Vagal Nerve Palsy After Transarterial Embolization of Transverse-Sigmoid Dural Arteriovenous Fistula Using Onyx

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Objective: We report a case of a 70-year-old man who developed a transverse-sigmoid dural arteriovenous fistula (TS-DAVF) that was successfully treated by transarterial embolization (TAE) with Onyx. *Case Presentation:* The patient presented with sudden and progressive disturbance of consciousness and left hemiparesis. Magnetic resonance imaging (MRI) revealed venous infarction and hemorrhagic changes with brain swelling in the right parietal lobe. Angiography revealed a right TS-DAVF and multiple occlusions with retrograde leptomeningeal venous drainage into the cortical veins. The TS-DAVF was graded as Borden type III and Cognard type IIa+b. Because of its progressive clinical nature and wide distribution of DAVF in the occluded sinus wall, he underwent emergent TAE with liquid embolic materials including *n*-butyl cyanoacrylate and Onyx under informed consent by his family. Complete obliteration of the TS-DAVF was achieved, leading to a marked amelioration of symptoms, and MRI after treatment confirmed a decrease in the brain swelling. However, he suffered transient dysphagia due to right vagal nerve palsy caused by occlusion of vasa nervorum of ascending pharyngeal artery. He returned home 5 months later with a modified Rankin Scale of 1. *Conclusions:* TAE with Onyx appears to be effective for aggressive TS-DAVF with a widely distributed shunt. However, the blood supply to the cranial nerves and potentially dangerous anastomoses between the external-internal carotid artery and vertebral artery should be taken into account to avoid serious complications.
Key Words: Dural arteriovenous fistula—transarterial embolization—Onyx—complication—vagal nerve palsy

Introduction

Transarterial embolization (TAE) using Onyx (Medtronic, Minneapolis, MN) has commonly been done for dural arteriovenous fistula (DAVF) particularly with retrograde leptomeningeal venous drainage (RLVD) that exhibited rapid deterioration of neurological symptoms. However, there is a potential risk of cranial nerve palsy in TAE with Onyx. Here, we report a case of a transverse-sigmoid sinus DAVF (TS-DAVF) treated by TAE with Onyx, which exhibited transient vagal nerve palsy after treatment.

Case Presentation

A 70-year-old man, who had a history of subcortical intracerebral hemorrhage (ICH) in the right frontal lobe 3 years ago, presented with a sudden onset of headache,

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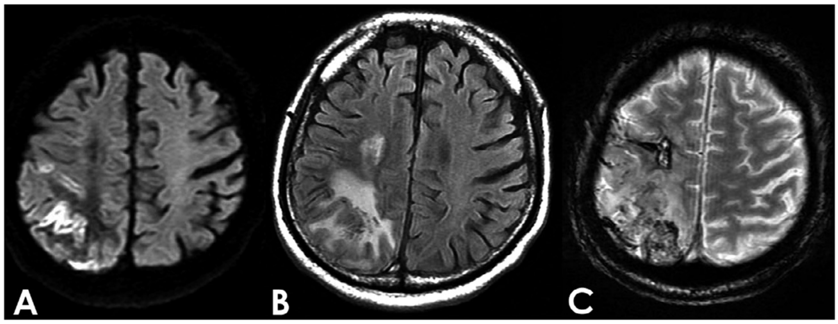

Figure 1. *Magnetic resonance images on arrival. (A) Diffusion-weighted image (DWI) showing acute ischemic changes in the right parietal lobe. (B) Fluid-attenuated inversion recovery (FLAIR) image showing brain swelling around the ischemic legion. (C) T2*-weighted image showing multiple hemorrhage in the right parietal lobe. A chronic lesion without changes in the DWI and FLAIR is also shown in the right frontal lobe.*

nausea, and the left hemisensory numbness. Neuroradiological examinations revealed the right TS-DAVF with brain swelling in the right parietal lobe at the local hospital. He was transferred to us for emergent treatment because of rapid progression of symptoms. On admission, he was somnolent, showing conjugate eye deviation to the right and the left hemiparesis (Manual Muscle Testing scores: 2/5)

Neuroradiological Findings

The MRI showed high signal intensity on diffusion-weighted images (DWI) (Fig 1A), as well as a high signal intensity region predominantly in the white matter on fluid-attenuated inversion recovery (FLAIR) (Fig 1B), and a low signal intensity region within this on T2*-weighted image (Fig 1C) in the right parietal lobe. Each of these findings indicated venous infarction and hemorrhagic changes with brain swelling. Another low signal intensity region within this on T2*-weighted image was observed in the right frontal lobe which was consistent with the past medical history of ICH. Digital subtraction angiography of the right external carotid artery indicated the right TS-DAVF fed by the right occipital artery (OA) and right middle meningeal artery (MMA) (Fig 2, A,B). Both ends of the dural sinus were occluded, showing isolated sinus, and shunted flow was drained into the cortical veins in the right parieto-occipital region, showing prominent RLVD (Fig 2B). The superior sagittal sinus and the right transverse and sigmoid sinuses were not visualized, while the deep veins and left transverse and sigmoid sinuses appeared normal. The right internal carotid arteriogram showed marked cork-screw sign indicating severe venous congestion in the right cerebral hemisphere (Fig 3). These findings were consistent with Borden type III[1] and Cognard type IIa+b[2] DAVF.

Endovascular Treatment

Because of rapid progression of acute venous congestion and hemorrhagic changes, we decided to perform transarterial embolization. We obtained written informed consent from the patient's family prior to the procedure including the off-label use of Onyx for DAVF at the time of treatment.

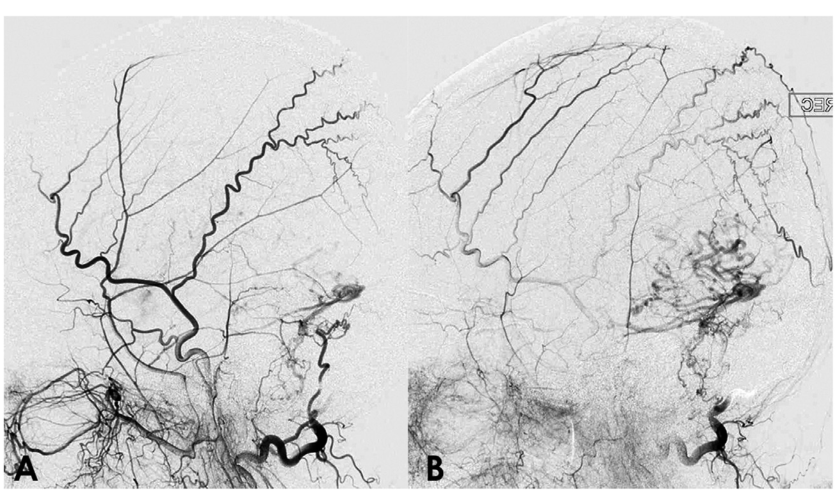

Figure 2. *(A) The right external carotid artery angiogram (ECAG; lateral view, early arterial phase), showing a dural arteriovenous fistula fed by the right occipital artery and right middle meningeal artery. (B) The late arterial phase the ECAG, showing prominent retrograde leptomeningeal venous drainage in the right temporoparietal area.*

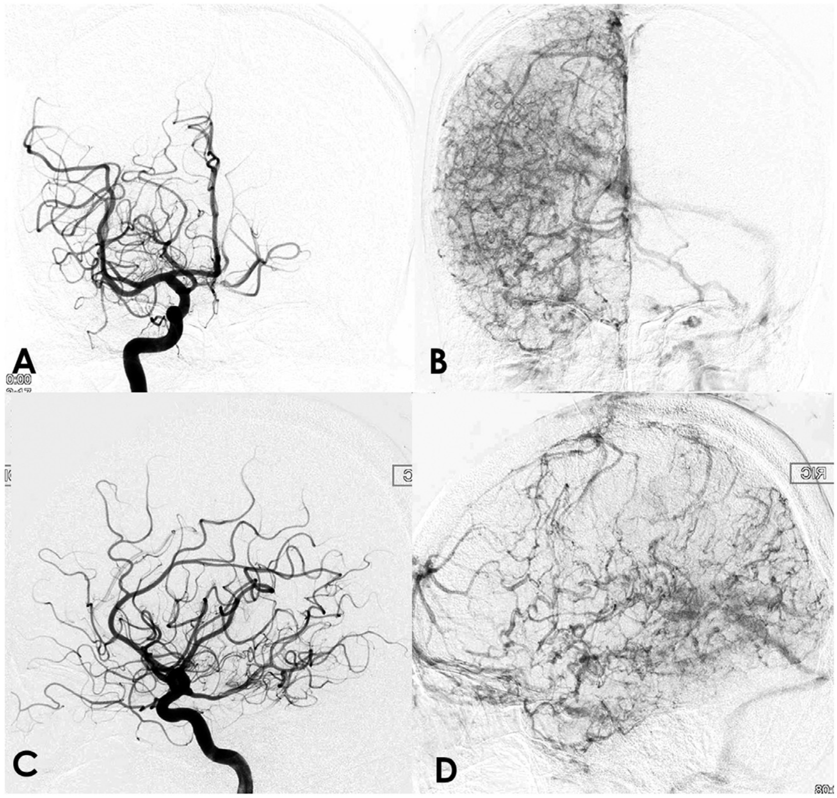

Figure 3. *The right internal carotid artery angiogram. Anteroposterior view ((A) Arterial phase, (B): venous phase) and lateral view ((C) arterial phase, (D) venous phase), showing dural sinus occlusion in the superior sagittal sinus and the right transverse sinus. Severe venous congestion is observed in the right internal carotid artery territory showing cork-screw sign.*

Under general anesthesia, a 7-Fr guiding catheter (RoadMaster 7Fr, 90 cm, Goodman, Aichi, Japan) was placed in the right external carotid artery (ECA). A microcatheter (Excelsior SL-10, Stryker, Kalamazoo, MI) was navigated to the right OA, which was occluded distal to the mastoid branch using four coils (one AXIUM 3D 3 mm/6 cm, one AXIUM HELIX 3 mm/8 cm, and two AXIUM HELIX 2 mm/6 cm; Medtronic) (Fig 4A), followed by the occlusion of OA at the origin of the mastoid branch (Fig 4B), using heated, low-concentration NBCA (Histoacryl; Aesculap AG, Tuttlingen, Germany; at concentrations of 15% NBCA 0.11 cc, 12% NBCA 1.31 cc, and 12% NBCA 0.21 cc) by the continuous column method (Fig 4C). Subsequently, we introduced a microcatheter (1.5-Fr Marathon; Medtronic) into the parietal branch of the MMA (Fig 5A) and advanced it to the immediate vicinity of the shunt point (Fig 5B). Then, the feeding artery and the affected venous sinus was embolized with Onyx-18 (a total of 1.2 cc; Medtronic) by the plug and push method[3] under digital subtraction angiography (DSA) and blank road-mapping guidance maintaining careful attention on the movement of Onyx. We intentionally penetrated a slight amount of Onyx into the drainage veins and the feeding arteries other than MMA

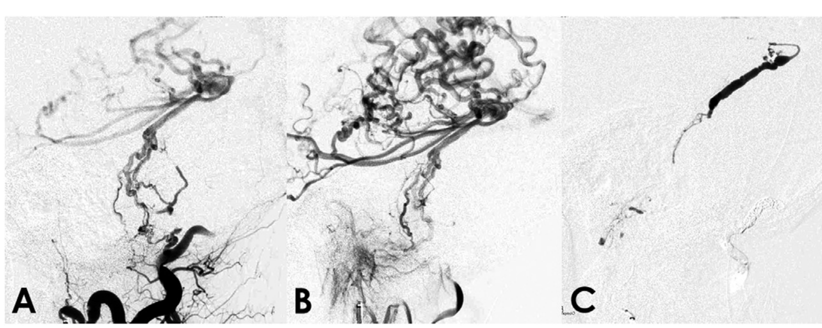

Figure 4. *(A) The right external carotid artery angiogram (lateral view, arterial phase) showing that the main trunk of the OA distal to the mastoid branch is occluded with coils. (B) The right external carotid artery angiogram (lateral view, late arterial phase) after coil embolization showing that the mastoid branch of the OA supplies the dural arteriovenous fistula causing severe retrograde leptomeningeal venous drainage. (C) Superselective injection during transarterial embolization of the mastoid branch of the OA using* n*-butyl cyanoacrylate.*

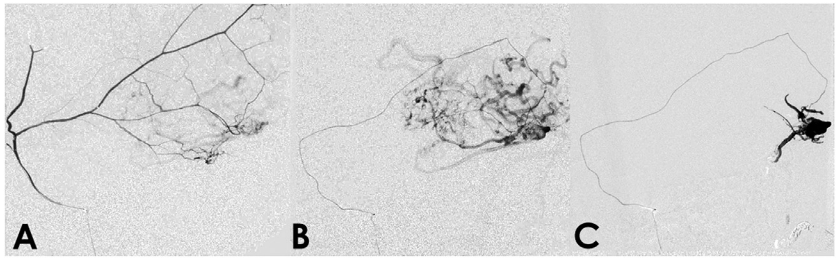

Figure 5. *Superselective angiogram (lateral view) of the middle meningeal artery (MMA). (A) Injection at the origin of the MMA, showing branches of MMA feeding the dural arteriovenous fistula. (B) Injection just proximal to the fistula, showing severe retrograde leptomeningeal venous drainage. (C) Superselective injection during transarterial embolization using Onyx.*

via the affected sinus (Fig 5C) to obtain complete obliteration (Fig 6, A,B).

Postoperative Course

General anesthesia was maintained until the next day and discontinued after confirming that there were no new lesions by head CT. The patient's symptoms improved to arousable (JCS 3) and left hemiparesis of MMT 4/5. On the seventh day, the patient became alert and his left arm and leg paralysis were assessed at MMT 5-/5, while he had suffered from dysphagia and hoarseness since the third day. No brainstem infarction was observed on MRI and an otorhinolaryngological examination revealed right vocal cord paralysis due to right vagal nerve palsy. An MRI performed on the 21st day showed significant improvement of

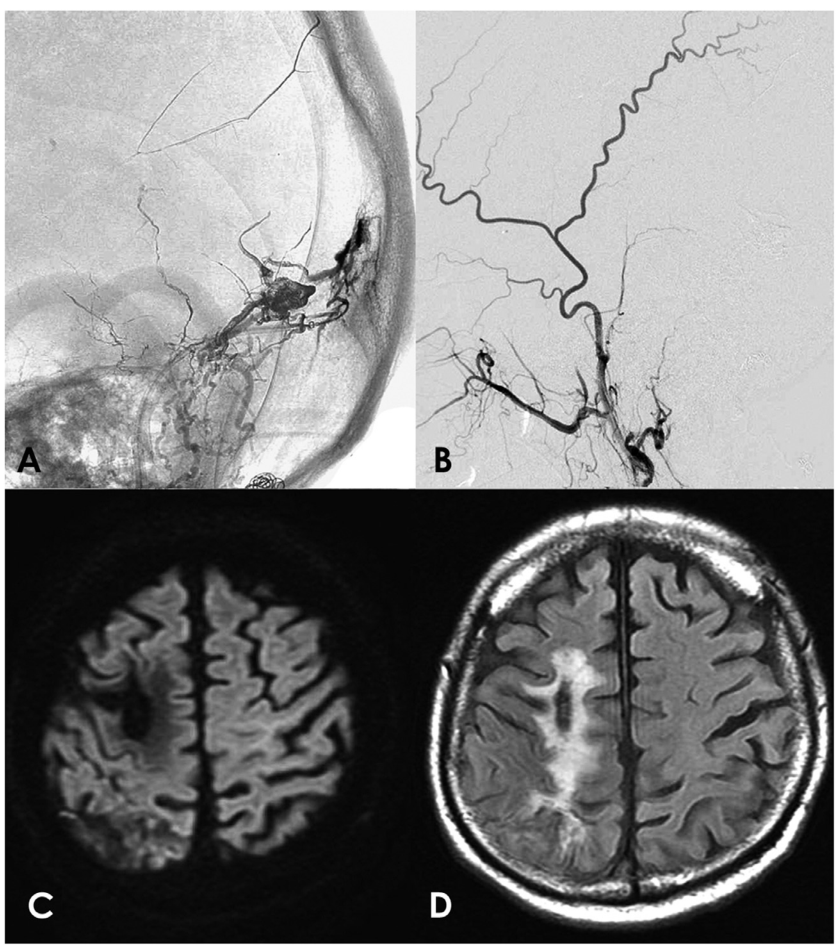

Figure 6. *(A) Plain craniogram (lateral view) after treatment, showing cast of* n*-butyl cyanoacrylate in the right occipital artery (OA) and cast of Onyx in the right middle meningeal artery (MMA). Note the penetration of Onyx into part of the draining veins and small feeding arteries in addition to the affected sinus. (B) The right external carotid artery angiogram (lateral view, arterial phase) after treatment, showing complete obliteration of the dural arteriovenous fistula. (C and D) MRI performed 21 days after treatment. (C) Diffusion weighted image. (D) Fluid-attenuated inversion recovery image), showing the disappearance of the ischemic area and amelioration of the brain swelling.*

the brain swelling in the right parietal lobe (Fig 6, C,D), and cerebral angiography performed on the 30th day of hospitalization indicated no recurrence of the DAVF. Because of continued dysphagia, a gastrostomy was performed on the 43rd day of hospitalization. Five months after onset, the dysphagia had improved to the extent that he was able to take food by mouths, and the gastrostomy was removed. He was discharged to his home by walk unassisted with a modified Rankin Scale of 1.

Discussion

DAVF takes a variety of forms depending on its location and venous drainage pattern. It has been reported that the annual mortality rate for the aggressive type of DAVF with RLVD is 10.4% and that the annual risk of central nervous system disorders is 15% (hemorrhagic 8.1%, non-hemorrhagic 6.9%).[4] Emergent treatment is required in cases with rapid progression of symptoms due to venous congestion and/or hemorrhage [3-7].

For an aggressive DAVF, it is usual to perform endovascular treatment using TAE/TVE or surgical treatment via direct surgery.[8] TVE includes procedures such as guiding a catheter through the occluded sinus and direct puncture of the affected sinus via a small craniotomy. Conventional TAE with NBCA is difficult to maintain its administration over the long term for curative treatment because the adhesive property. Therefore, the cure rate ranges from 37% to 75%, indicating limited effectiveness[9,10] by the conventional therapy. Onyx, which is a modification of ethylene vinyl alcohol copolymer (EVAL)[11,12], compared to NBCA, is less adhesive to the catheter and offers high permeability to the lesion. Because these characteristics allow a large amount of embolic agent to be introduced into a single thin blood vessel, Onyx is extremely effective when used in the endovascular treatment of DAVF.[3,13,14] Because it is possible for Onyx to occlude shunts that cover a wide area, reports on its use for embolization of aggressive DAVFs have indicated a cure rate of 80%, as demonstrated by angiograms, and that Onyx monotherapy was the type of treatment[15-17] and selected in 83% of cases in a report.[17] Recently, the Japanese government has just approved the use of Onyx for formidable DAVF.

Although the patient in this study recovered 5 months after surgery, he suffered long-term dysphagia due to right vagal nerve palsy. There are some reports of cranial nerve palsy after TAE with Onyx.[18-21] The most affected cranial nerves are facial nerve and trigeminal nerve, followed by abducent nerve and hypoglossal nerve.[18-21] In these reports, it is mentioned that the cranial nerve palsy recovered spontaneously within several months. The causative mechanism of cranial nerve palsy after TAE using Onyx remained unclear, but some hypotheses have been argued. Nyberg et al[22] reported two cases of cranial nerve palsy, in which they mentioned that these neuropathies are caused by axonometric traction injury related to microcatheter extraction from Onyx cast. The other possibility may be toxicity of Onyx or dimethyl sulfoxide which is contained in Onyx mixture. The most likely cause of cranial nerve palsy is mentioned that penetration of Onyx into vasa nervorum.[20,21,23] Even though we could not find any report of vagal nerve palsy after TAE with Onyx for DAVF, the vagal nerve palsy in this case may have been caused by the back-flow into the neuromeningeal branch of ascending pharyngeal artery via the arterial network around the shunt, either by NBCA from OA or Onyx from MMA. It is necessary to pay close attention to the possibility of cranial nerve palsy due to occlusion of the vasa nervorum, as well as the possibility of dangerous anastomosis between the external and internal carotid arteries or the vertebral artery.

We used coils and NBCA for TAE of OA mastoid branch prior to TAE for MMA to reduce the shunt flow via OA. However, such an assist procedure, particularly using the liquid embolic material, may increase the risk of complication. Considering the excellent nature of Onyx to penetrate into the shunt with reflux to other feeders, assist embolization may not be necessary for Onyx embolization for DAVF via the main target feeder.

Conclusions

We have reported a case in which the use of TAE with Onyx resulted in the successful treatment of TS-DAVF, which had resulted in the rapid deterioration of neurological symptoms. Consideration should be given to the possibility that reflux of Onyx into the vasa nervorum of the ascending pharyngeal artery may cause the lower cranial nerve palsy, while Onyx TAE can be an effective therapeutic option in cases of aggressive types of DAVF.

Conflict of interest

There are no conflicts of interest to declare in all authors.

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