

Case Report

Direct Puncture Preoperative Embolization for Cavernous Venous Malformation of the Orbit

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Abstract

Orbital cavernous venous malformation -previously called hemangioma-, is a common adult orbital space occupying lesion. Large symptomatic lesions may warrant surgical removal. Intraoperative bleeding however may extend the intraoperative time and prevent complete excision. Pre-operative embolization of the external carotid artery supply may be of help. However, some lesions have significant ophthalmic artery supply. In this particular scenario, surgical exposure of the lesion and direct puncture followed by liquid embolic injection have been described. We report a case of fluoroscopic guided direct puncture of the malformation and almost complete obliteration with n-butyl cyanoacrylate. Direct puncture embolization allowed a rapid surgical removal of the malformation with minimal blood loss.

INTRODUCTION

Cavernous venous malformation (CVM), previously known as orbital cavernous haemangioma, is the most common orbital vascular lesion in adults [1,2]. It is classified as type 3 low-flow arteriovenous malformation [3]. CVM can remain asymptomatic for a long time or present with progressive painless proptosis. Other presentations include diplopia, ocular pain, lid swelling and optic nerve damage [4,5]. Usually, CVM can be treated successfully with complete surgical excision [6]. Embolization before surgery to reduce the lesion never evolved to a standard procedure [7]. We present a case of orbital CVM which was successfully treated with Direct Puncture Preoperative Embolization.

CASE REPORT

A 39 years old male patient complaining of right eye protrusion and horizontal diplopia was admitted to our service (Figure 1A and B). The exophthalmometry was: RE-LE= 8 mm, and visual acuity: 40/20 (RE), and 20/20 (LE). A contrast-enhanced computerized tomography scan showed a densely hypervascular extraconal mass laterally located in the right orbit. The mass measured 5.2, 4.1 and 2,2 cm in its anteroposterior, height and width respectively, and had a multilobulated aspect without a clear capsule (Figure 2A). The 3D reformatted contrast enhanced CT angiography demonstrated a cluster of dilated dysplastic vessels with pseudo aneurysms, anticipating potentially significant intraoperative blood loss (Figure 1C). The MR imaging unveiled small serpiginous flow voids at the margins of the lesion (not shown). The patient was scheduled for embolization prior

to surgical excision. After informed consent, the patient was given general anesthesia and a 5 French introducer sheath 10 cm long was inserted in the right femoral artery. A 5 French guiding catheter 100 cm long was placed proximally in the trunk of the right external carotid artery assisted by a 0,035" hydrophilic guidewire 150 cm long. By injecting isoosmolar nonionic iodine contrast media, angiographic runs in anteroposterior and lateral projections were taken which showed a right intraorbital low flow hypervascular polilobulated lesion fed by external carotid arterial branches (angular, zygomatic-orbital and sphenopalatines) and the ophthalmic artery, branch of the internal carotid artery (Figure 2B). Then, a 2,1 French tipped microcatheter with a 0.014" microguidewire was navigated distally in each of the external carotid artery branches. After a microangiography demonstrating the blush of the lesion and absence of dangerous anastomoses, 250 – 300 microns polyvinyl alcohol – PVA - particles were injected superselectively, obtaining complete devascularization of the external arterial feeders (not shown). A common carotid artery angiogram showed that 50% of the lesion blush remained due to the ophthalmic artery supply (Figure 2C). Then, the right eye was washed with saline solution while the periocular skin and antisepsis was administered to the visible portion of the lesion with betadine solution and the tumor was punctured with a 1,5-inch-long, 21 G -Gauge- hypodermic needle. After obtaining venous blood return, contrast media was injected directly, verifying that the needle tip was in the vascular lumen of the lesion and ruling out retrograde flow to the ophthalmic artery and an anterograde flow towards the



Figure 1 Patient's pictures: (a) and (b) Before CVM surgical excision. Notice right exophthalmos, palpebral distension, chemosis, conjunctival hyperemia. (c) A 3-D reformatted contrasted CT angiography. The lateral orbit wall has been digitally removed. Noticed a large tangle of abnormal vessels with ectatic dilatations. (d) Direct puncture of the CVM and injection of the embolic mixture: n-bca and lipiodol at 50%. (e) and (f) Three months after surgery the exophthalmos and the redundant palpebral skin gradually decrease and conjunctival hyperemia is gone.

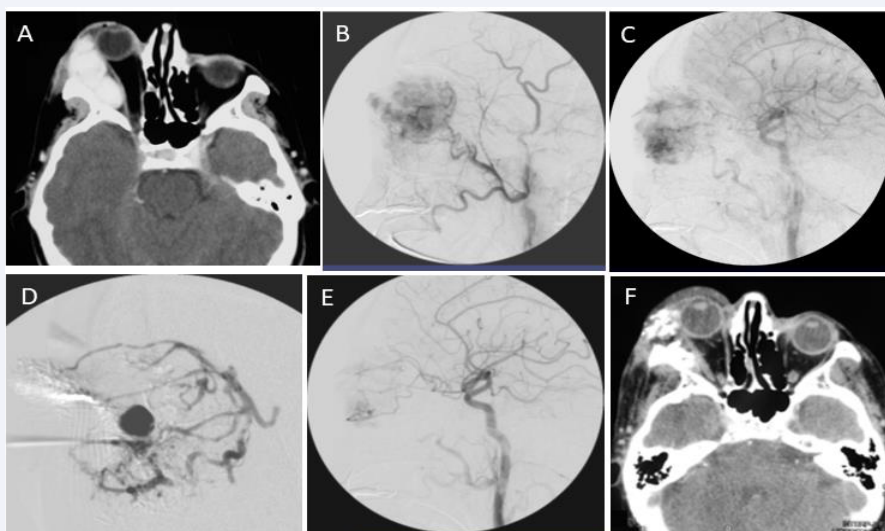


Figure 2 (a) Axial cranial contrast-enhanced CT scan, axial. Notice a lesion located laterally in the extraconal space in the right orbit with bright enhancement showing a multilobulated pattern. (b) External carotid artery. Notice a round blush with multilobulated pattern and delayed venous drainage (c) Common carotid artery angiogram after complete external carotid artery supply embolization with polyvinyl alcohol particles of 250-300 μ. CVM blush remnant was due to ophthalmic artery supply. (d) Second and last direct puncture with gentle injection of n-bca. The injection was stopped as soon as retrograde flow of the embolic mixture to the main terminal branches of the ophthalmic artery was adverted. (e) Common carotid angiogram shows ≥ 95% embolization. (f) Common carotid angiogram shows ≥ 95% embolization.

ophthalmic veins, the needle lumen was flushed with normal saline and purged with 2 cc of 5% dextrose in water. Then, a total amount of 0,8 cc of an embolic mixture of 50% n-butyl cyanoacrylate - n-bca - in lipiodol was injected in the lesion in two different injections (Figure 1D and 2D). Post-embolization common carotid artery angiogram showed almost complete obliteration of the hemangioma blush (Figure 2E). The needle and catheters were removed and manual hemostasis of both puncture sites: in the lesion and in the right femoral artery were done. Then, the CVM was surgically removed by an anterior orbitotomy via incision of the skin without damage to the normal orbital structures. The intraoperative blood loss was 50 cc.

Postoperatively, the surgical wound healed uneventfully and the patient discharged. At 2 months of follow up proptosis had rapidly regressed and the diplopia disappeared (Figure 1E and F). The exophthalmometry was RE-LE= 3 mms. The visual acuity was 24/20 (RE), and 20/20 (LE). A follow-up contrast-enhanced CT scan showed complete lesion removal with a small superficial remain of embolic material in the connective tissue at the outer cantus (Figure 2F).

DISCUSSION

We report a case of a 39 years old male patient with large CVM situated laterally to the extraconal compartment in the right

orbit. We used endoarterial particles followed by direct puncture with n-butyl cyanoacrylate embolization to devascularize the lesion completely. The CVM was easily removed by an anterior orbitotomy via incision of the skin without complications and a minimal blood loss of 30 cm³.

CVM can be treated successfully with complete surgical excision [6]. However, complications have been reported. The most serious are bleeding and visual loss because of the damage to the optic nerve or lesion of its vascular supply [8,9].

Endovascular embolization techniques are traditionally described for many high-flow vascular malformations of the orbit. Nonetheless, embolization after angiography of an intraosseous CVM has been reported prior to surgical excision. This technique can decrease the arterial supply to the lesion which facilitates the procedure by reducing intraoperative time and blood loss [10,11].

The blood supply to the malformation may be provided dominantly by the ophthalmic artery (OA), branch of the internal carotid artery. In this scenario, microcatheterization of the OA is difficult and with risk of retinal ischemia and visual loss. One useful alternative has been described by Lin T et al., using direct puncture of the lesions after surgical exposure and injection of medical glue in 31 orbital vascular malformations, including 28 CVMs. CVM obliteration with n-butyl cyanoacrylate reduces the risk of haemorrhage, facilitates the complete resection of the lesion and reduces post-surgical recurrence [12].

Direct puncture injection of periocular capillary haemangiomas has been associated with the risk of ophthalmic artery emboli, ischemia and visual loss [13]. Therefore orbital and periorbital embolization procedures should be performed by an experienced neurointerventionist and assisted by a high resolution digital subtraction angiography. We report one case of preoperative embolization by direct puncture and injection of the embolic mixture of n-butyl cyanoacrylate 50% in lipiodol guided by a digital subtraction C arm. Preoperative direct puncture embolization was a useful tool to decrease intraoperative bleeding facilitating surgical removal.

Although larger studies with more number of cases are required to validate the role of direct puncture preoperative embolization in the management of a cavernous venous malformation, our case demonstrates this technique may be useful prior to surgery in order to facilitate the surgical resection.

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