A Patient with a Cavernous Sinus Dural Arteriovenous Fistula in Whom Direct Puncture of the Superior Ophthalmic Vein Led to Rapidly Progressing Thrombosis and Postoperative Non-arteritic Ischemic Optic Neuropathy: Pathogenesis with Respect to a Drainage Route

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Objective: We report a patient in whom direct puncture of the superior ophthalmic vein for a cavernous sinus dural arteriovenous fistula led to rapidly progressing thrombosis and postoperative non-arteritic ischemic optic neuropathy (NA-ION), and review the pathogenesis.

Case Presentation: A 74-year-old female. Detailed examination of diplopia and visual disorder suggested a cavernous sinus dural arteriovenous fistula. As approaching via a posterior route was difficult, transvenous embolization by direct puncture of the superior ophthalmic vein was performed. As drainage routes were aggregated around this vein, thrombosis of this vein occurred, inducing postoperative NA-ION through a rapid change in hemodynamics.

Conclusion: When performing direct puncture of the superior ophthalmic vein, puncture methods and heparinization should be considered after sufficiently investigating drainage routes.

Keywords ▶ cavernous sinus dural arteriovenous fistula, superior ocular vein direct puncture, ischemic optic neuropathy, thrombosis

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Introduction

For endovascular treatment for cavernous sinus dural arteriovenous fistulas (CSdAVFs), coil packing of a shunt site or sinus where a shunt is present via a transvenous approach is routinely performed. A catheter is inserted into the cavernous sinus (CS) through the inferior petrosal sinus (IPS) using a transfemoral approach. When approaching is difficult, a catheter is sometimes inserted through the superior ophthalmic vein (SOV) via the superior petrosal sinus (SPS), facial vein (FV), or middle temporal vein. If approaching is impossible using these routes, direct puncture of the SOV or Sylvian vein under craniotomy must be considered.^{1,2} Concerning heparinization at the time of SOV puncture, many studies reported hemorrhagic complications, and systemic heparinization is frequently avoided. In the present case, we also performed CSdAVF

Fig. 1 Right common carotid angiograms (A) anteroposterior view, (B) lateral view revealing a right CSdAVF fed by MHTs, artery of foramen rotundum and right MMA, and draining into bilateral SOV. (C and D) Preoperative time-of-flight magnetic resonance angiography. CSdAVF: cavernous sinus dural arteriovenous fistula; MHT: meningohypophyseal trunk; MMA: middle meningeal artery; SOV: superior ophthalmic vein

treatment by direct puncture of the SOV in the absence of systemic heparinization, but rapidly progressing thrombosis of the SOV occurred during treatment. We considered that puncture methods, as well as the necessity of heparinization, should be reviewed. If rapidly progressing thrombosis occurs during treatment, it may involve the orbital level, leading to blindness. In the present case, visual disorder deteriorated after surgery, but thrombosis did not involve the retinal level. As the etiology of visual disorder, thrombosis was not a direct etiological factor, but a rapid change in hemodynamics may have induced non-arteritic ischemic optic neuropathy (NA-ION) based on the results of the ophthalmological examination.³⁾ When performing direct puncture of the SOV, where drainage routes are aggregated, mechanically or physically induced return disorder may rapidly influence hemodynamics, resulting in thrombosis. Even if thrombosis does not involve the retinal level, optic neuropathy may occur, as demonstrated in the present case. To prevent such a complication, puncture methods, as well as the necessity of heparinization, should be discussed prior to surgery. For direct puncture of the SOV, the pretreatment assessment of drainage routes may reduce thrombosis or complications although this procedure is not frequently adopted.

Case Presentation

Patient: A 74-year-old female.

Complaints: Diplopia, low vision.

Medical history: Hypertension.

Family history: Not contributory.

Present illness: She consulted a local ophthalmological clinic with the above complaints. An eye drop preparation was prescribed, but the symptoms gradually exacerbated.

Activities of daily living (ADL) reduced, requiring assistance. In another hospital, cephalic MRI was performed, suggesting a CSdAVF. For detailed examination and treatment, she was referred to our hospital.

Findings: Consciousness was clear. The visual acuities of the bilateral eyes were 0.1 (corrected visual acuity: 0.1 on the left and right sides). Conjunctival hyperemia was noted. Ocular movement was restricted in all directions. In particular, bilateral abduction limits were marked. Cerebral angiography showed that a shunt pouch had converged on the medial side of the right CS, with the bilateral meningohypophyseal trunks (MHTs), bilateral arteries of the foramen rotundum, right middle meningeal artery (MMA), and left accessory meningeal artery (AMA) as feeders. An outflow tract from the left CS to the left SOV via the right SOV and intercavernous sinus was observed. Based on these findings, a diagnosis of a Barrow type D, Cognard type IIa CSdAVF was made (**Fig. 1**).

Course: Under general anesthesia, treatment was performed through the right internal jugular vein via the femoral vein. Complete occlusion of the IPS and SPS was observed, and various catheters and wires were inserted, but recanalization was not achieved (**Fig. 2**). Furthermore, the FV was considered as an approach, but it could not be selected due to occlusion of its periphery (**Fig. 3**). Occlusion of the left sigmoid sinus or deeper was noted, and an approach from the left side was abandoned. Therefore, the strategy was switched to flow reduction, and transarterial embolization (TAE) of the right MMA and left AMA was performed. There was a slight reduction in shunt flow, but this did not lead to radical cure.

There was no reduction of neurological symptoms, suggesting the necessity of additional treatment. We considered treatment via an intracranial vein under craniotomy or

Fig. 2 (A) The venous phase of right common carotid angiography. (B) Catheter venography for the assessment of internal jugular veins.

Fig. 3 (A and B) Facial vein was occluded at the distal portion (arrowheads).

through the FV or SOV by direct puncture. However, our hospital had no hybrid room, which may affect the safety, and the angular vein was markedly tortuous; therefore, we selected treatment by direct puncture of the SOV, which may facilitate the selection of an access route. Under general anesthesia, a 5 Fr diagnostic catheter was inserted into the right common carotid artery. After preparing a 3D road map, direct puncture of the SOV was performed. Regarding a supraorbital incisure as a benchmark, cutting-down was not initially conducted, and a 20G elaster needle was percutaneously punctured. Blood reflux was noted, suggesting that SOV puncture in the superficial layer was successful. However, subsequently, reflux disappeared, making microcatheter or -wire insertion difficult. A skin incision measuring approximately 2 cm was established in a medial area below the eyebrow for direct puncture of the SOV under direct vision. Macroscopically, the SOV was dilated (**Fig. 4A**), and Doppler ultrasonography showed arterial

blood reflux (**Fig. 4B**). Additionally, puncture was conducted using a 20G elaster needle to confirm pulsatile reflux (**Fig. 4C**). An Excelsior SL-10 STR (Stryker, Kalamazoo, MI, USA) and CHIKAI black 0.014 (Asahi Intecc, Tokyo, Japan) were inserted so that they might reach the CS, but the microguidewire could not be smoothly inserted (**Fig. 4D**). Additional imaging through a diagnostic catheter revealed the residual shunt, but there was no visualization of the SOV as an outflow tract (**Fig. 4E**). The elaster needle was removed. On Doppler ultrasonography, there was no SOV blood flow, suggesting thrombosis (**Fig. 4F**). Heparinization was not performed, considering the risk of hemorrhagic complications. Although the SOV was punctured in the visible range several times, there was no reflux. The deep orbital SOV was punctured by exfoliating a deeper area. As blind operations were required for the deep SOV, puncture was conducted toward the orbital roof, and the SOV was punctured by

Fig. 4 (A) The image showing the over-swelling right SOV. (B) Doppler ultrasonography showed reversed flow. (C) A 20-gauge puncture needle was inserted into the SOV. (D) Plain X-ray lateral view films. (E) The peripheral part of the SOV could not be visualized (arrowheads), but the shunting point remained (arrow). (F) Doppler ultrasonography showed thrombosed SOV. (G) Plain X-ray films showing placed coils to the shunting point. (H) Right fundus photograph revealed normal results. (I) DWI shows a high signal intensity area and ADC image shows a decreased signal intensity area on the right optic nerve. ADC: apparent diffusion coefficient; DWI: diffusion-weighted image; SOV: superior ophthalmic vein

gradually turning a needle onto the bulbar side. Slight blood reflux was noted, and the Excelsior SL-10 STR and CHIKAI black 0.014 were again inserted through the deep SOV. At the SOV-CS junction, marked stenosis was observed, making insertion difficult. The microguidewire was switched to a GT wire 0.012 (Terumo Corporation, Tokyo, Japan), and it was possible to guide the microcatheter to the CS shunt site. A Target 360 Soft (Stryker) 6 mm × 20 cm, seven Target 360 Ultra 5 mm × 15 cm, one Target 360 Ultra 4 mm × 15 cm, four Target Helical Ultra 4 mm × 15 cm, and two Target Helical Ultra 3 mm × 10 cm were placed at the shunt site; embolization with a total of 15 coils was performed (**Fig. 4G**). The lesion disappeared, and this procedure was completed.

Postoperative course: The visual acuity of the right eye further deteriorated immediately after surgery, suggesting intraoperative-thrombosis-related occlusion of the central retinal vein. The patient consulted the Department

of Ophthalmology the day after surgery. There was an improvement in the visual acuity of the left eye from 0.1 to 0.2 (corrected visual acuity: 0.6), but that of the right eye had deteriorated, reaching the light perception level. There was an improvement in bilateral abducens nerve disorder, and slight right gaze was noted. Ocular motility disorder of the right eye persisted. However, funduscopy did not show any circulation disorder of the central retinal artery or vein (**Fig. 4H**). The ocular pressure was normal, leading to a diagnosis of circulation disorder at the optic nerve canal level. Furthermore, diffusion-weighted MRI showed an increase in the brightness of the right optic nerve, and there was a reduction on an apparent diffusion coefficient image (**Fig. 4I**). Based on these findings, a diagnosis of NA-ION was made. Furthermore, the lesion site was present at the optic nerve canal level, leading to a diagnosis of non-arteritic posterior ischemic optic neuropathy (NA-PION). Anticoagulant therapy, steroid therapy, and vitamin supplementation

Fig. 5 Cerebral angiogram after embolization (A) AP view, (B) lateral view showing complete obliteration of the CSdAVF. (C and D) Postoperative time-of-flight magnetic resonance angiography. AP: anterior-posterior; CSdAVF: cavernous sinus dural arteriovenous fistula

were temporarily performed from the day after treatment, but there was no reduction of visual disorder. Improvements in the visual acuity of the left eye and ocular motility disorder were achieved, and diplopia disappeared, facilitating assistance-free daily living. After confirming the absence of recurrence using cerebral angiography and MR angiography (**Fig. 5**), the patient was discharged.

Discussion

In CSdAVFs, occlusion of the IPS, as a posterior route, initially occurs after shunt outgrowth, followed by occlusion of the SOV and inferior ophthalmic vein (IOV) as anterior routes.^{4,5} In the phase of advanced posterior-route occlusion and marked anterior drainage involving the SOV, neurological symptoms, such as abducens nerve paralysis, appear in addition to three CCF signs. Occlusion of the outlet from the SOV to the FV induces high-pressure congestion of the ophthalmic veins, rapidly increasing the ocular pressure and causing visual disorder. In this phase, a shunt is present, but the outflow tract is occluded, rapidly promoting thrombosis and leading to the state of paradoxical worsening. After the appearance of thrombosis, spontaneous cure is achieved in some cases, but irreversible glaucoma or retinal blood flow disorder may occur, resulting in blindness or cerebral hemorrhage through intracranial reflux; therefore, emergency shunt occlusion is necessary.

In the present case, drainage route return disorder mechanically or physically occurred immediately after SOV puncture, suggesting thrombosis of the main drainage route (SOV) through a rapid change in hemodynamics. Two case reports in which direct puncture of the thrombosed SOV led to treatment have been published,^{6,7} but thrombosis occurred during the spontaneous course in the two patients. No patient with thrombosis during treatment has been

presented. Treatment by direct puncture of the SOV is not frequently selected, but it is adopted when only an anterior drainage route remains due to occlusion of all posterior routes. Satomi et al.⁴ classified CSdAVFs into three stages based on the grade of outflow venous tract occlusion, but we further subclassified the staging that they proposed (**Fig. 6**). When performing direct puncture of the SOV in the presence of posterior route occlusion, especially in stage 2A in which drainage routes are aggregated around the SOV, drainage route return disorder may mechanically or physically occur. It must also be recognized that thrombosis of a drainage route may make treatment difficult, inducing unexpected intra- or postoperative complications.

As SOV-puncture-related complications, puncture-related intra-orbital hemorrhage, ocular injury, optic nerve injury, oculomotor nerve injury, and internal carotid artery injury have been reported. For endovascular treatment, heparinization is routinely performed. However, according to some studies,^{8,9} systemic heparinization was not conducted in patients treated by direct puncture of the SOV, considering the risk of hemorrhagic complications. With respect to direct puncture of the SOV in the presence of aggregated drainage routes, as demonstrated in the present case, the risk of thrombosis may be high. We considered that systemic heparinization might reduce the risk of thrombosis to some degree. Furthermore, to reduce mechanical stimuli related to several sessions of puncture, we should have performed puncture while examining the SOV course under direct vision after establishing a skin incision.

In the present case, initially, thrombosis-related occlusion of the central retinal vein was suspected as an etiological factor for the post-treatment deterioration of optic neuropathy, but funduscopy showed that return at the retinal level was maintained. Based on funduscopy and MR findings, a diagnosis of NA-PION was made. ION

Fig. 6 The classification of venous drainage patterns. In Type 1, both anterior and posterior drainage routes are open; in Type 2, the posterior drainage route is closed, whereas the anterior drainage route is open (Type 2A: the venous drainage converges on SOV, Type 2B: the venous drainage does not converge on SOV); and in Type 3, both the posterior and anterior drainage routes are closed. IOV: inferior ophthalmic vein; IPS: inferior petrosal sinus; PP: pterygoid plexus; SOV: superior ophthalmic vein; SPS: superior petrosal sinus

is related to the acute circulatory disorder of optic nerve papilla feeders. It is classified into two types: arteritic ION (A-ION), which is related to angitis, and NA-ION, which is associated with arteriosclerosis or hemodynamic changes. Based on sites, it is classified into anterior ION (A-ION), which is characterized by papillary edema related to the disturbance at the retinal level, and posterior ION (PION), in which papillary edema is absent due to the disturbance at the optic nerve level.¹⁰⁾ For the diagnosis of PION, normal funduscopy findings and assessment using diffusion-weighted MR and diffusion coefficient images are useful.¹¹⁾ In the present case, funduscopy did not reveal any retinal arteriovenous disorder, and the ocular pressure was normal. In addition, there was an increase in the brightness of the right optic nerve on diffusion-weighted MRI, whereas there was a reduction on a diffusion coefficient image, leading to a diagnosis of NA-PION. Risk factors for NA-ION include hypertension, diabetes mellitus, dyslipidemia, and smoking, but its pathogenesis remains to be clarified. Previous studies reported the onset of NA-ION after carotid artery stenting,¹²⁾ after TAE of dAVFs,¹³⁾ or after aortic arch replacement,¹⁴⁾ suggesting its association with marked changes in hemodynamics. In the present case, a rapid change in venous return may have been closely involved in the onset of NA-ION. As treatment methods, steroids and anti-edema therapy have been selected, but no treatment method has been established.

Conclusion

We encountered a patient in whom rapidly progressing thrombosis immediately after SOV puncture influenced treatment, as well as the post-treatment course. Although the procedure of SOV puncture is not complex, it should be recognized that rapidly progressing thrombosis may occur, depending on the state of a drainage route. Prior to treatment, it may be necessary to evaluate drainage routes and examine the necessity of heparinization. If a skin incision is initially established before puncture, mechanical stimuli related to several sessions of SOV puncture may be reduced. It must always be considered that rapid changes in venous return may cause thrombosis of the central retinal vein and ocular lesions, such as NA-ION.

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Disclosure Statement

There is no conflict of interest regarding this article for the first author and coauthors.

References

- 1) Kazekawa K, Iko M, Sakamoto S, et al: Dural AVFs of the cavernous sinus: transvenous embolization using a direct superficial temporal vein approach. *Radiat Med* 2003; 21: 138–141.
- 2) Kuwayama N, Endo S, Kitabayashi M, et al: Surgical transvenous embolization of a cortically draining carotid cavernous fistula via a vein of the sylvian fissure. *AJNR Am J Neuroradiol* 1998; 19: 1329–1332.
- 3) Hashimoto M, Ohtsuka K, Suzuki Y, et al: A case of posterior ischemic optic neuropathy in a posterior-draining dural cavernous sinus fistula. *J Neuroophthalmol* 2005; 25: 176–179.
- 4) Satomi J, Satoh K, Matsubara S, et al: Angiographic changes in venous drainage of cavernous sinus dural arteriovenous fistulae after palliative transarterial embolization or observational management: a proposed stage classification. *Neurosurgery* 2005; 56: 494–502.
- 5) Suh DC, Lee JH, Kim SJ, et al: New concept in cavernous sinus dural arteriovenous fistula: correlation with presenting symptom and venous drainage patterns. *Stroke* 2005; 36: 1134–1139.
- 6) Wakui D, Terada T, Oshima K, et al: [A case of carotid cavernous fistula treated with a direct puncture to the thrombosed superior ophthalmic vein]. *JNET* 2013; 7: 345–350. (in Japanese)
- 7) Chen WH, Tsai IC, Huang HC, et al: Transcutaneous puncture of the superior ophthalmic vein for embolization of dural carotid-ophthalmic fistula. *Interv Neuroradiol* 2008; 14: 9–11.
- 8) Leibovitch I, Modjtahedi S, Duckwiler GR, et al: Lessons learned from difficult or unsuccessful cannulations of the superior ophthalmic vein in the treatment of cavernous sinus dural fistulas. *Ophthalmology* 2006; 113: 1220–1226.
- 9) Quiñones D, Duckwiler G, Gobin PY, et al: Embolization of dural cavernous fistulas via superior ophthalmic vein approach. *AJNR Am J Neuroradiol* 1997; 18: 921–928.
- 10) Hayreh SS: Ischemic optic neuropathy. *Prog Retin Eye Res* 2009; 28: 34–62.
- 11) Al-Shafai LS, Mikulis DJ: Diffusion MR imaging in a case of acute ischemic optic neuropathy. *AJNR Am J Neuroradiol* 2006; 27: 255–257.
- 12) Nanto M, Tanigawa S, Takado M, et al: [A case of ischemic optic neuropathy after carotid artery stenting]. *JNET* 2014; 8: 52–57. (in Japanese)
- 13) Yamamoto M, Yoshida K, Nonaka S, et al: [A case of ischemic optic neuropathy after transarterial embolizations for cavernous sinus dural arteriovenous fistula]. *JNET* 2013; 7: 18–23. (in Japanese)
- 14) Kanabuchi K, Yamaguchi M, Ueda T, et al: A case of ischemic optic neuropathy developed eleven days after an aortic arch replacement. *Tokai J Exp Clin Med* 2011; 36: 112–115.