

Hearing Disturbance After Transvenous Embolization of Dural Arteriovenous Fistula Involving the Anterior Condylar Confluence: case report

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●Abstract●

Objective: We report a case of hearing disturbance occurring as a rare complication of transvenous embolization of a dural arteriovenous fistula (AVF) involving the anterior condylar confluence (ACC).

Case presentation: A 61-year-old man presented with hearing disturbance after transvenous embolization of a dural AVF involving the ACC. The manifestations of the dural AVF were left ocular symptoms such as conjunctival injection, chemosis, and diplopia. An angiogram located the fistula in the left ACC. And the ACC dural AVF was supplied mainly by the left ascending pharyngeal artery and drained from the ACC into the left inferior petrosal sinus, cavernous sinus, and superior ophthalmic vein retrogradely. It had no other drainage pathway angiographically. We attempted but failed to advance a microcatheter into the fistula at the ACC by the transvenous approach. Therefore we inserted the microcatheter into the left inferior petrosal sinus (IPS) and occluded the left IPS (the only available drainage route from the ACC) with platinum detachable coils. The final angiogram revealed almost no AVF remained. Soon thereafter left ocular symptoms improved, however, a left neurosensory hearing disturbance appeared 3 days later. The hearing disturbance was treated with steroid therapy and systemic heparinization immediately but was unresponsive to this treatment.

Conclusion: The hearing disturbance may be due to venous circulatory failure of the inner ear after occlusion of the IPS. The fistula, and not the IPS, should be occluded in the treatment of ACC dural AVF to avoid hearing complications.

●Key Words●

anterior condylar confluence, dural arteriovenous fistula, hearing disturbance, embolization, inferior petrosal sinus

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Introduction

Dural arteriovenous fistulas (AVFs) commonly occur in the cavernous sinus, transverse sinus, and sigmoid sinus. Recently, dural AVFs involving anterior condylar confluence (ACC dural AVFs) have also been recognized and treated mostly by transvenous embolization (TVE)^{1,4-7,9-12)}. Since the exact location of the fistula can now be determined with three-dimensional angiography using recently developed digital subtraction angiography routines, selective TVE of the fistula with coils has been performed. One of the main complications of TVE of ACC

dural AVFs is hypoglossal nerve palsy, when the dural AVF involves both the ACC and the hypoglossal canal^{6,7,9,10,12)}. Here we present a case of hearing disturbance following TVE of the ACC dural AVF, which is a rare complication of this procedure.

Case report

A 61-year-old man suffered from idiopathic left pulsatile tinnitus three months ago but it disappeared in a month. Then, left conjunctival injection, chemosis, and diplopia appeared. He was referred to our hospital with suspected left carotid-cavernous fistula (CCF). Neurological findings

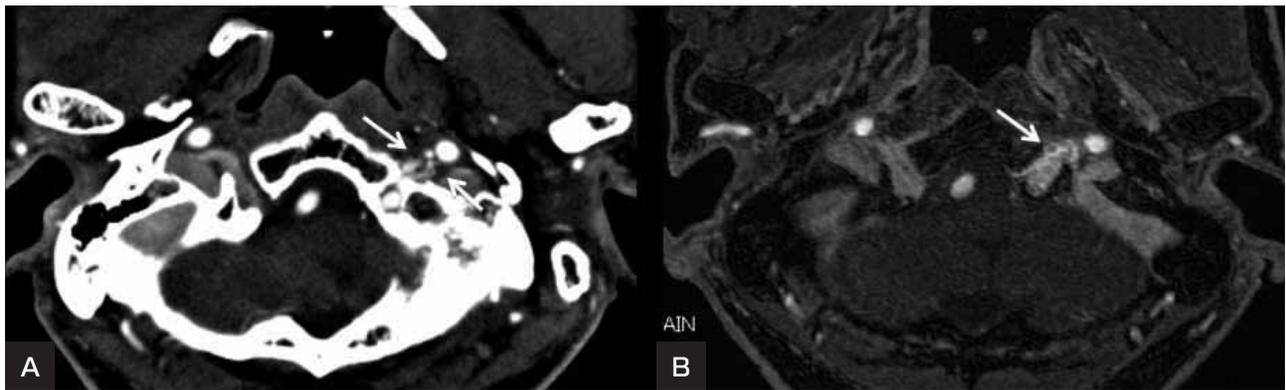


Fig. 1 Source images of computed tomography (A) and magnetic resonance imaging after contrast enhancement (B) revealed small multiple arteries (arrows) at the outer side of the left hypoglossal canal.

were chemosis of the left eye, left abducens nerve palsy, and increased intraocular pressure (30 mmHg). Dural AVF involving the left cavernous sinus (CS) was suspected from the medical history and neurological findings.

Source images of computed tomography and magnetic resonance imaging after contrast enhancement revealed small multiple arteries at the outer side of the left hypoglossal canal (Fig. 1A, B). Left external carotid angiography revealed the presence of a dural AVF adjacent to the left jugular bulb, which was supplied mainly by the left ascending pharyngeal artery (Fig. 2, 3A, B). The right ascending pharyngeal artery was not involved. The fistula was located at the anterior condylar confluence (ACC). The drainage route was the left superior ophthalmic vein (SOV) via the left inferior petrosal sinus (IPS) and CS retrogradely and no other drainage pathway was identified on angiography.

TVE was performed via the right femoral vein. Using a 5 Fr. guiding catheter positioned in the left jugular vein, we first attempted to insert a microcatheter directly into the fistula at the ACC, but failed. Then we attempted to advance it from the jugular vein through the IPS into the fistula at the ACC, but failed again. Therefore the microcatheter was placed into the left IPS, which we occluded with detachable coils since it seemed to be the only drainage route from the ACC. Angiography still showed a small residual fistula, which we treated with transarterial embolization from the left pharyngeal artery

using Spongel (Yamanouchi, Tokyo, Japan). The final angiography revealed a minimal residual ACC dural AVF with almost no drainage (Fig. 3C, D).

Though his left ocular symptoms (conjunctival injection, chemosis) improved within a few days, a left neurosensory hearing disturbance occurred 3 days after the treatment (postoperative day 3; POD3). He was immediately treated with steroids and systemic heparinization. Mild dysphagia and dysarthria due to hypoglossal nerve palsy occurred POD6. Furthermore episodes of peripheral vertigo occurred POD16 and 26, both of which improved within days. Angiography demonstrated complete obliteration of the ACC dural AVF 1 month after the treatment (Fig. 4). Though the left hypoglossal nerve palsy and vertigo disappeared within 1 month, hearing disturbance did not improve and remained 2 years after the treatment.

Discussion

ACC dural AVF has recently become one of the major topics in neurointerventional radiology^{1,4-7,9-12}. This is due to the rapid technological progress in digital subtraction angiography devices. Selective three-dimensional angiography and tomographic imaging enables us to understand the angioarchitecture of the vascular lesions accurately, especially in the craniocervical region¹¹. Using this knowledge, most cases of ACC dural AVF have been treated by selective TVE of the fistula with less coils and a shorter operative time than before.

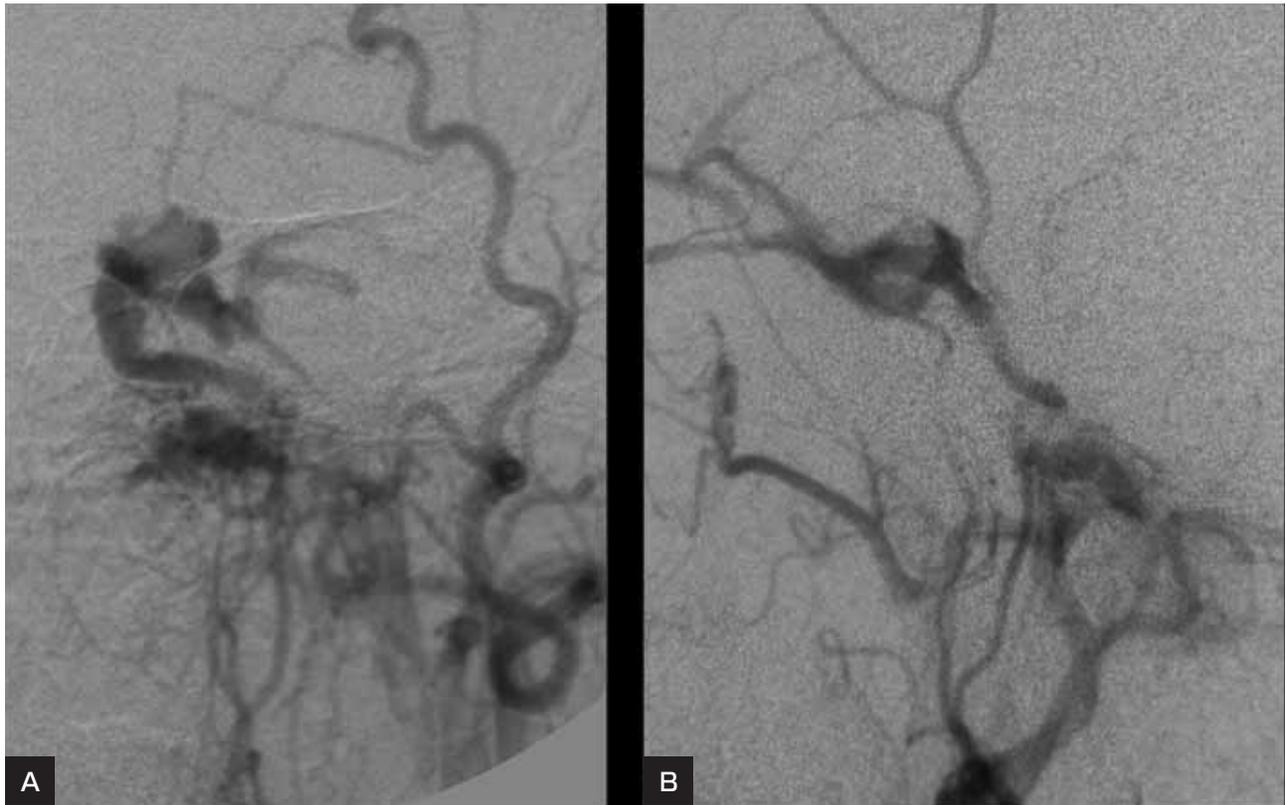


Fig. 2

Frontal (A) and lateral (B) views of the left external carotid artery angiogram showing a dural AVF adjacent to the jugular bulb with retrograde venous drainage to the SOV via the left IPS and left cavernous sinus. Antegrade venous drainage to the jugular vein is hardly seen.

AVF: arteriovenous fistula, IPS: inferior petrosal sinus, SOV: superior ophthalmic vein

The symptomatology of dural AVF is understandable in terms of anatomical knowledge of the craniocervical venous system¹¹⁾. In general, symptoms of dural AVF are related to venous drainage pattern^{1,4-7,9,10,12)}. ACC dural AVF drains into the ipsilateral jugular vein antegradely, resulting in symptoms of pulse-synchronous tinnitus. Occasionally ACC dural AVF drains into the ipsilateral IPS, CS, and SOV retrogradely, resulting in ipsilateral ocular symptoms (such as proptosis, chemosis, and diplopia due to abducens and/or oculomotor nerve palsy), which are the same as the symptoms of CS dural AVF. An ACC dural AVF draining into the ipsilateral anterior condylar vein in the hypoglossal canal uncommonly results in symptoms of ipsilateral hypoglossal nerve palsy.

In our case, pulsatile tinnitus was recognized at first, and disappeared later. It was speculated that an initial direct drainage tract from the ACC into the jugular vein was occluded due to thrombosis caused by continuous venous

hypertension. Preoperative symptoms were ocular symptoms because drainage from the AVF flowed only retrogradely into the ipsilateral IPS, CS, and SOV. Our attempts to introduce a microcatheter into the fistula at the ACC from the jugular vein directly or from the IPS via the jugular vein, failed. Consequently, we occluded the IPS with detachable coils without occluding the fistula at the ACC. Final angiography revealed a minimal residual ACC dural AVF. His ocular symptoms improved immediately (POD2), but later a hearing disturbance (POD3), dysphagia / dysarthria (POD6), and vertigo (POD16, 26) occurred. We speculated that the hearing disturbance and vertigo were due to venous circulatory failure of the inner ear caused by occlusion of the IPS involving the normal venous tract of the inner ear. A textbook (i.e., the Atlas of Otology) demonstrates that the venous return from the cochlea (the organ of hearing) drains into the IPS and/or jugular bulb passing through

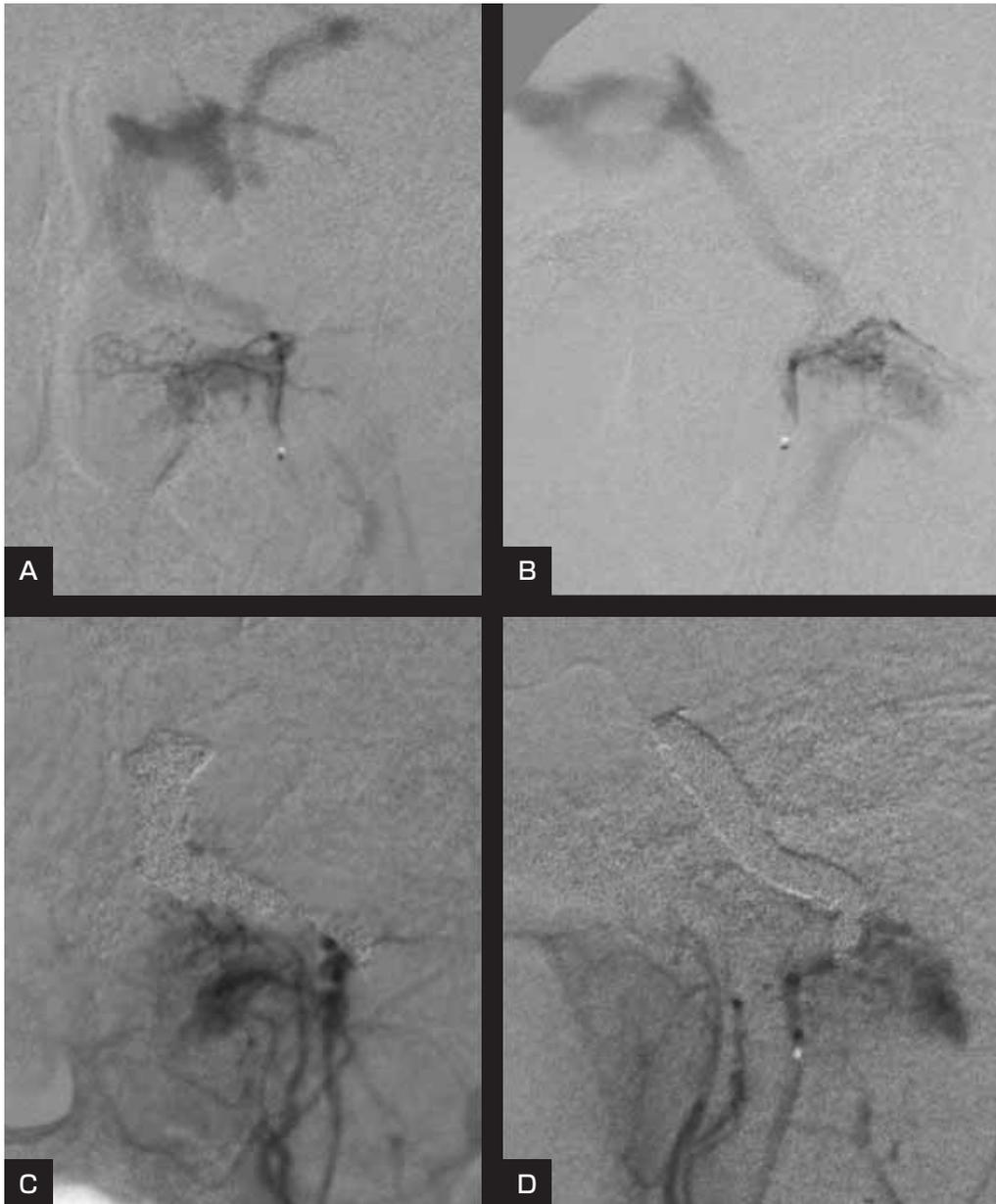


Fig. 3

Frontal (A) and lateral (B) views of an angiogram of the left ascending pharyngeal artery showing a dural AVF adjacent to the jugular bulb with retrograde venous drainage to the SOV via left IPS. Antegrade venous drainage to the jugular vein is hardly seen.

Frontal (C) and lateral (D) views of an angiogram of the left external carotid artery after embolization of the left IPS showing a residual dural AVF adjacent to the jugular bulb without any venous drainage.

AVF: arteriovenous fistula, IPS: inferior petrosal sinus, SOV: superior ophthalmic vein

the vein of the cochlear aqueduct (inferior cochlear vein), and that the venous return from the semicircular canals (mediating the labyrinthine sense, vertigo, and dizziness) drains into the sigmoid sinus passing through the vein of the vestibular aqueduct, and/or drains into the IPS

passing through the vestibulocochlear vein and the vein of the cochlear aqueduct (Fig. 5)⁸⁾. Therefore it is speculated that occlusion of the IPS could cause hearing disturbance and vertigo/dizziness due to the venous hypertension of the cochlea and vestibular labyrinth.

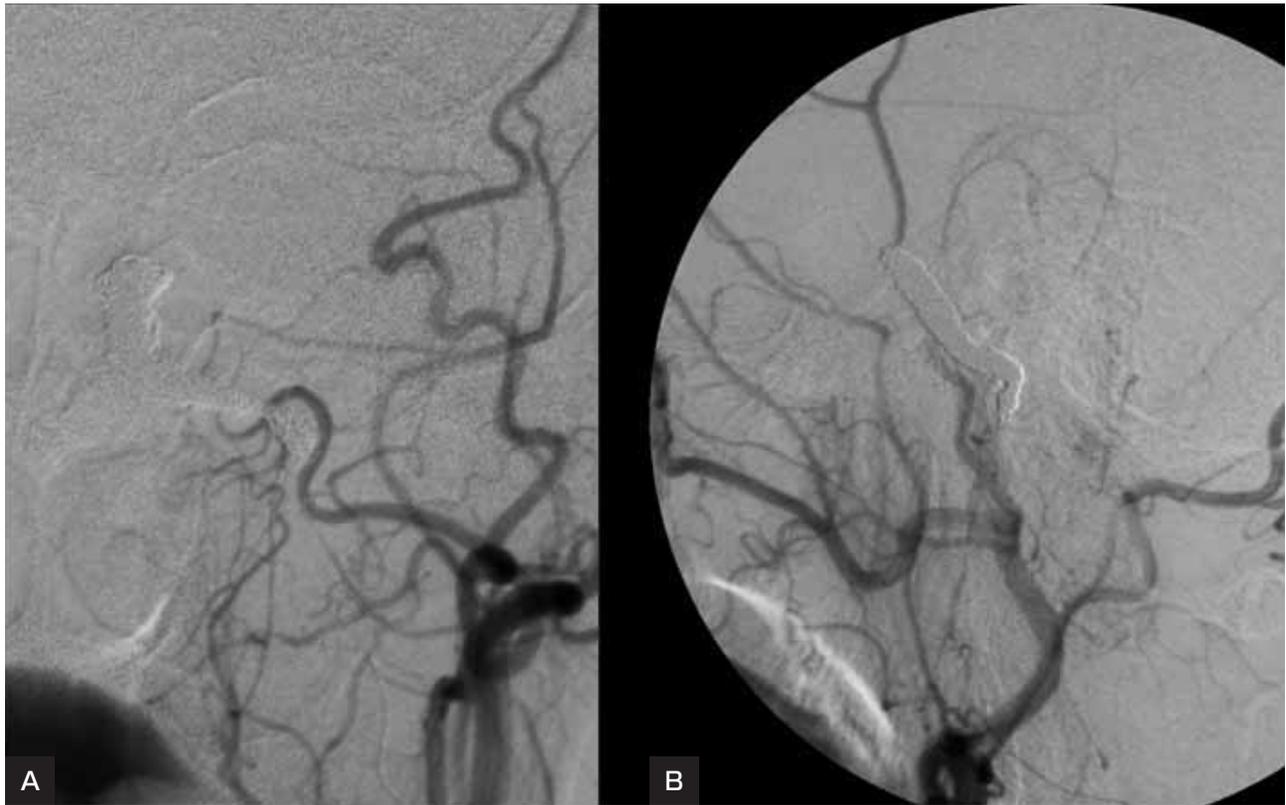


Fig. 4
Frontal (A) and lateral (B) views of the left external carotid artery angiogram after embolization a month later revealing complete occlusion of a residual dural AVF.
AVF: arteriovenous fistula

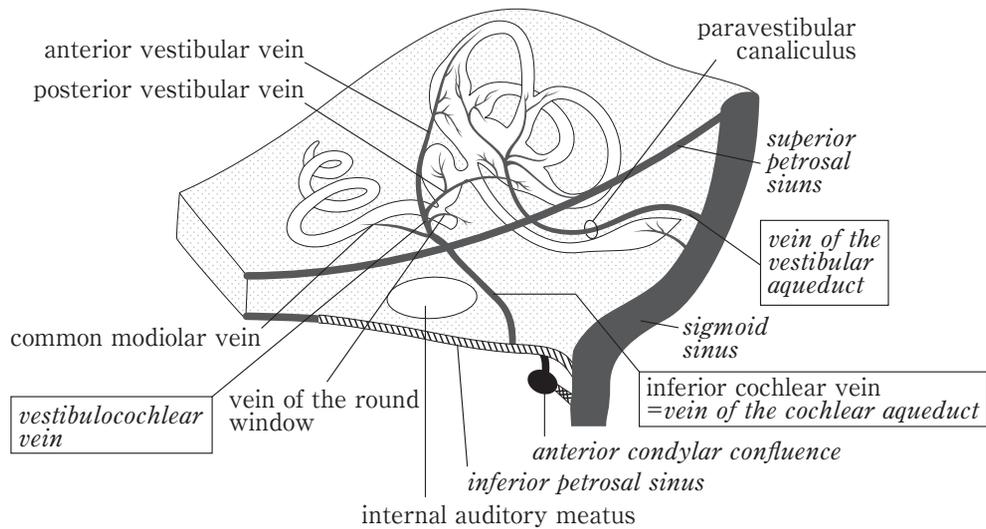


Fig. 5
Schematic representation of veins of the internal ear (Modified from Nomura K⁸⁾). Part of the embolized IPS (hatched area) and the direct tract into the ACC occluded before the treatment (shaded area).

ACC: anterior condylar confluence IPS: inferior petrosal sinus

Venous circulatory failure in the semicircular canals was gradually improved by venous return into sigmoid sinus within one month, causing the vertigo/dizziness to disappear. However, improving the venous circulatory failure in the cochlea may not improve the hearing disturbance. Another possibility is suggested by the fact that the vein of the cochlear aqueduct drains into the ACC. Thus, occlusion of the IPS may result in venous hypertension of the ACC, which then causes a retrograde shunt flow from the ACC into the vein of the cochlear aqueduct. This venous hypertension of the cochlear aqueduct might be the cause of hearing disturbance even though the residual AVF was minimal immediately after the treatment. In addition, difference in recovery of individual neurons after damage may determine whether the neuropathy is temporary or permanent. Therefore the site of ACC dural AVF occlusion should be the shunting point (i.e., the ACC in this case) but not the draining vein (i.e., the IPS in this case).

Dysphagia and dysarthria (POD6) caused by hypoglossal nerve palsy could be due to thrombotic occlusion of the anterior condylar vein (ACV) at the hypoglossal canal. It was speculated that the mechanism underlying delayed hypoglossal nerve palsy is inflammation in the hypoglossal nerve and the surrounding thrombosed ACV. This phenomenon was similar to the one that occurs in abducens and/or oculomotor nerve palsy after treatment of the ICA (C4) giant aneurysm by ICA proximal occlusion or intraluminal occlusion²³). Another explanation might be occlusion at an inadequate site (e.g., the IPS) causing worsening of venous hypertension of the ACV, leading to hypoglossal nerve palsy. In our case, hypoglossal nerve palsy improved completely within 1 month with steroid treatment.

We emphasize here that TVE of dural AVF should be performed by occlusion of the fistula or drainage site (vein) involving the fistula, and not by occlusion of the drainage site (vein) alone. Placement of coils in an inadequate site may cause worsening of venous hypertension and may lead to an unfavorable outcome⁹). The IPS, especially, should not be occluded because it

forms a tract of venous circulation between the cochlea and vestibular apparatus. Understanding the venous circulation of the cochlea and vestibular apparatus is mandatory for safe treatment of ACC dural AVF.

Conclusion

A case of ACC dural AVF with ocular symptoms was treated by transvenous occlusion of the ipsilateral IPS, the only drainage tract, but not by occlusion of the fistula of the ACC. Consequently, though the ocular symptoms immediately improved, hearing disturbance, vertigo/dizziness, and dysphagia/dysarthria occurred because of venous circulatory failure, and ipsilateral hearing disturbance persisted. It is extremely important to occlude the fistula itself by TVE of the dural AVF.

The authors declare that they have no conflict of interest.

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