Clinical Case Report

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Amelioration of acute orbital compartment syndrome following transvenous embolization for an indirect carotid-cavernous fistula

A case report

Yun-Hsiu Hsieh, MD, Tzu-Heng Weng, MD, Ming-Cheng Tai, MD, Ke-Hung Chien, MD*

Abstract

Rationale: Orbital compartment syndrome (OCS) is a rare occurrence after transvenous embolization of indirect carotid-cavernous fistula (CCF). A lateral canthotomy and cantholysis are the most commonly performed surgical interventions. In our case, as the acute OCS occurred immediately after an uneventful transvenous embolization, an orbital floor orbitectomy was performed.

Patient concerns: Here, we present a rare case of a 59-year-old patient who required a transvenous embolization of an indirect CCF and subsequently immediately developed an acute OCS.

Diagnoses: An indirect CCF was revealed using brain magnetic resonance angiography and a transvenous embolization of the fistula was performed using coils. Post-embolization angiograms revealed an occlusion of the CCF.

Interventions: After the development of a relative afferent pupillary defect and acute OCS, we performed a lateral canthotomy, superior and inferior cantholysis, and an orbital floor orbitectomy. Subsequently, visual acuity and intraocular pressure improved.

Lessons: Our case is the first report of acute OCS occurring after transvenous embolization of a CCF that required further orbital floor decompression to prevent permanent visual loss. Moreover, our case demonstrates that acute OCS may rapidly develop after transvenous embolization due to superior ophthalmic venous (SOV) thrombosis and that an early intervention may reduce the risk of visual impairment.

Abbreviations: BCVA = best-corrected visual acuity, CCF = carotid-cavernous fistula, ECA = external carotid arteries, ICA = internal carotid arteries, IPS = inferior petrosal sinus, OCS = orbital compartment syndrome, RAPD = relative afferent pupillary defect, SOV = superior ophthalmic venous.

Keywords: carotid cavernous fistula, orbital compartment syndrome, superior ophthalmic venous thrombosis, transvenous embolization

1. Introduction

An indirect carotid-cavernous fistula (CCF) is the abnormal communication between the meningeal branches of internal and/or external carotid arteries (ICA and/or ECA) and the cavernous sinus. Indirect CCF has a benign prognosis and may spontaneously resolve.[1] However, in some cases, intervention is required, and the first-line treatment is endovascular embolization. Here, we present a rare case of acute orbital compartment syndrome (OCS) development after transvenous embolization of an indirect CCF that required an orbital floor orbitectomy, which is the first reported case of such a condition. The patient had given consent for the use photographs for publication.

2. Case presentation

A 59-year-old man presented to our emergency room with a 3-month history of progressive swelling, tenderness, and redness of right orbital region (Fig. 1A). His best-corrected visual acuity (BCVA) of the right eye decreased from 6/8.6 to 6/20 on Snellen chart, and intraocular pressure increased from 18 to 27 mmHg. Brain magnetic resonance angiography revealed an indirect CCF, Barrow classification type D, on the right hemisphere. The patient subsequently received transvenous embolization of the fistula using coils by a radiologist. An intraoperative angiogram revealed multiple tiny arterial feeders from the ICA and ECA (Fig. 2A and B). Because the ipsilateral inferior petrosal sinus (IPS) was thrombosed, the cavernous sinus was accessed via pterygoid plexus. Matrix2 Helical UltraSoft SR Coils 5 mm × 15 cm (MicroPlex Helical 10 Coil 5 mm × 15 cm) were placed in the outlet of the right superior ophthalmic vein and right cavernous sinus. Post-embolization angiograms demonstrated occlusion of the CCF (Fig. 3). Immediately after the procedure, severe proptosis and conjunctival vascular congestion developed with a positive relative afferent pupillary defect (RAPD) of the right eye

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Department of Ophthalmology, Tri-Service General Hospital, Taipei City, Taiwan (R.O.C.).

** Correspondence: Ke-Hung Chien, Department of Ophthalmology, Tri-Service General Hospital, 2F., No. 325, Sec. 2, Chenggong Rd., Neihu Dist., Taipei City 114, Taiwan (R.O.C.) (e-mail: yred8530@gmail.com).*

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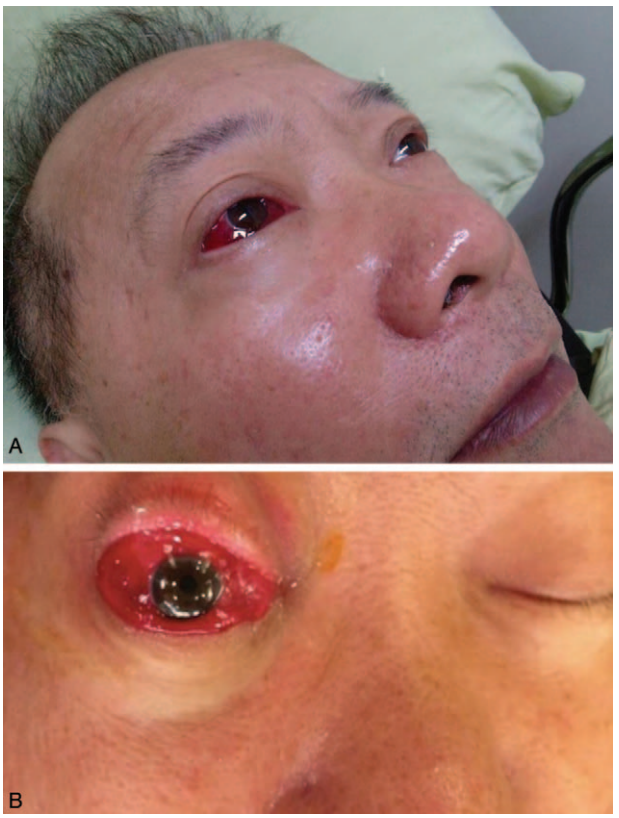

Figure 1. (A) Before transvenous embolization. Photograph of the patient before the procedure showing proptosis and subconjunctival hemorrhage of the right eye. (B) After transvenous embolzation. Photograph of the patient immediately after transvenous embolization showing severe chemosis, subconjunctival hemorrhage, and proptosis of the right eye.

(Fig. 1B). Within 30 minutes of the development of acute OCS, lateral canthotomy and inferior and superior cantholysis were performed. However, the RAPD persisted even after the procedure; thus, we performed an orbital floor orbitectomy. Subsequent brain magnetic resonance imaging revealed a dilated and hyperintense right superior ophthalmic venous (SOV) on T2 scan (Fig. 4), confirming the diagnosis of venous thrombosis. On postoperative day 1, the patient showed improved visual acuity, with normal global tactile tension and a sluggish light reflex of the right eye. Two weeks later, the BCVA of the right eye returned to 6/6 and intraocular pressure improved to 18 mmHg. There was no complication such as enophthalmos, hypoglobus, nor extraocular muscle movement limitation of the right eye 2 months later during postoperative follow up (Fig. 5).

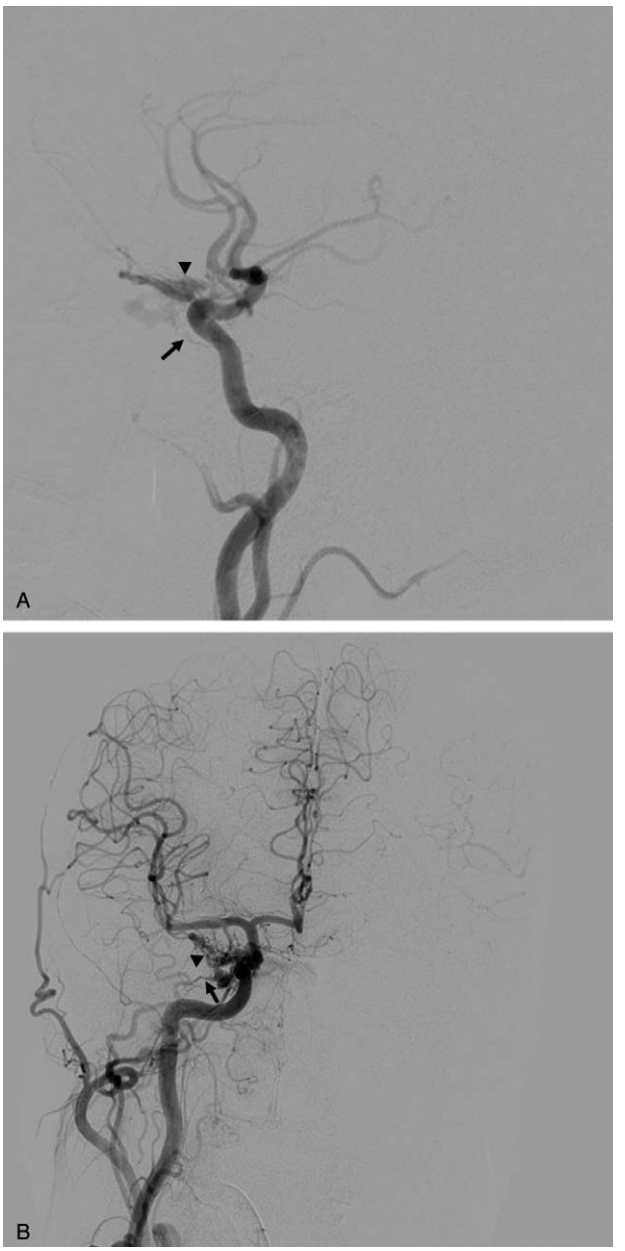

Figure 2. (A) Pre-embolization angiogram. Angiogram of the internal carotid artery. Lateral view of a right common carotid angiogram showing the right cavernous sinus (arrowhead) filled with tiny branches of the right internal carotid artery (arrow). (B) Pre-embolization angiogram. Anteroposterior view of a right common carotid angiogram showing the right cavernous sinus (arrowhead) filled with tiny branches of the left external carotid artery (arrow).

3. Discussion

OCS, a rare but devastating orbital complication, can develop due to a variety of causes, including trauma, neoplasms, infection, retrobulbar hemorrhage, and even transvascular embolization of a CCF, which is also associated with SOV thrombosis.[2,3] Transvenous embolization of a CCF is preferred because of the much higher rates of occlusion of the fistula, compared with transarterial embolization[4]; there is only one reported case of OCS following transvenous embolization of CCF.[2] For that case, OCS occurred 12 hours after embolization but immediately developed after embolization in our case. The occurrence of OCS within minutes in our case may have been due to placing the coil to the entrance of the SOV and complete occluding the cavernous sinus thus causing SOV thrombosis. The paradoxical worsening of signs result from SOV thrombosis may cause OCS, as suggested in a previous report.[2,5] We did not perform an angiogram to confirm this supposition because an imaging evaluation would have delayed the OCS intervention.[3] The natural history following SOV thrombosis as seen in previously reported cases is subsequent spontaneous resolution when collateral vessels develop to drainage the venous outflow.[2] The present case may serve as a reminder to radiologists and neurosurgeons that an OCS can progress rapidly and complete

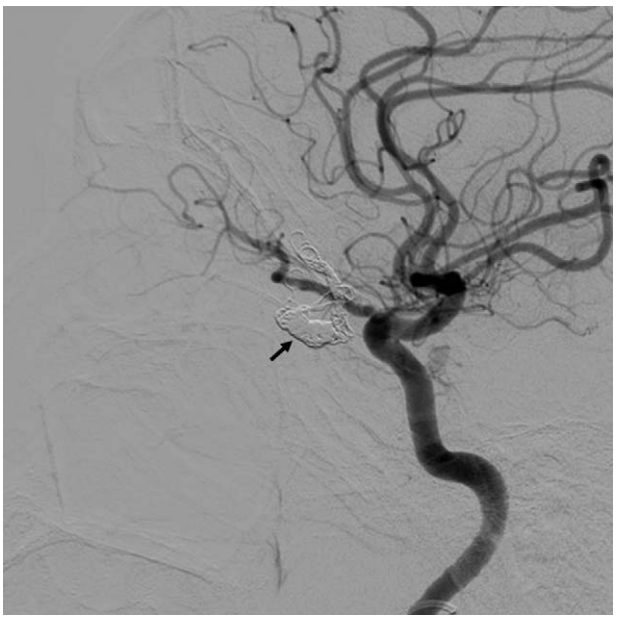

Figure 3. Post-embolization angiogram. Post-embolization angiogram showed the right internal carotid artery was patent and the fistula (arrow) was occluded.

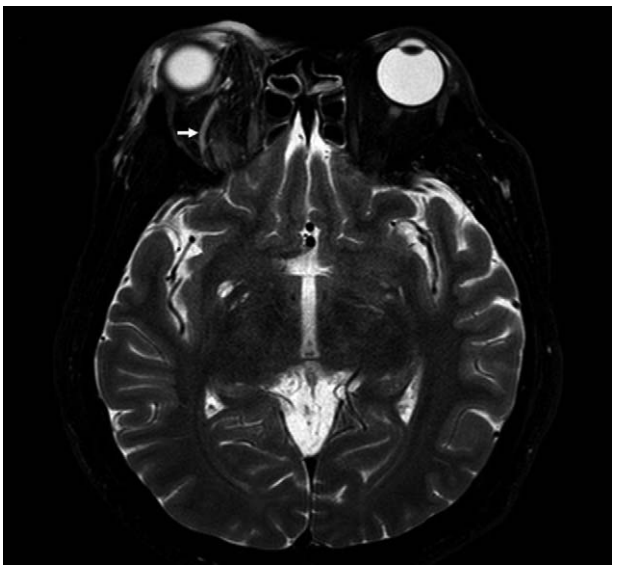

Figure 4. Magnetic resonance imaging. T2-weighted image showing dilated and hyperintense right superior ophthalmic venous (arrow).

cavernous sinus occlusion should be avoided as suggested in the previous report.[5]

A lateral canthotomy and cantholysis are the most commonly performed procedure for releasing the orbital pressure in OCS; however, orbital floor decompression surgery is suggested in more severe cases.[6] Our case of OCS required orbital floor orbitectomy surgery to completely release the "tight orbit" and RAPD sign; this condition has not been previously reported for transvenous embolization. An orbital floor orbitectomy is more invasive and time-consuming[6]; however, our case of OCS occurred immediately after the embolization while the patient was still in the operating room and under general anesthesia.

A limitation of our case is that we did not determine whether the orbital decompression surgery needed to be carried out within 30 minutes or if the CCF would have spontaneously resolved as is commonly observed. Ischemia for 30 minutes to 2 hours can induce optic nerve damage that results in OCS.[3,6] In our case, the acute OCS occurred immediately after an uneventful transvenous embolization for CCF, resulting in a poor prognosis due to the presence of the RAPD sign and clinical expression of a tight orbit. We believe that our urgent ophthalmology review and management successfully saved the vision of the patient. Although the best time for performing orbital decompression

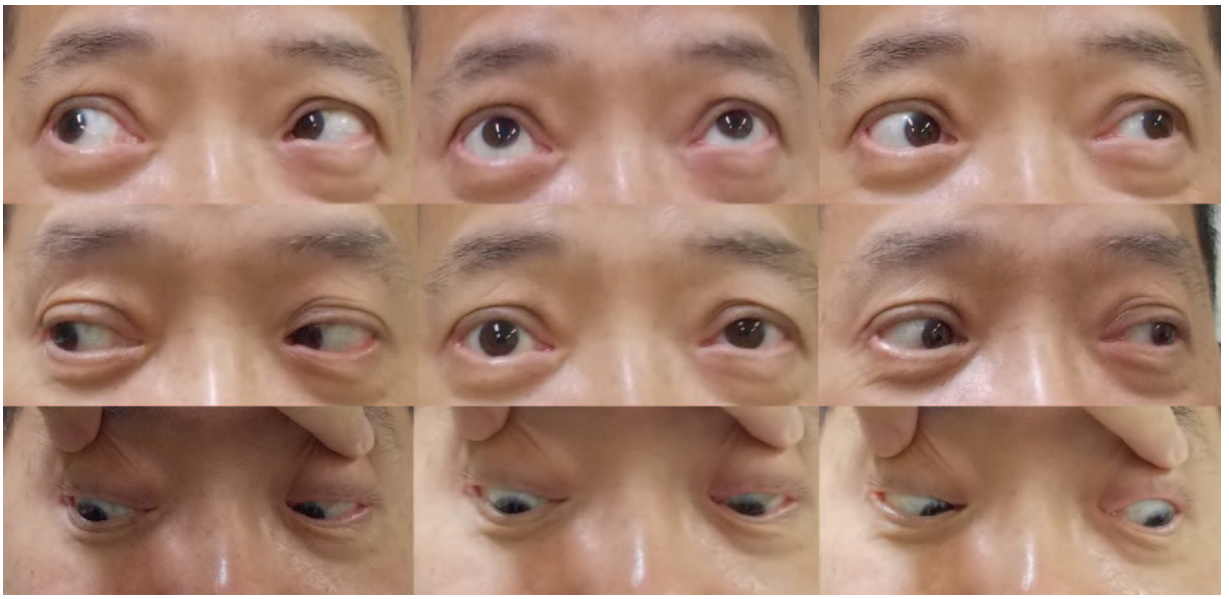

Figure 5. Post-orbitectomy. Photograph of the patient after orbital floor orbitectomy 2 months later showing orthotropia without enophthalmos, hypoglobus, nor extraocular muscle movement limitation of the right eye.

surgery remains controversial, our results suggest that a good outcome resslies on early treatment.

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