Transvenous embolization via the facial vein for intraorbital dural arteriovenous fistula: illustrative case

Jumpei Ienaga, MD,1 Tetsuya Tsukada, MD, PhD,1 Toru Watanabe, MD,1 Yosuke Sakai, MD,1 Kenji Uda, MD, PhD,2 Kazunori Shintai, MD, PhD,1 Yoshio Araki, MD, PhD,1,2 Tetsuya Nagatani, MD, PhD,1 and Yukio Seki, MD, PhD1

1Department of Neurosurgery, Japanese Red Cross Aichi Medical Center Nagoya Daini Hospital, Aichi, Japan; and 2Department of Neurosurgery, Nagoya University Graduate School of Medicine, Aichi, Japan

BACKGROUND Intraorbital arteriovenous fistula (IOAVF) is a rare type of intracranial fistula that presents with ocular signs similar to those of cavernous sinus dural arteriovenous fistula. The treatment of IOAVF is based on the vascular architecture of each case due to its infrequent occurrence. The authors report the case of an IOAVF that was successfully treated with embolization via the facial vein, with good outcomes.

OBSERVATIONS A 78-year-old woman presented with left eyelid swelling, pulsatile ocular protrusion, and left ocular conjunctival hyperemia. Ophthalmological evaluation revealed elevated intraocular pressure; time-of-flight magnetic resonance angiography revealed a dilated left superior ophthalmic vein. Digital subtraction angiography showed an arteriovenous shunt in the left superior orbital fissure, which was treated using transvenous coil embolization. The patient experienced immediate improvement in left ocular protrusion and conjunctival hyperemia. Ophthalmological evaluation 1 month after treatment showed normal intraocular pressure in the left eye. No neurological symptoms were observed, and there was no recurrence of the arteriovenous shunt 3 months postoperatively.

LESSONS The authors report a rare case of IOAVF treated with embolization via the facial vein with a good outcome. A thorough understanding of the vascular architecture using three-dimensional images is useful for determining endovascular access and procedures.

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KEYWORDS arteriovenous shunt; embolization; facial vein

Dural arteriovenous fistulas (dAVFs) account for 10%–15% of intracranial vascular malformations. They can occur in any part of the dura mater; however, they are most common in the cavernous sinus (CS) and transverse-sigmoid sinus regions. Intraorbital arteriovenous fistulas (IOAVFs) are a rare type of intracranial fistula that often present with ocular signs similar to those of CS dAVFs.1 Spontaneous IOAVF is found mostly in the aged population (mean: 61 yrs old), with a male predominance.2 It is associated with a relatively high risk of visual deterioration (33%)2 on the affected side, and the choice of treatment depends on the angioarchitecture of the fistula in each case. Herein, we report the case of a dural IOAVF treated with embolization via the facial vein, which showed a good outcome.

Illustrative Case A 78-year-old woman presented with a 1-month history of left eyelid swelling, pulsatile ocular protrusion, and conjunctival hyperemia (Fig. 1A). The patient had no history of trauma. Ophthalmological evaluation revealed elevated intraocular pressure (IOP; 15 mm Hg in the right eye and 27 mm Hg in the left eye), but there was no disturbance in visual acuity. Time-of-flight magnetic resonance angiography revealed a dilated left superior ophthalmic vein (SOV; Fig. 1B). A left internal carotid angiogram revealed a small dAVF at the posterior end of the opacified SOV (on the superior orbital fissure) supplied by a branch of the meningohipophyseal trunk. A left external carotid angiogram revealed another fistula point at the middle of the dilated SOV (within the orbit) supplied by the meningoacralveal artery.
branching from the middle meningeal artery. The venous phase of a left internal carotid angiogram showed that shunt flow in the SOV drained only into the facial vein and that normal cerebral venous blood flow to the inferior petrosal sinus through the CS was preserved (Fig. 2). Multiplanar reconstruction (MPR) images of the left internal carotid angiogram showed separation of the posterior end of the SOV from the superficial middle cerebral vein by some intervening tissue at the anterior part of the CS. A three-dimensional (3D) image of the external carotid angiography revealed two shunt points: (1) the anterior fistula point was fed by the meningolacrimal artery through the cranio-orbital foramen and the recurrent meningeal artery through the superior orbital fissure and (2) the posterior fistula point was fed by the accessory meningeal artery (Fig. 3). The 3D configuration image of contrast-enhanced computed tomography (CT) showed that the facial vein was not connected to the internal jugular vein but to the external and anterior jugular veins via the retromandibular vein (Fig. 4).

Considering the possibility of visual disturbances in the left eye due to elevated IOP, embolization of the arteriovenous shunt was indicated. Transvenous coil embolization of the involved SOV via the facial vein was planned. With the patient under general anesthesia, a 6-Fr Fubuki sheath (Asahi Intecc Co., Ltd.) was introduced into the left external jugular vein through the right femoral vein because CT findings indicated that the external jugular vein had lower flexibility than the anterior jugular vein. Afterward, a 130-cm 3.4-Fr Tactics (Technocrat Corp.) was placed into the facial vein as a distal access catheter (DAC). The left SOV was approached using a Headway Duo microcatheter (Terumo) and an Asahi Chikai guidewire (Asahi Intecc Co., Ltd.). The shunt points and SOV were embolized using 8 coils (Fig. 5A). Complete occlusion of the shunt was confirmed on digital subtraction angiography (DSA; Fig. 5B and C), and normal perfusion of the left eye was demonstrated by a retinal brush without any delay on left internal carotid angiogram. Venous return through the left CS was unaffected by the procedure (Fig. 5D).

The patient’s left ocular protrusion and conjunctival hyperemia began to improve immediately after embolization, and ophthalmological evaluation 1 month later showed normalization of the left IOP. The patient had no neurological symptoms, and no recurrence of the arteriovenous shunt was observed 2 years postoperatively.

**Patient Informed Consent**

The necessary patient informed consent was obtained in this study.
Lv et al.\textsuperscript{2} reviewed case reports and literature reviews on the rarity of IOAVF and the evolution of its treatment. They reported 26 IOAVFs, 19 of which drained into the SOV or inferior ophthalmic vein.\textsuperscript{1–16} Furthermore, eight of the reported cases were specified as having no connection between the CS and draining vein.\textsuperscript{2,6,8,14,15,16} Thus, it can be deduced that IOAVF is a rare clinical condition.

With regard to the anatomical isolation of this condition, Yamamoto et al.,\textsuperscript{17} in their case of IOAVF, considered that thrombus formation between the SOV and CS may have resulted in the intraorbital isolation of the drainage route. In the present case, MPR images from the left internal carotid angiogram showed some intervening tissue separating the SOV from the superficial middle cerebral vein at the anterior CS. Such an anatomical variation may have contributed to the isolation of shunted flow in the SOV from the CS in addition to the possibility of thrombus formation in the posterior end of the SOV.

Recently, transvenous embolization (TVE) has become the mainstay of IOAVF treatment.\textsuperscript{2} In the present case, the fistula was embolized using the transfemoral venous approach via the facial vein. Other possible methods for accessing the shunt point include reaching it from the inferior petrosal sinus via the CS and directly puncturing the SOV. Our case was characterized by the separation of the SOV from the shunt point and CS and percutaneous transorbital puncture of the SOV. In addition, our case was characterized by isolation of the drainer (SOV) from the CS, in which normal venous flow had been preserved. A potential risk of the trans-CS approach is that the access route through the thrombosed zone of the SOV may create a new draining route of the fistula to the CS, making additional embolization necessary. Direct catheterization of the surgically exposed SOV could skip over tortuous venous structures to the orbit; however, it is associated with the difficulty of exposing a small-caliber SOV. Percutaneous transorbital puncture of the SOV would be the last choice when other approaches are impossible because it is invasive and accompanied by potential complications, such as eyeball damage, intraorbital nerve injury, retrobulbar hematoma, internal carotid artery injury, and infection.\textsuperscript{18} Transarterial embolization (TAE) using liquid embolic agents is an effective treatment; however, it can cause visual impairment and external ophthalmoplegia in IOAVF.\textsuperscript{5} Therefore, TAE is considered an option when TVE is unsuccessful.\textsuperscript{19} In the present case, a less invasive transfemoral venous approach via the facial vein was selected, wherein a DAC improved the stability and handling of a microcatheter in the tortuous venous architecture. The combination of 3D DSA in the arterial phase in the external carotid artery and MPR image of the internal carotid angiography in the venous phase allowed for a detailed understanding of the vascular architecture. By understanding the detailed anatomical structures and accurately selecting the fistula point using a DAC, embolization was performed without using...
liquid embolic agents, which eliminated the risk of neurological complications.

Lessons
We reported a rare case of IOAVF treated with embolization via the facial vein, which showed a good outcome. A thorough understanding of the angioarchitecture of the fistula with MPR images helps in determining endovascular access and procedures.

References

Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions
Conception and design: Tsukada, Ienaga. Acquisition of data: Tsukada, Ienaga, Sakai, Uda. Analysis and interpretation of data: Ienaga, Araki. Drafting of the article: Ienaga, Nagatani, Seki. Critically revising the article: Tsukada, Ienaga, Seki. Reviewed submitted version of the manuscript: Tsukada, Watanabe, Nagatani. Approved the final version of the manuscript on behalf of all authors: Tsukada. Study supervision: Tsukada, Shintai.

Correspondence
Tetsuya Tsukada: Japanese Red Cross Aichi Medical Center Nagoya Daini Hospital, Aichi, Japan. tetsuya7.3wake@gmail.com.