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Case Report

Multiple Spontaneous Arteriovenous Fistulas of the Superficial Temporal Artery

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Objective: Multiple spontaneous arteriovenous fistulas (AVFs) of the external carotid artery (ECA) are rare. We present a case of multiple spontaneous AVFs treated by coil embolization using a combination of the transarterial and transvenous approach.

Case Presentation: A 59-year-old woman complained of right pulsatile tinnitus and a mass lesion at the ventral region of the right ear. 3D CTA and cerebral angiography revealed two AVFs on the superficial temporal artery (STA) with an aneurysm 9.8 mm in diameter. A balloon guiding catheter was navigated to the right STA via the right femoral artery. Another balloon guiding catheter was navigated to the right external jugular vein. The STA distal to the aneurysm was embolized with platinum coils by a transvenous approach. The STA proximal to the aneurysm was embolized transarterially and draining veins were embolized transvenously. Her symptoms were cured after endovascular embolization. MRA at 1 day and 6 months postoperatively showed no recurrence of AVFs or aneurysm.

Conclusion: Coil embolization of multiple spontaneous AVFs of the ECA using a combined transarterial and transvenous approach is a curable treatment option. Transvenous embolization of an STA distal to an aneurysm is useful.

Keywords coil embolization, multiple arteriovenous fistulas, superficial temporal artery

Introduction

Spontaneous arteriovenous fistulas (AVFs) of the external carotid artery (ECA) are rare, with more than 95% of such cases caused by traumatic or iatrogenic accident.[1–5] Multiple spontaneous cases are particularly rare. To the best of our knowledge, only four cases of multiple spontaneous AVFs of the ECA have been reported.[6,7] Two cases were treated by Onyx (Medtronic, Minneapolis, MN, USA) embolization, while the other two cases were treated by surgical resection.[6,7] We present a case of multiple spontaneous AVFs of the superficial temporal artery (STA) treated by coil embolization using a combined transarterial and transvenous approach.

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Case Presentation

A 59-year-old woman complained of right pulsatile tinnitus that originated 2 years ago and a pulsatile mass at the ventral region of the right ear 1 year ago. No other neurological sign was detected. She had no history of craniofacial injury, hypertension, or connective tissue disease. CTA revealed a right temporal AVF combined with STA aneurysm 9.8 mm in diameter (**Fig. 1A**). Right external carotid angiography revealed two AVFs of the STA (**Fig. 1B** and **1C**). The proximal AVF combined with the STA aneurysm drained into the superficial temporal vein (STV). The distal AVF situated at the suboccipital region drained into the muscular vein.

The patient selected endovascular embolization of the lesion after written informed consent. We planned coil embolization of lesions by combining transarterial and transvenous approaches (**Fig. 1D**). From the right femoral artery, a 6-Fr Optimo EPD balloon guiding catheter (Tokai Medical Products, Aichi, Japan) was navigated to the orifice of right STA (**Fig. 2A** and **2B**). From the left femoral vein, a 7-Fr Optimo EPD balloon guiding catheter was navigated to the right external jugular vein. Using a CHIKAI black 14 (Asahi Intecc, Aichi, Japan), an Excelsior SL-10

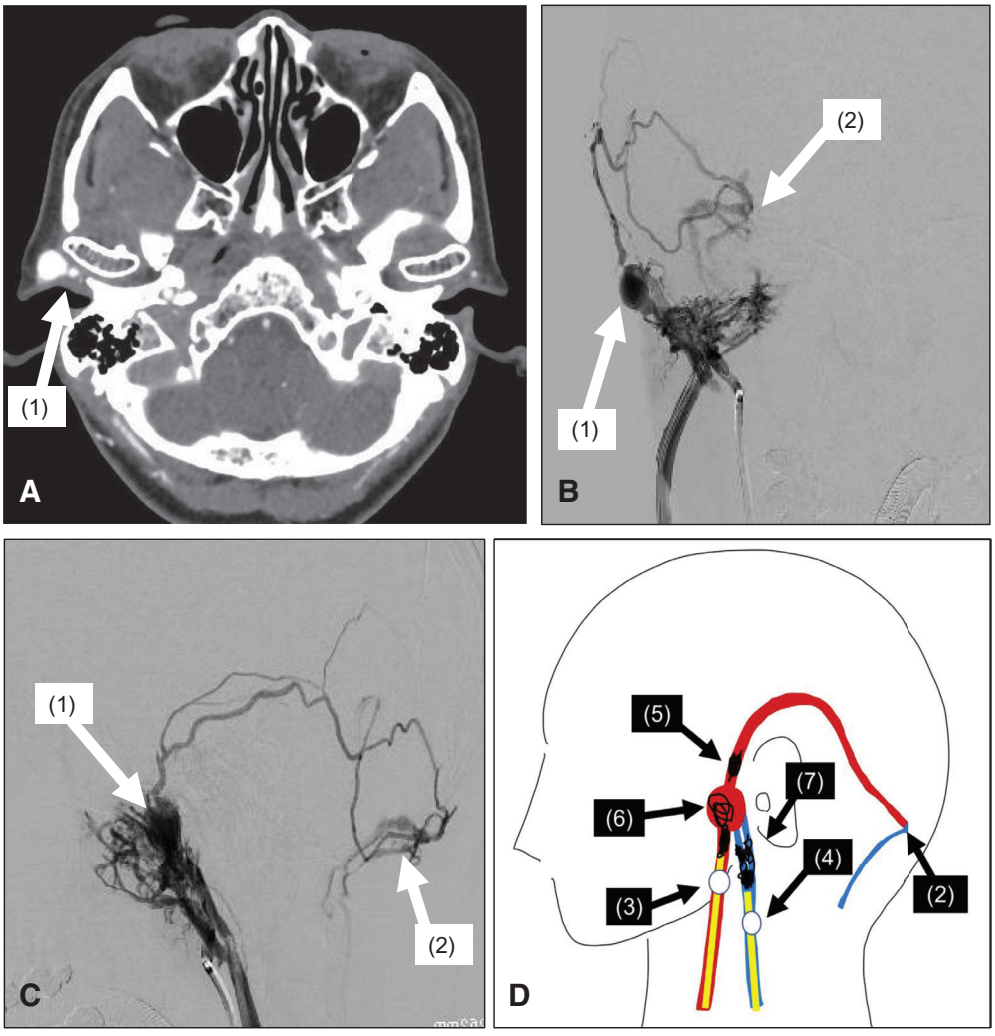

Fig. 1 Contrast CT, preoperative digital subtraction angiography, and operative strategy. (**A**) Contrast-enhanced CT. (**B** and **C**) Anterior–posterior view and lateral view of preoperative digital subtraction angiography. (**D**) Operative strategy. (1) The STA aneurysm and AVF. This aneurysm is situated in the ventral region of right external ear canal. (2) AVF situated in suboccipital region. (3) 6-Fr Optimo EPD balloon guiding catheter is navigated to the right STA. (4) 7-Fr Optimo EPD balloon guiding catheter in the right external jugular vein. (5) Using a transvenous-transaneurysmal approach, the distal STA is occluded by platinum coils under blood flow reduction by arterial balloon inflation. (6) Three loops are inserted into the aneurysmal dome and then the remainder is inserted into the STA proximal to the aneurysm. (7) Two draining veins are occluded transvenously. AVF: arteriovenous fistula; STA: superficial temporal artery

pre-shaped 45° (Stryker, Kalamazoo, MI, USA) was guided to the STA distal to the aneurysm by a transvenous and transaneurysm approach, and this distal STA was occluded by platinum coils under blood flow reduction by arterial balloon inflation (**Fig. 2A** and **2B**). Another Excelsior SL-10 pre-shaped 45° was guided into the aneurysmal dome transarterially. Three loops of coil were inserted into the aneurysmal dome, and the remainder was then inserted into the STA proximal to the aneurysm (**Fig. 2C** and **2D**). The Excelsior SL-10 pre-shaped 45° and the Headway 17 45° (Terumo, Tokyo, Japan) were guided into the two draining veins transvenously. Finally, two draining veins were occluded with coils (**Fig. 2E** and **2F**). After embolization, angiography showed complete obliteration of the STA aneurysm and cure of two fistulas (**Fig. 3A** and **3B**).

The pulsatile tinnitus and a pulsatile mass at the ventral region of the right ear disappeared, and the coil mass was not palpable at the body surface after the endovascular embolization. MRA 1 day after endovascular embolization showed no right AVF (**Fig. 3C**). She was discharged on the fourth postoperative day without any complication. An MRA taken at 6 months after endovascular embolization showed no recurrence of AVF (**Fig. 3D**).

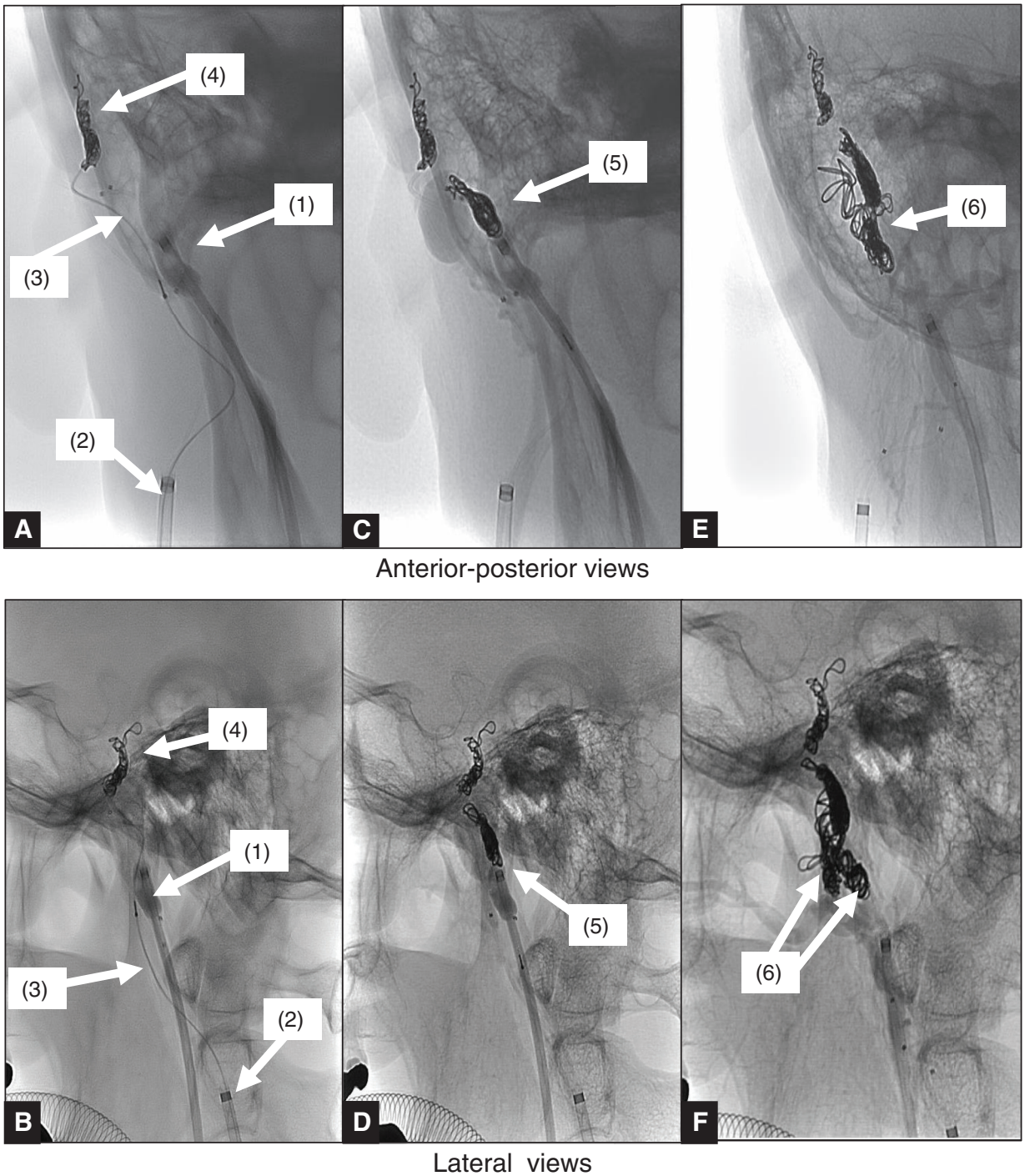

Fig. 2 Intraoperative findings. (**A** and **B**) Coil embolization of the STA distal to the aneurysm. (**C** and **D**) Coil embolization of the STA proximal to the aneurysm. (**E** and **F**) Coil embolization of two draining veins. (1) 6-Fr Optimo EPD balloon guiding catheter in the right STA, the balloon of which is inflated for blood flow reduction. (2) 7-Fr Optimo EPD balloon guiding catheter in the right external jugular vein. (3) Excelsior SL-10 pre-shaped 45° in the STA distal to the aneurysm by a transvenous-transaneurysmal approach. (4) Coil mass in the distal STA. (5) Coil mass in the aneurysmal dome and proximal STA. (6) Coil mass in the two draining veins. STA: superficial temporal artery

Discussion

This is the first report on multiple spontaneous AVFs of the STA. Definitive mechanisms for spontaneous AVFs have not been revealed.[1,2,4,8] Several reports have suggested that spontaneous AVFs are related to congenital factors and to acquired factors such as hypertension and arteriosclerosis. Pathological studies of spontaneous AVFs revealed

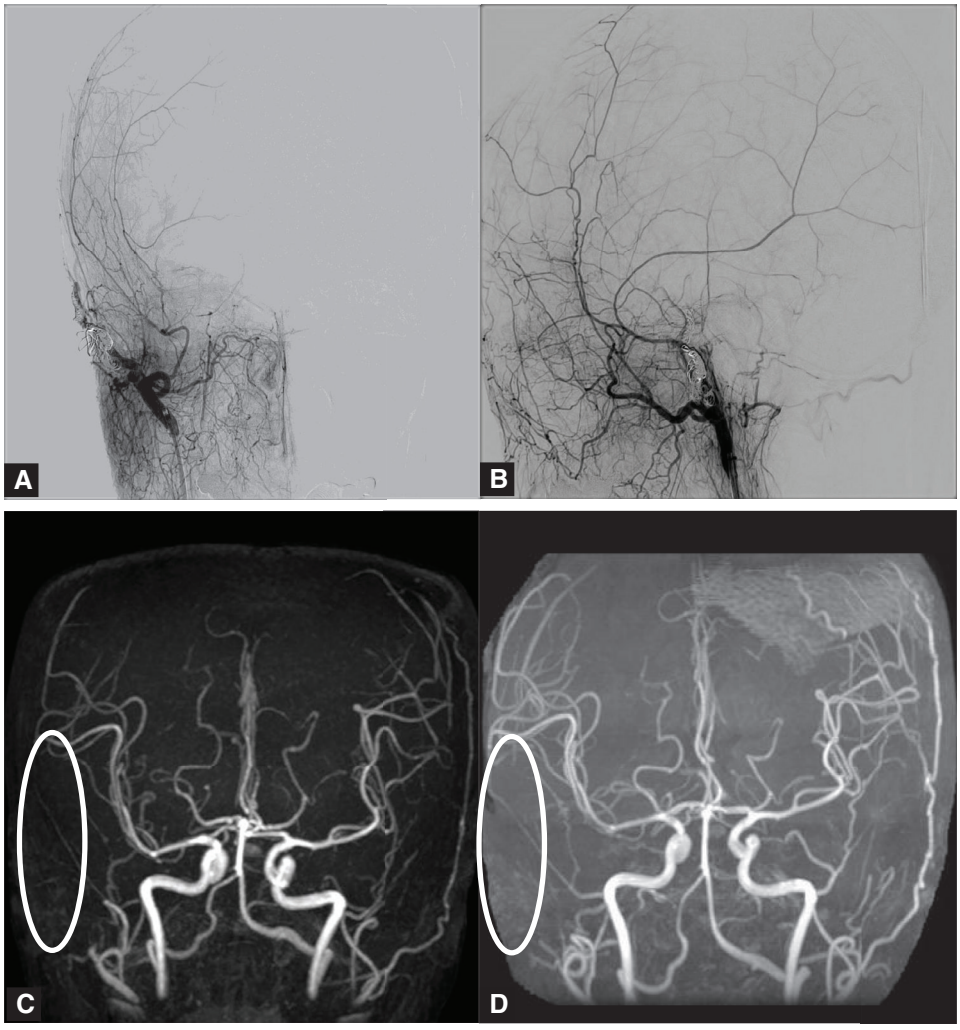

Fig. 3 Postoperative right external carotid angiography and MRA. (**A** and **B**) Anterior–posterior view and lateral view of postoperative digital subtraction angiography. Disappearance of the aneurysm and two AVFs. (**C**) MRA 1 day after endovascular embolization. No demonstration of a right AVF (white circle). (**D**) MRA 6 months after endovascular embolization. No recurrence of a right AVF (white circle). AVF: arteriovenous fistula

hyperplasia of the intima-media complex and adventitia as well as partial indistinctness of the internal elastic lamina, mimicking atherosclerotic vascular disease.[1,2] On the other hand, the pathological description of traumatic AVFs is that of a single, well-formed vascular channel created between an artery and a vein through canalization of a thrombus or an aneurismal sac.[9] A pathological analysis was not possible for our case. The patient had no history or physical evidence of trauma, and she also had no collagen disease or other diseases. These factors suggest that the AVFs were spontaneous and two AVFs existed independently.

Treatment indications are cosmetic deformity, tinnitus, pain, skin erosion with the possibility of rupture of the lesion, and high-output cardiac failure resulting from an arteriovenous shunt.[3] There are two treatment options: surgical resection and endovascular embolization. Curative excision can be done by surgical resection of the fistula, but this method needs a facial-skin incision and includes a massive bleeding risk. A proximal ligation of the feeding artery may cause ischemic necrosis in distal tissue, or recurrence or patency of the AVF may occur due to the collateral supply secondary to an inadequate excision.[1,3,9–11] These risks, however, are similar to proximal embolization in endovascular embolization. Endovascular treatment is less invasive and more useful than surgical resection. A combination of transarterial and transvenous approach is curative method, and transvenous-transaneurysmal approach distal to the STA is a useful treatment option.

As embolization materials have advanced, endovascular treatment has become an important therapeutic modality for

AVFs of the STA. Embolization materials include metallic coils, N-butyl-2-cyanoacrylate (NBCA), and Onyx.[6,12,13] NBCA can be injected via a percutaneous or transarterial route, although this has been noted to lead to an intense inflammatory response that can cause skin necrosis.[12] The properties of the Onyx allow the surgeon to control the developing cast during injection. The disadvantages to the Onyx are pigmentation and a limited ability to penetrate the arterial vascular network as it decreases in caliber and increases in number of finer branches.[12–14] There are several complications after embolization by liquid embolic materials, such as tenderness and hyperemia at the embolization site, skin necrosis over the lesion and permanent, and patchy hair loss that may not improve and can lead to cosmetic problems.[3,12,13] Therefore, the indications for liquid embolic material are limited. In embolization using metallic coils, if too many coils are embolized a palpable mass develops, this may cause tenderness.[3,15] In addition, a serious complication of coil embolization by the transvenous approach is pulmonary embolization due to migration into the systemic circulation.[6,12,13,15] In our case, we avoided forming a palpable mass by using as few coils as possible in the aneurysm. There were two reasons for using a balloon guiding catheter on the venous side. The first reason was to prevent coil migration into the systemic circulation, and the second was to stabilize the catheters.

Coil embolization of the proximal AVF resulted in the disappearance of the distal AVF. We speculate the mechanism of this result to be that the reduction in blood pressure and flow induced the closing of the shunt. There is the possibility that collateral circulation to the distal AVF will develop in the future. The patient has had no recurrence of the AVF on MRA at 6 months after surgery, and there are no skin problems. We will require long-term follow-up that includes checking for any AVF recurrence or skin problems.

Conclusion

We report on a patient who had multiple spontaneous AVFs of the ECA that were successfully treated by coil embolization. Coil embolization of multiple spontaneous AVFs of the ECA using a combined transarterial and transvenous approach can be considered a useful treatment option.

Disclosure Statement

The authors declare no conflicts of interest.

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