Case Report

Worsening of cavernous sinus dural arteriovenous fistula with incomplete superior ophthalmic thrombosis after palliative transarterial embolization

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Worsening of ocular symptoms in cavernous sinus dural arteriovenous fistulae (CSDAVF) is rarely due to superior ophthalmic vein (SOV) thrombosis after palliative transarterial embolization and may resolve spontaneously. We present a unique case of a 38-year-old female whose ocular symptoms worsened three days after palliative embolization. Repeat angiography revealed incomplete thrombus of SOV and severe orbital venous congestion. Complete occlusion of the CSDAVF was achieved by urgent transvenous embolization through the SOV and the patient’s ocular symptoms and visual impairment recovered. In such situations repeat angiogram may be needed in evaluating the subtle angiographic changes and curative embolization may be necessary.

**Key words:** Cavernous sinus, dural arteriovenous fistulae, superior ophthalmic vein

Dural arteriovenous fistulae (DAVF) are abnormal arteriovenous shunts developing in the dura mater, usually within or near the walls of a dural sinus and are most common in the cavernous and transverse or sigmoid sinuses. A variety of treatment strategies for aggressive cavernous sinus dural arteriovenous fistulae (CSDAVF) are offered in the literature, including transarterial and/or transvenous embolization, radiation therapy and surgery. Paradoxical worsening with subsequent spontaneous resolution, has been observed in patients after transarterial embolization. Gamma knife radiosurgery or conservative treatment due to complete thrombosis of the superior ophthalmic vein (SOV) may occur. We present a unique patient with worsening of ocular symptoms due to incomplete thrombosis of SOV after palliative transarterial embolization, in which urgent complete embolization as well as anticoagulation therapy may be necessary to avoid irreversible visual impairment.

A 38-year-old female presented with six months of progressive left proptosis with pain, chemosis and diplopia starting in the puerperium. She denied a history of cranial or orbital trauma. Two months before presentation to our hospital she was treated as infectious conjunctivitis in a local clinic, without improvement. Physical examination demonstrated left abducens nerve palsy, proptosis, chemosis and a clearly audible bruit on the left side.

Cerebral angiography revealed a left-sided CSDAVF fed by the internal carotid artery (ICA) and external carotid branches [Figures 1A-C]. Venous drainage was mainly retrograde through the enlarged ipsilateral SOV and facial vein. For treatment a transfemoral route via jugular vein, facial vein and SOV was tried, but the microcatheter could not be passed through the tortuous facial vein and angular vein. A SOV approach by direct percutaneous puncture or surgical exposure was refused by the patient. Accordingly, transarterial embolization was planned with the aim of symptom palliation. Under local anesthesia a microcatheter was superselectively navigated into the feeders arising from the left external carotid artery (ECA) via the middle meningeal artery and ascending pharyngeal artery and we injected 20% N-butyl-2-cyanoacrylate (NBCA) mixed with ethiodol (Lipiodol) for embolization. Transarterial catheterization for coil placement from the left ICA was not successful. Immediate postembolization angiograms demonstrated occlusion of the ECA feeders, while the fistula supplied from the left ICA remained [Figure 1D]. Conservative management including manual left carotid compression was continued following this endovascular procedure.

No obvious change in the patient’s ocular symptoms was observed after embolization, but three days later she developed worsening left-sided proptosis.
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Figure 1: Cerebral angiograms before and immediately after the first embolization. A. Lateral left common carotid artery (CCA) injection before embolization showing opacification of the left cavernous sinus (thin arrow) with drainage to dilated superior ophthalmic vein (SOV) (thick arrow) and facial vein (arrowheads). Note that the cavernous sinus was partially thrombosed and the SOV was the predominant drainage. B. Lateral left internal carotid artery (ICA) injection showing a small fistula from the ICA. C. Lateral left external carotid artery (ECA) injection before embolization, revealing a CSDAVF with predominant venous drainage toward the SOV and facial vein. D. Lateral left ECA view immediately after superselective transarterial feeder embolization, showing obliteration of the fistula supplied by the ECA.

Figure 2: Cerebral angiograms before and immediately after the second embolization. A. Lateral left ICA injection before embolization showing incomplete thrombosis in the distal superior ophthalmic vein (arrow) and no facial vein opacification. B. Lateral venous phase of the left ICA angiogram showing obvious slowing of the ocular and orbital venous outflow. C. Lateral view of the left ICA injection showing position of coils within the cavernous sinus through the microcatheter introduced from the SOV. D. Lateral view of immediate left ICA injection after embolization showing complete disappearance of the shunt.

Discussion

Superior ophthalmic vein thrombosis with paradoxical worsening of symptoms is rare after transarterial embolization of CSDAVF.

In one study it was thought that the thrombogenic Yvalon had reached the feeders to the SOV, causing complete SOV thrombosis, confirmed by color Doppler imaging. The ocular symptoms of that patient improved with conservative management.

Paradoxical worsening with SOV thrombosis was also reported in a patient with CSDAVF one month after gamma knife radiosurgery, with increasing proptosis and visual deterioration in the involved eye. Magnetic resonance scanning showed absence of a flow void in the SOV and soft tissue signal in its lumen, indicating thrombus formation. The patient's symptoms improved within one month with conservative treatment and MR angiography revealed complete obliteration of the fistula. The mechanism of worsening of symptoms and

and chemosis. Urgent cerebral angiography revealed thrombus formation in the distal left SOV and loss of the drainage via the facial vein [Figures 2A, B]. There was severe congestion with slow flow around the orbital area. Therefore, further treatment via an SOV approach after operative exposure was chosen. Via a 2cm incision in the upper eyelid the thrombosed distal SOV was exposed. An 18-gauge cannula was used to puncture the SOV through the thrombosed part and an Excel-14 microcatheter (Target Therapeutics/Boston Scientific, Watertown, MA) was inserted, easily reaching the anterior portion of the cavernous sinus. Three GDCs (Boston Scientific, Fremont, CA) were then delivered through the microcatheter for embolization [Figures 2C, D]. At the end of the intervention total occlusion of the fistula was achieved. The patient’s painful proptosis, chemosis and diplopia resolved rapidly and her vision improved gradually. No symptoms to suggest recurrence developed during clinical follow-up of 18 months.
signs before the onset of clinical improvement was thought to be due to SOV thrombosis and extension of thrombus from the SOV to dural shunts.

However, irreversible visual impairment may also result from changes in venous drainage after palliative transarterial embolization. Satomi et al.\cite{7} reported a patient with a left CSDAVF draining into the ipsilateral inferior petrosal sinus and SOV who developed worsening conjunctival chemosis and visual acuity impairment two months after palliative embolization. Repeat angiography demonstrated occlusion of the left inferior petrosal sinus and stagnation in the SOV. Although closure of the CSDAVF was confirmed the patient continued to be visually impaired.

Where treatment is concerned, DAVF with cortical venous drainage must be completely obliterated because of the high risk of hemorrhage or focal neurological deficits. However, deterioration of ocular symptoms following palliative embolization or after stereotactic radiosurgery may be transient and is not an absolute indication for urgent and complete obliteration of the fistula.\cite{4-7} In the present case, the distal portion of the left SOV as the only venous outflow was occluded for incomplete partial thrombosis and serious orbital congestion was not only due to drainage obstruction but also due to the high-pressured orbital venous reflux. Because of the severity, it was justified to obtain complete occlusion of the CSDAVF by transvenous embolization through the SOV and the patient's ocular symptoms and visual impairment recovered.

Our case is thus unique for its partial SOV thrombosis and severe orbital congestion with risk of irreversible visual loss due to high intraocular pressure. Noninvasive MRI and Doppler imaging are widely used in the radiological follow-up of CSDAVF, but detailed hemodynamic changes including orbital congestion may then go undetected.\cite{5-8} The observation of this case suggests that in worsening of CSDAVF, repeated angiogram may be needed to evaluate subtle angiographic changes and curative embolization should be considered even though the combination of worsening signs and evidence of thrombosis may indicate impending resolution of a cavernous-dural shunt.

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References


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