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Case Report

Endovascular Treatment of Hemifacial Spasm Associated with a Tentorial DAVF Using Transarterial Onyx Embolization: A Case Report

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Objective: We describe a patient treated with transarterial Onyx embolization for a tentorial dural arteriovenous fistula (DAVF) who presented with hemifacial spasm (HFS).

Case Presentation: A 56-year-old man suffered from right blepharospasm for 4 years, and the symptom gradually spread to the right side of his face with oculo-oral synkinesis. MRI of the brain revealed abnormal multiple flow voids at the surface of brainstem and cerebellar hemisphere. MRA (time of flight) and spoiled gradient recalled echo-revealed abnormal vessels at the posterior fossa indicated arteriovenous shunting. 3D-MRI fusion images showed that a dilated vein was in contact with the root exit zone (REZ) of the right facial nerve. The right carotid angiography displayed a complex tentorial DAVF on the right side. There were multiple feeding vessels drained to the tentorial sinus at the point where the inferior cerebellar vermian vein met, and severe venous congestion was noted. We diagnosed a tentorial DAVF and thought that this was responsible for the right HFS. We used neuroendovascular treatment for this lesion. After transarterial Onyx embolization, his right HFS diminished. MRI after treatment showed that the vein in contact with the REZ of the right facial nerve had shrank.

Conclusion: We experienced a rare case of HFS associated with a DAVF. Our case supports that transarterial Onyx embolization can treat HFS associated with a tentorial DAVF. It is the first description of successful treatment that could be confirmed through postoperative MRI.

Keywords dural arteriovenous fistula, hemifacial spasm, MRI, transarterial Onyx embolization

Introduction

Tentorial dural arteriovenous fistulas (DAVFs) are rare but those with leptomeningeal venous drainage are of high risk. They might result in symptom onset including pulsatile tinnitus, a progressive neurological deficit from venous hypertension and ischemia, or intracranial hemorrhage. We report a rare case of a tentorial DAVF manifesting with a hemifacial spasm (HFS) and the experience of treatment for this patient by transarterial Onyx embolization.

Case Presentation

A 56-year-old man suffered from right blepharospasm for 4 years along with pulsatile tinnitus, with the spasm having gradually spread to the right side of his face.

His neurological examination demonstrated normal facial sensation to light touch and pinprick. Facial twitching was overtly obvious, but there was no facial weakness. Oculo-oral synkinesis of the right side was observed. He had no other complications such as hypertension, diabetes mellitus, and hyperlipidemia, and no history of peripheral facial nerve palsy. Blood analysis did not reveal any abnormal finding.

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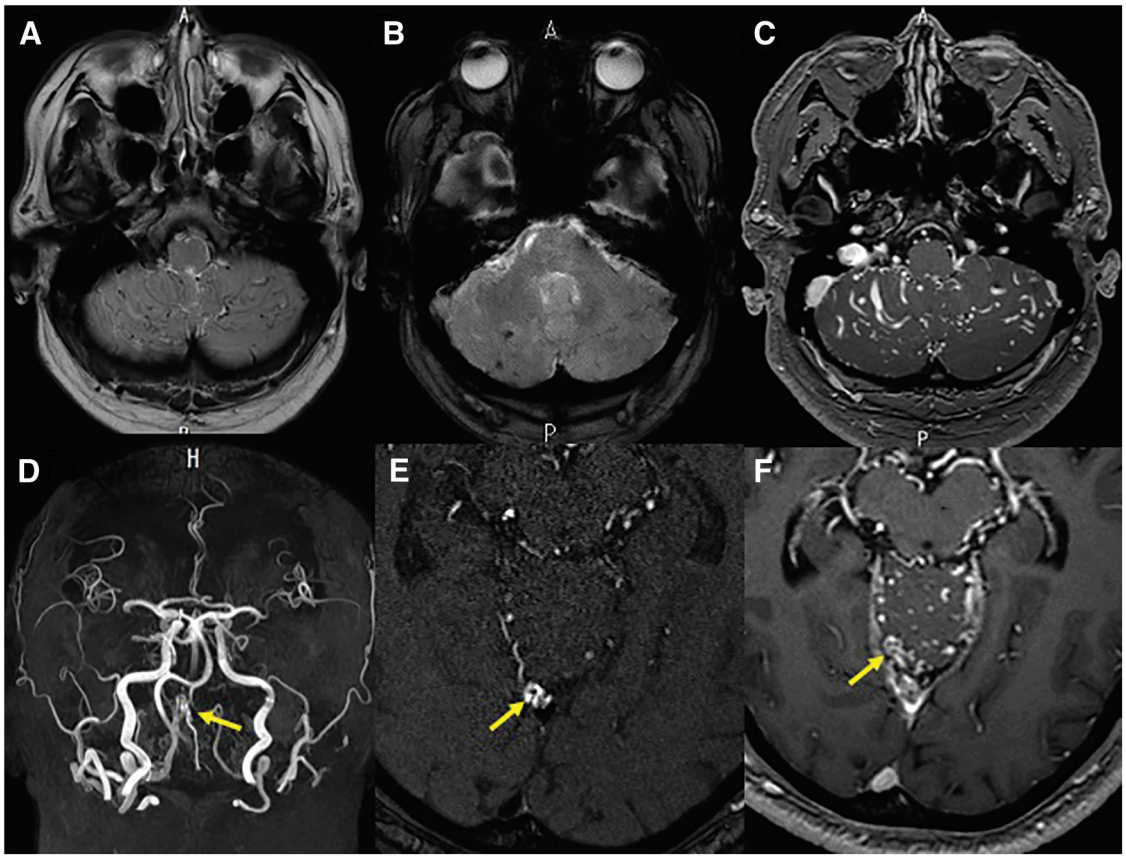

Fig. 1 Pre-treatment MRI. (**A**) T2WI showed abnormal multiple flow voids at the surface of brainstem and cerebellar hemisphere. (**B**) T2*WI showed microbleeds at the cerebellar hemisphere. (**C**) SPGR showed multiple dilated vessels at the posterior fossa. (**D**–**F**) Time of flight and SPGR revealed arteriovenous shunt point (yellow arrows) drain to the inferior vermian vein. SPGR: spoiled gradient recalled echo; T2*WI: T2* weighted image

MRI of the brain revealed abnormal extra-axial multiple flow voids at the surface of the brainstem and cerebellar hemisphere (**Fig. 1A**). T2* weighted image (T2*WI) showed microbleeds at the cerebellar hemisphere (**Fig. 1B**). MRA (time of flight) and spoiled gradient recalled echo-revealed abnormal vessels at the posterior fossa indicated arteriovenous shunting and the shunting point was presumed the tentorial sinus at the point where the inferior cerebellar vermian vein (**Fig. 1C**–**1F**).

3D-MRI fusion images formed by integrating MRA with images of facial nerve extracted by diffusion tensor image (DTI) showed that a dilated vein was in contact with the root exit zone (REZ) of the right facial nerve, and the right posterior inferior cerebellar artery was found outside (**Fig. 2A** and **2B**).

The right carotid angiography displayed a complex tentorial DAVF on the right side. There were multiple feeding vessels including the right occipital artery, middle meningeal artery, bilateral tentorial artery, and bilateral posterior meningeal artery. These arteries drained to the inferior cerebellar vermian vein and severe venous congestion was noted (**Fig. 3A**–**3F**). We diagnosed a tentorial DAVF (Borden type III, Cognard type III) and thought that this was responsible for the right HFS. We planned neuroendovascular treatment for this lesion.

Under general anesthesia, 8Fr Launcher (Medtronic, Minneapolis, MN, USA) was placed at the right common carotid artery, 6Fr FUBUKI (Asahi Intecc, Aichi, Japan) was placed at the right occipital artery as a support catheter, and DeFrictor Nano Catheter (Medico's Hirata, Osaka, Japan) was navigated close to the fistula site with ASAHI CHIKAI 008 (Asahi Intecc). Transarterial embolization (TAE) was initiated by the plug and push technique with Onyx 18 (Covidien, Minneapolis, MN, USA). Onyx penetrated to the area considered to be beyond the fistula point around the cerebellar vermian vein (**Fig. 4A**–**4C**). In all, 2.14 ml of Onyx 18 was injected and we confirmed the disappearance of the shunt. We confirmed no clear arteriovenous shunt by angiography of the external carotid artery, internal carotid artery, and vertebral artery. We had no adverse events during our procedure.

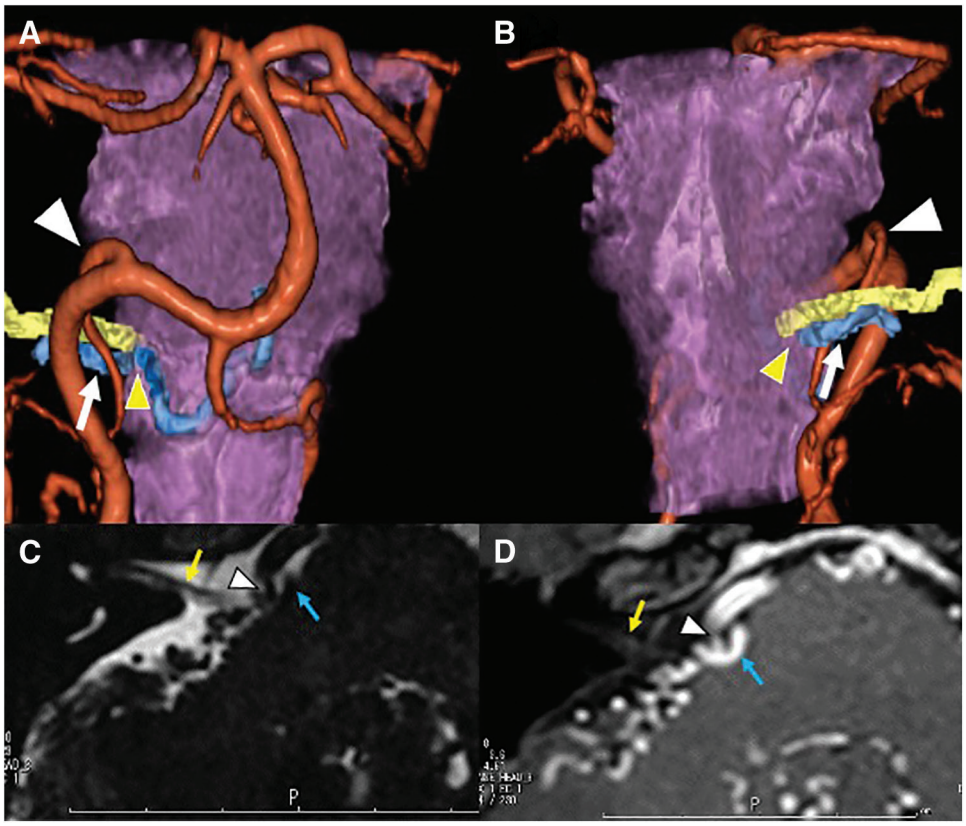

Fig. 2 3D fusion images of MRA and DTI, and heavy CISS and SPGR. Images showed that a dilated vein was in contact with the REZ of the right facial nerve. (**A**) Ventral and (**B**) dorsal views, yellow: facial nerve, blue: dilated vein (white arrows), red: vertebrobasilar system, white arrowheads: posterior inferior cerebellar artery, and yellow arrowheads: REZ of facial nerve. (**C** and **D**) CISS and SPGR images, blue arrows: dilated vein, yellow arrows: facial nerve, and white arrowheads: posterior inferior cerebellar artery. CISS: constructive interference in steady state; DTI: diffusion tensor image; REZ: root exit zone; SPGR: spoiled gradient recalled echo

After neuroendovascular treatment, his right HFS diminished over a week and tinnitus disappeared. MRI after treatment showed that the dilated vein in contact with the REZ of the right facial nerve had shrank (**Fig. 4D** and **4E**).

We confirmed that this pathology was the cause of HFSs.

Discussion

Tentorial DAVFs of nonsinus type have flow pattern that varies among patients, reflecting that the tentorial sinuses intrinsically receive an influx of venous blood from both above and below. DAVF had uniform leptomeningeal venous drainage and caused severe venous congestion. Therefore, they are considered high-risk fistulas as they have progressive features.[1–4] Congestion of the deep venous system causes hemorrhagic or ischemic events at the brainstem, cerebellum, thalamus, and occipital lobe. These appear as gait disturbance, diplopia, visual field defect, and progressive cognitive dysfunction.[5–7] Myelopathy is caused by congestive spinal veins,[8] while sensory disturbance by direct compression of brainstem caused by varices or dilated veins[9] has been reported.

HFS has been reported to be related to vascular compression of the facial nerve at its REZ in the majority of patients.[10–12] HFS is also associated with other pathological conditions such as cerebellopontine tumor and arteriovenous malformation or aneurysm.[13,14]

Recently, MRI sequences have been developed, enabling preoperative analysis of the neurovascular anatomy in the cerebellopontine angle.[15–18] We extracted the REZ of facial nerve with a 3D model by MRA and DTI and created a fusion image of vessels and nerve to evaluate the offending vessel. This method was considered useful in the evaluation of pretreatment.

It is very rare for DAVF to be the cause of HFS, and there have only been two reports thus far (**Table 1**).[19,20]

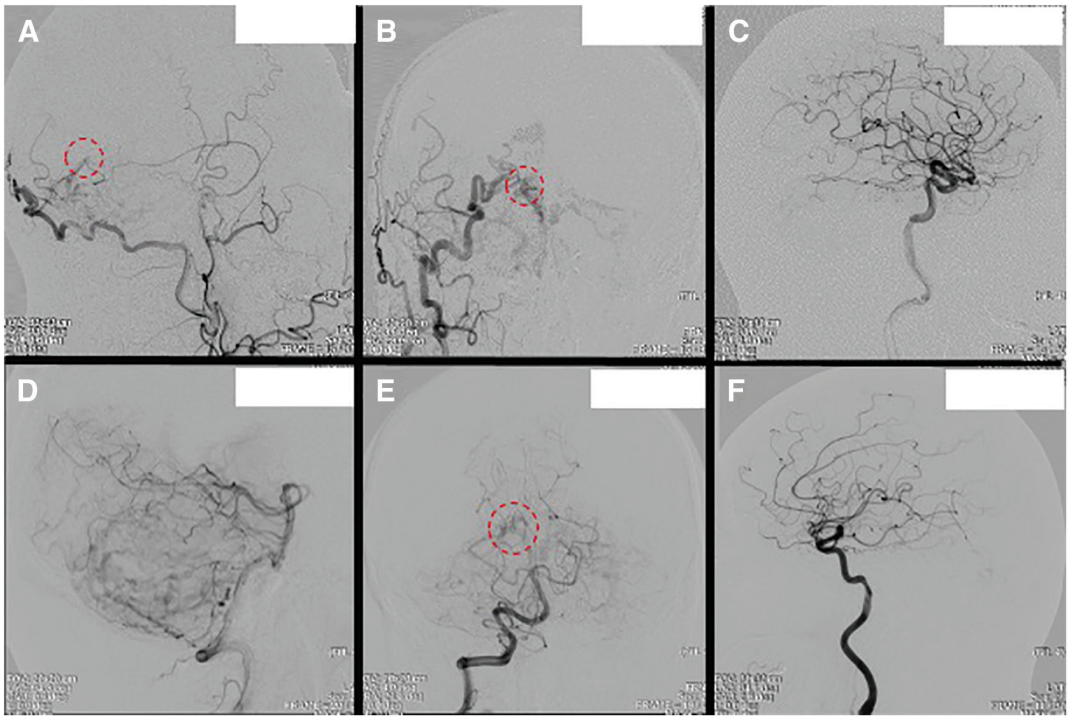

Fig. 3 Pretreatment cerebral angiography. (**A** and **B**) Right external carotid artery angiogram in lateral (**A**) and anteroposterior (**B**) views shows the right tentorial DAVF supplied from the right occipital artery and middle meningeal artery. (**D** and **E**) Right vertebral artery angiogram in lateral (**D**) and anteroposterior (**E**) views shows the right tentorial DAVF supplied from the posterior meningeal artery and venous congestion. Red dotted circles: shunting point of DAVF. (**C**) Right internal carotid artery angiogram. (**F**) Left internal carotid artery angiogram. Bilateral tentorial arteries supply to the tentorial DAVF. DAVF: dural arteriovenous fistula

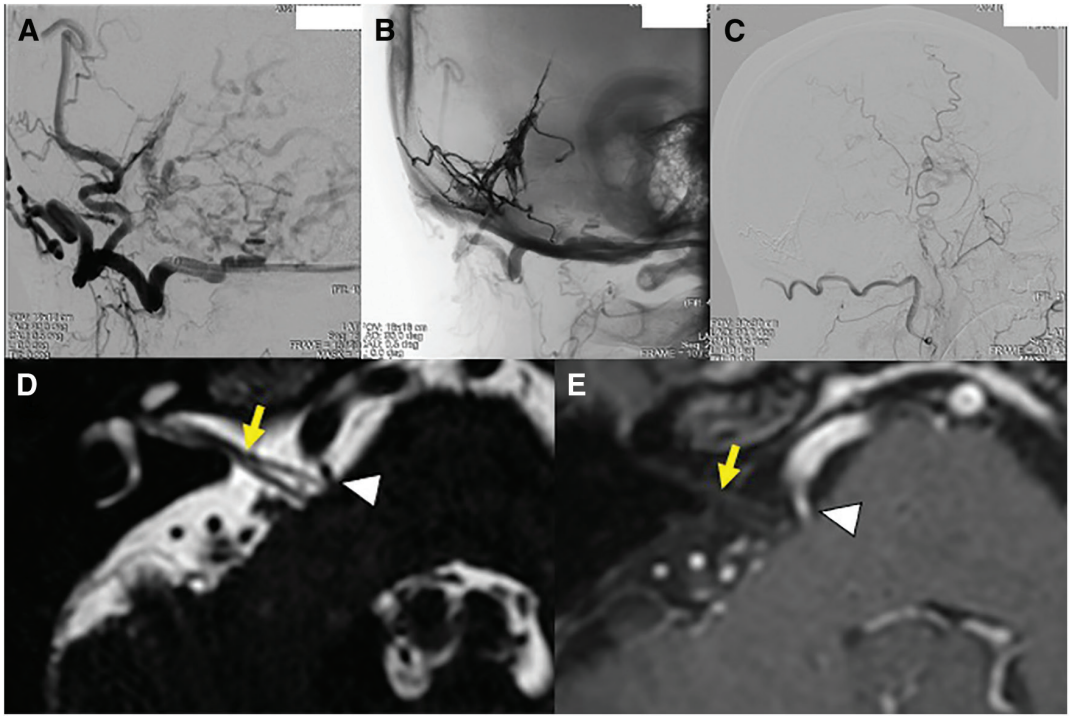

Fig. 4 Upper: The shunt point was depicted from occipital artery angiography (**A**) Transarterial Onyx embolization was performed (**B**) and the shunt has disappeared (**C**). Lower: Posttreatment MRI CISS (**E**) and SPGR with contrast medium (**D**). In posttreatment MRI, the dilated vein was not seen near the REZ of the right facial nerve, and abnormal dilated veins around brainstem and cerebellum were diminished. Yellow arrows: right facial nerve and white arrowheads: posterior inferior cerebellar artery. CISS: constructive interference in steady state; REZ: root exit zone; SPGR: spoiled gradient recalled echo

Table 1 Summary of DAVF associated with HFS

No.	References	Age (years)	Sex	Duration	Other symptom	Location	Side	Varices	Borden/ Cognard type	Treatment	Outcome
1	Deshmukh et al.[19]	50	Man	6 months	Left limb numbness, weakness (transient)	Tentorial	Left	+	III/IV	Craniotomy	Improve
2	Li et al.[20]	56	Man	6 years	Tinnitus	Petrosal	Right	–	III/III	Onyx embolization	Improve
3	Present case	59	Man	5 years	Tinnitus	Tentorial	Right	–	III/III	Onyx embolization	Improve

DAVF: dural arteriovenous fistula; HFS: hemifacial spasm

Deshmukh et al. first described the association of HFS with a tentorial DAVF.[19] A 50-year-old man suffered from progressive left facial twitching. Radiological examinations revealed tentorial DAVF with this pathology being diagnosed as a cause of HFS and craniotomy being performed. His symptom resolved after surgery.

Li et al. reported of HFS associated with the dilated petrosal vein of the DAVF, which had compressed and indented the facial nerve at the REZ, and treated using transarterial Onyx embolization, and that post-embolization, the patient had a prompt improvement in the spasm.[20] Purely venous compression is rarely encountered. El Refaee et al. reported that only one of 249 cases of facial spasms caused by venous compression.[21] Our case was speculated that the right posterior inferior cerebellar vein (PICA) also run in the vicinity of the facial nerve but was somewhat distant from its REZ, and that facial spasms were caused by compression of dilated veins that existed near the REZ. The fact that the facial spasm disappeared as a result of the DAVF treatment suggested the validity of this speculation.

We also used transarterial Onyx embolization and got a good posttreatment condition.

Her facial spasm has disappeared at three months post treatment, but further long-term follow-up is necessary.

Our case is the second case of HFS caused by DAVF treated by transarterial Onyx embolization and is the first report with a detailed description of pre- and posttreatment MRI findings.

Conclusion

We experienced a rare case of HFS associated with a tentorial DAVF. 3D fusion image of MRI and MRA is useful for evaluating offending vessels of REZ of facial nerve. Endovascular treatment is effective for DAVF associated with HFS.

Disclosure Statement

None of the authors have any commercial or financial involvement in connection with this study that represents or appears to represent any conflicts of interest.

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