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Abstract

Facial ArterioVenous Malformations (AVM) are rare lesions and present great difficulty in their diagnosis and treatment. We report a case of a 24-year-old male who has been diagnosed a right facial AVM that underwent endovascular embolization with a liquid embolic device and consequently surgical resection. The type of liquid embolic device used has given advantage for both treatment techniques.

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INTRODUCTION

Facial ArterioVenous Malformations (AVMs) are unusual lesions and are the consequence of abnormal development of vascular angiogenesis during embryogenesis (4th - 6th week of gestation) ⁽¹⁾. So, facial AVMs are present at birth, but they become clinically evident with aging. Facial AVMs may have a progressive, invasive and destructive course. In these cases, an early diagnosis and a definitive treatment are necessary⁽²⁻⁴⁾.

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In these cases is generally done a two or three steps treatment: endovascular embolization, followed by surgical excision and follow-up cosmetic surgery if needed ⁽²⁾.

We present a case of 24-year-old male with a right facial AVM that was treated in two steps (endovascular embolization and surgical excision) using a liquid embolic device, which guaranteed a double advantage for both techniques.

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CASE REPORT

A 24-year-old male presented to our hospital for a painful right cheek mass adjacent the mouth corner, which has an evident grown over the past six months. Clinical examination revealed oval, with well-defined margins, mobile, stretched-elastic consistency and pulsatile mass in the right cheek, suspect for an AVM. Consequent Magnetic Resonance Imaging (MRI) angiography of the head-neck revealed a 3.5 cm enhancing mass into the right cheek, anterior to the upper arch teeth, confirming the diagnosis of facial AVM (Figure 1). After a multidisciplinary consultation, patient was candidate for a two steps treatment: first endovascular embolization to close the arterial afferents to the AVM and second a whole surgical resection of the mass.

Institutional review board approval and written informed consent from the patient were obtained. Under general anaesthesia, from the right common femoral artery he underwent Digital Subtraction Angiography (DSA) with a 5-Fr introducer (Terumo, Tokyo, Japan). Selective DSA of the right external carotid artery done with a 5-Fr angulated diagnostic catheter (Cordis, Milpitas, CA-USA) confirmed facial AVM with vascular afferences from right facial artery (Figure 2). After coaxial super-selective DSA of the right facial artery with a Progreat microcatheter 2.7 Fr (Terumo, Tokyo, Japan) and a previous mixing of liquid embolic device Squid-12 (Emboflu, Gland, Switzerland) with a "shaker machine" (time = 20 minutes), the "nidus" and surrounding AVM vessels were embolized with "roadmap" and "stop-and-go" technique. Final DSA control from the external right carotid confirmed the complete embolization of the facial AVM. 24 hours later, under general anaesthesia, the patient underwent surgical resection of the embolized AVM through a trans-oral incision. The distal third of right facial artery and the external margins of the AVM (delimited by the marginal vessels of the lesion) were well identified for their hyperpigmentation given by liquid embolic device (Figure 3). Therefore the right facial artery and the AVM were isolated from the surrounding tissue and whole lesion was removed from the soft tissue bed. The surgical site was closed with resorbable suture (Vicyl 3-0 - Ethicon, Hamburg, Germany) and satisfactory haemostasis was confirmed. The postoperative course was uneventful and patient was discharged on day 3.

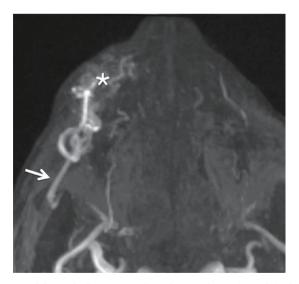


Figure 1. Magnetic Resonance Imaging angiography axial view that demonstrates a 3.5 cm hyper-vascular oval mass (*) at the level of the right cheek with vascular afference from omolateral hypertrophic facial artery (arrow).

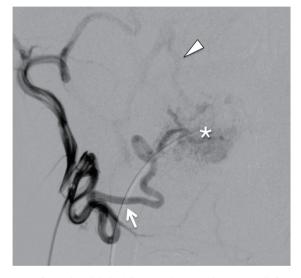


Figure 2. Selective Digital Subtraction Angiography (DSA) of the right external carotid artery that demonstrates a hypertrophic with a tortuous course of omolateral facial artery (arrow), which determines arterial vessels in-flow to the ArterioVenous Malformations (AVM) (*) and with an initial visualization of the early venous out-flow (arrow-head).

Histological evaluation confirmed the diagnosis of AVM and in the various histological sections was clearly visible inside the vessels the presence of thrombus and liquid embolic device (Figure 4). Follow-up till 24 months after endovascular embolization and surgical excision (including clinical observation, RMI) demonstrated no clinical complications and no signs of residual or recurrence of the right facial AVM (Figure 5, 6).

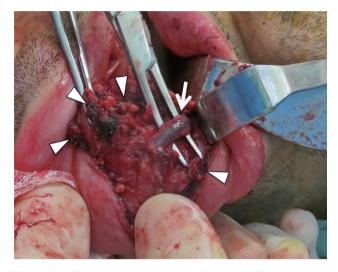


Figure 3. AVM into the right check exposed from a transoral incision, note the distal third of the right facial artery (arrow) and the external margins of the AVM (arrow-heads) as hyperpigmentation vessels due to liquid embolic device.

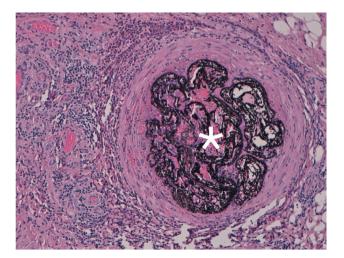


Figure 4. Histologic specimen of surgical resected AVM, note into the small arterial vessel presence of thrombus and liquid embolic device (hyperpigmented component) (*).

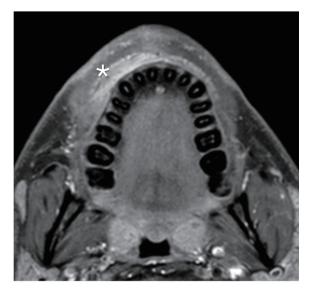


Figure 5. MRI axial view at 24 months that demonstrates at the level of the right cheek a minimum thickening of the soft tissues (*), in the absence of images referable to AVM.



Figure 6. Patient' photo demonstrating the presence of a subcutaneous mass at the level of the right cheek adjacent the mouth corner, with inflamed skin, suspect for an AVM (arrow). B) Patient' photo at 24-months after endovascular embolization and surgical excision demonstrated no clinical complications and signs at the level of the right cheek adjacent the mouth corner (arrow)

DISCUSSION

AVM is an abnormal communication between the arterial and venous system without the normal capillary bed^(2.4). The natural course of an AVM is progressive, invasive and destructive with aging. Thus, multidisciplinary preoperative planning is mandatory to choose the most effective and safe therapeutic techniques^(2,5-9). Nowadays, facial AVMs generally follow a combined treatment consisted of super-selective endovascular embolization and completed by surgical resection (24-48 hours later)^(2-4,9). If needed cosmetic surgery can be carried out during the follow-up^(2,3).

Facial AVM super-selective endovascular embolization has the aim to occlude the "nidus" and the surround feeding vessels. When there is not an adequate vascular access to the feeding AVM vessels, a direct percutaneous puncture with consequent embolization can be a valid alternative⁽¹⁰⁾. This manoeuvre is done with the aim of reducing a possible risk of haemorrhage during the second surgery step. But, the true "limit" of surgery remains of how much the resection has to be extended. It is difficult to assess the whole extension of the lesion. A less invasive surgical intervention may lead a possible recurrence; a more invasive surgical intervention may lead to an irreversible lesion of surround structures^(2,4-12). Theoretically, therefore a complete embolization of the "nidus" and of the peripheral micro-vessels of the facial AVM, associated with a good peripheral demarcation of the lesion allows performing a satisfactory resection. But in the real world, this results is very difficult to obtain, since traditional embolic agents are able to close AVM vessels, but do not allow a precise peripheral delimitation of the lesion.

Embolic agents are classified as solid (example metallic devices) and liquids⁽¹³⁻¹⁷⁾. Liquids embolic materials include adhesive agents (example acrylates) and non-adhesive agents (example ethanol). Last generation of non-adhesive liquid embolic materials are made of ethylene-vinyl alcohol copolymer as the Squid (Emboflu, Gland, Switzerland). The Squid as new non-adhesive liquid embolic material guarantee a slower solidification inside the vessel, a more prolonger injection time and therefore allowing a complete embolization of the "target" vessels throughout their course⁽¹⁵⁻¹⁷⁾. Moreover, the Squid has two technical advantages. The first one that is commercially available in two forms: Squid-18 with more viscosity and Squid-12 with less viscosity. This last form, due to its lower viscosity, has a more penetration capability with an additional advantage in AVMs embolization for their "nidus" and also of their peripheral micro-vessels. Its radiopaque material, the tantalum powder, does the second advantage. The tantalum powder has been inserted with the aim of making the embolic agent clearly visible during the angiographic manoeuvre. But in our opinion, from a visual point of view, the tantalum powder appears dark and this makes it visible macroscopically inside the embolized vessels. Therefore, the Squid-12 can also embolized small peripheral vessels, even those of an AVM, associated with their macroscopic intraoperative visualization (in our

In our opinion in this case of facial AVM, these two advantages have made embolization effective and complete, and have allowed the well-defined resection of the margins during the surgical act without injuring the surrounding healthy tissues.

opinion like a "tattoo"), guarantees a double advantage to

the procedure.

In conclusion, the two steps technique with endovascular embolization and followed by surgical excision for facial AVMs treatment it is considered the safest and most effective. The choice of a non-adhesive liquid embolic device with visible pigmentation can be crucial to perform a complete embolization and surgical excision of the AVM itself.

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