procedure was finished after confirming complete obliteration of the shunt point by bilateral common carotid artery and vertebral artery angiography. The disappearance of tinnitus was confirmed shortly after the procedure. No reddening, necrosis, or hair loss was noted in the scalp.
Case 2
The patient was a 63-year-old female with no particular clinical history including trauma. She had the onset with left-sided pulsatile tinnitus.

On admission, no neurologic abnormality was noted, but pulsatile vascular murmur was heard in the left occipital region.

By cerebral angiography, a diagnosis of scalp AVF with a direct AV shunt between the left occipital vein (OV) and left STV with the left OA and left PAA as feeders was made (Fig. 4A–4B).

Endovascular treatment was initiated under general anesthesia. After an 8 Fr sheath was placed in the right femoral artery, systemic heparinization was made by intravenous administration of 5000 units. An 8 Fr FUBUKI was placed at the origin of the left external carotid artery as a guiding catheter, and a 4.2 Fr FUBUKI was placed in the left OA as an intermediate catheter. We attempted to

Fig. 2  Selective angiography of the left PAA in Case 1. (A) Lateral view. The black arrow shows the tip of Marathon navigated in the posterior auricular artery as close to the shunt point. Black arrow heads show the 7 mm × 30 cm Axium coil placed in the draining vein adjacent to the shunt to prevent from the migration of the Onyx into the pulmonary artery. The white arrow head shows the coils (Axium 3 mm × 10 cm and ED coil 2 mm × 6 cm) placed at the just proximal portion of the tip of the Marathon to make the plug by injecting 25% NBCA from the SL-10 located at the proximal portion of the coils. (B) Lateral view after injection of 25% NBCA from SL-10. The white arrow shows the injected NBCA. (C) Lateral view. Injection of Onyx34 from Marathon. Onyx passed through the shunt point and entered into the distal part of PAA (black arrow). (D) Lateral view at the late phase of Onyx injection. Onyx entered into the OA through the shunt point and occluded the distal part of the OA contributing the shunt (black arrow). Axium: eV3 Covidien, Irvine, CA, USA; Marathon: eV3 Covidien; NBCA: n-butyl-2-cianoarylate; OA: occipital artery; PAA: posterior auricular artery; SL-10: Stryker, Kalamazoo, MI, USA
approach the shunt point using a Marathon via the mastoid branch of the left OA, but we could not navigate it through a small curve before the shunt point. A small dural branch running medioinferiorly on its peripheral side was noted, and as there was a possibility of its anastomosis with a vessel supplying the facial nerve, a Marathon was inserted into this branch, and it was occluded using an ED coil Extrasoft 1.5 mm × 1 cm. Next, through the 8 Fr FUBUKI, guiding catheter, an Excelsior SL-10 Pre-Shaped J was inserted for coil embolization and placed about 1 cm before the Marathon. Coil embolization was performed from this site by inserting GDC 10 soft 360 3 mm × 6 cm (Stryker) and ED Extrasoft 2 mm × 6 cm (Kaneka Medix Corp., Osaka, Japan) (Fig. 4C), a plug was formed by injecting 0.02 mL of 25% NBCA through the Excelsior SL-10 Pre-Shaped J. (Fig. 5). The Excelsior SL-10 Pre-Shaped J was removed, 0.4 mL of Onyx18 was injected over 2–3 minutes through the remaining Marathon (Fig. 4D), Onyx was made to flow back to the peripheral side of the left OA and into the left PAA, and the AV shunt was completely occluded (Fig. 4E). The Marathon could be withdrawn without resistance. The procedure was ended after confirming the obliteration of the shunt point by angiography of the entire external carotid artery system (Fig. 4F). The disappearance of tinnitus was confirmed shortly after the procedure. No reddening, necrosis, or hair loss was noted in the scalp.

Discussion

Scalp AVF was first reported in 1757 by Hunter, and its treatment by circumferential ligation was reported in 1829 by Brodie. The disorder has been called by various terms
including cirrhotic aneurysms, arteriovenous (AV) aneurysms, aneurysm by anastomosis, aneurysmal varix, plexiform angioma, and scalp AV malformations. Grimes defined lesions with a small number of simple communicating branches between the artery and vein as AVFs, varix-like lesions with a large number of complex communicating branches as cirrhotic aneurysms, and the two types combined as scalp AV malformations (scalp AVMs).

Etiologically, the lesions are classified into congenital and acquired, and acquired lesions are often caused by blunt trauma but have also been reported in connection with scalp grafting, craniotomy, and arthroscopic examination of the temporomandibular joint. Treatments are indicated for aesthetic improvement of pulsatile masses, prevention of bleeding, and management of exacerbation of symptoms such as headache and tinnitus. No clear history of trauma or surgery was reported by the two cases presented here, but

Fig. 4  Lateral view of the left external carotid angiogram in Case 2. (A) Preoperative external carotid angiogram, early artery phase. The arrow shows the shunt point. (B) Preoperative external carotid angiogram, capillary phase. The arrow shows the shunt point. (C) Selective angiogram from the SL-10 placed 1 cm proximal of the tip of the Marathon in the occipital artery. Arrow shows coils (GDC 10 soft 360 3 mm × 6 cm, ED Extrasoft 2 mm × 6 cm) placed at just proximal of the Marathon. (D) Injection of Onyx from the Marathon. Arrow shows the coil introduced into the meningeal branch of the mastoid branch of the occipital artery to prevent Onyx migration into the branches supplying cranial nerves. Arrow heads show 25% NBCA injected from the SL-10 to make the plug. Onyx penetrated through the shunt and occluded the distal branch of the OA and venous side. (E and F) Postoperative external and common carotid angiogram. The arteriovenous shunt was completely obliterated. ED Extrasoft: Kaneka Medix Corp., Osaka, Japan; GDC: Stryker, Kalamazoo, MI, USA; Marathon: eV3 Covidien, Irvine, CA, USA; NBCA: n-butyl-2-cyanomethlyate; OA: occipital artery; SL-10: Stryker

Fig. 5  Schematic diagram of the scalp AVF in Case 2. AVF: arteriovenous fistula; Marathon: eV3 Covidien, Irvine, CA, USA; NBCA: n-butyl-2-cyanomethlyate; OA: occipital artery; OV: occipital vein; PAA: posterior auricular artery; STV: superficial temporal vein
the possibility that the lesions were caused by minor trauma cannot be excluded. Radical surgical resection used to be the most common treatment, but it was difficult to treat all feeders, and there was a strong tendency to recur. In addition, complications such as scalp necrosis and postoperative bleeding due to incomplete treatment were possible. According to a review of the literature over 20 years until 2014 by Xue et al., endovascular treatment was performed in 29 patients, resulting in recurrence in 3 (10.3%) and complications such as tenderness of the site of operation, reddening, skin necrosis, migration of the embolic agent into the vein, and loss of hair in 4 (13.8%), and surgical treatment was performed in 65 patients, resulting in recurrence in none but complications such as intraoperative massive hemorrhage and skin necrosis in 2 (3.1%). They also reported that the five patients treated by a combination of endovascular treatment + surgical treatment were free of recurrence or complications.\(^{11}\) Various endovascular procedures have been reported, embolic agents such as Onyx, NBCA, coils, polyvinyl acetate, and ethanol have been used, and techniques including direct puncture using a circular compression device (CCD)\(^{12}\) and transvenous embolization by Onyx injection using a balloon\(^{13}\) have been reported.

In both of our patients, the vasculature of the lesion was simple with a small number of feeders, a single shunt point, and 2–3 draining veins. We selected endovascular treatment because the possibility that the shunt area would be completely occluded from the arterial side was high. We selected Onyx as the embolic agent due to its reliability because it was necessary to completely occlude the shunt area and part of venous drainers with a liquid embolic agent from a single feeder for radical cure. The treatment was performed with careful informed consent by providing sufficient information including dark discoloration of the skin at the site of Onyx injection, necessity of resection if a bulge should form at the injection site, and off-label use of Onyx. Methods for Onyx embolization include the simple push technique, plug and push technique, balloon assist technique, and PCT, but we selected the PCT because a balloon catheter could not be inserted due to marked tortuosity, and because it was necessary to ensure delivery of Onyx from the shunt to veins with strong pressure. The catheters compatible to dimethyl sulfoxide (DMSO) available in Japan for Onyx injection were the Marathon, Echelon 10 (eV3 Covidien), Echelon 14, Headway 17A Advanced (Terumo Corporation, Tokyo, Japan), Rebar18 (eV3 Covidien), and Scepter C/XC (Terumo Corporation), and detachable catheters such as Sonic (Balt, Montmorency, France) and Apollo (eV3 Covidien) are widely used abroad. We selected the Marathon as it was available in Japan, deliverable to peripheral regions, and highly manipulable. Also, while the Marathon was not detachable, we performed the PCT because, in our experience, the catheter could be extracted after its placement in the blood vessel of the external carotid artery system for a few minutes even when a small amount of NBCA at a concentration of 20%–25% was allowed to flow back to the proximal side of the catheter. In addition, we used this method because the catheter can be cut easily in the OA by making a small incision should its removal become difficult.

The procedure was performed under general anesthesia because of vascular pain in Onyx injection. The tip of the Marathon was placed as close as possible to the shunt point, and the Excelsior SL-10 for coil embolization was placed about 10 mm from the tip of the Marathon. A coil was placed through the Excelsior SL-10 for plug formation, and a plug was formed by injecting NBCA 25% at the coil through the same Excelsior SL-10. The point of plug formation is, following the original method of Chapot, to place the MC for NBCA injection as close as possible to the coil rather than in the coil mass and to stop the injection when NBCA begins to slightly flow back toward the feeder after filling the coil mass. Thereafter, Onyx was injected continuously through the Marathon (Fig. 3). Advantages of the PCT over the plug and push technique are that a safety margin for reflux need not be considered and that a large amount of Onyx can be injected in a short period with pressure. However, as pressure is used for injection, Onyx also retrogradely enters feeders that are not delineated on angiography, and it is necessary to collect information about feeders before the procedure. In Case 1, a coil was also placed on the venous side to prevent Onyx that migrated into the vein from flowing into the pulmonary vein. In Case 2, additional coil embolization was also performed in a branch distal to the plug to prevent Onyx from entering the vessel supplying the facial nerve.

Disadvantages of the PCT include complexity of the procedure, which requires insertion of two MCs and the use of coils, NBCA, and Onyx, the possibility of reflux if plug formation is inadequate, and dilatation of the blood vessel if excessive pressure is applied for Onyx injection, permitting reflux of Onyx through the gap between the plug and vessel wall. For this reason, accurate measurement of the diameter of the target vessel and appropriate selection of the coil are necessary. In addition, extraction of the Marathon is occasionally prevented by the NBCA or
Onyx embolus. It can be removed in most cases by gently and slowly pulling it. However, according to the package insert, the risk of catheter separation increases if the Marathon, which is flexible over 25 cm from its end, is likely to be broken at the junction part, where the polyamide used in the shaft changes to polyether block amide, made more flexible by the addition of polyether. However, if, in the worst case, the device breaks, the broken segment can be left in the external carotid artery or is considered to be recovered by making an incision under local anesthesia. Presently, at our hospital, we perform the PCT only in the external carotid artery system in consideration of the possibility of rupture of the Marathon. Also, according to the report by Chapot et al., the proximal side of the detachable MC was embolized with 33%–50% NBCA, but, using a non-detachable catheter, we embolized the proximal part of the catheter from the beginning with 25% NBCA and have experienced no case in which catheter extraction was difficult after Onyx injection over a few minutes. The PCT is expected to become more widely applicable as a very useful procedure by the introduction of detachable MCs such as Sonic and Apollo.

## Conclusion

We treated two cases of scalp AVF by the PCT using a non-detachable MC and successfully occluded the lesions. This technique could be performed safely by forming a small plug at a site as close as possible to the catheter using a coil and low-concentration NBCA and injecting Onyx within a few minutes by limiting indications to lesions in the external carotid artery system. It is expected to become a useful technique with the introduction of detachable MCs.

## Disclosure Statement

There are no conflicts of interest to be disclosed concerning this paper.

## References