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Research Article

MASSIVE CEPHALIC ARTERIOVENOUS MALFORMATION MANAGEMENT

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A B S T R A C T

Arteriovenous malformations (AVMs) of the frontal region are rare lesions. These vascular anomalies are due to abnormal communications between the nourishing arteries and the draining veins. Due to the hemodynamic nature of these vascular lesions and, sometimes their unpredictable development, they are very difficult to treat. Their multidisciplinary management, which generally requires collaboration between the radiologist, the dermatologist and the surgeon, has grown, thanks to imaging progress and specially endovascular methods.

We report the case of a patient operated in our service for a massive AVM of the frontal region, extended to the scalp. We will discuss the difficulties in the management of this pathology in an emergency situation, as well as a review of the literature.

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INTRODUCTION

Arteriovenous malformations (AVMs) are vascular abnormalities characterized by arteries and veins in direct communication with each other, which leads to the bypass of the normal capillary bed, with rapid and high rate of flow. The abnormal vascular network formed thus is called "nidus". This is a fairly rare birth defect; its exact cause is still unknown. However, an error during intra uterine vascular embryogenesis is very often mentioned.[1,2]

With the progress made in medical imaging techniques, we nowadays, have access to a more precise diagnosis of this pathology.

The treatment of this anomaly is multidisciplinary and depends on several factors that we will discuss in this work. It is also herein presented, the surgical treatment of a massive frontal AVM and the constraints associated with this intervention.[2]

Clinical Case

A 24-year-old patient with no notable pathological history was admitted to our service for bleeding from the frontal region on an arteriovenous malformation (AVM) evolving from birth. This congenital lesion discovered in childhood gradually increased in size with reported bleeding episodes. The patient was referred to us for further emergency management of his bleeding.

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back towards the vertex and descending into the orbito-nasal region. This swelling is dystrophic as a whole, slightly painful on palpation with a throbbing perception. On general examination, the patient is very anxious, prostrate and not very expressive.

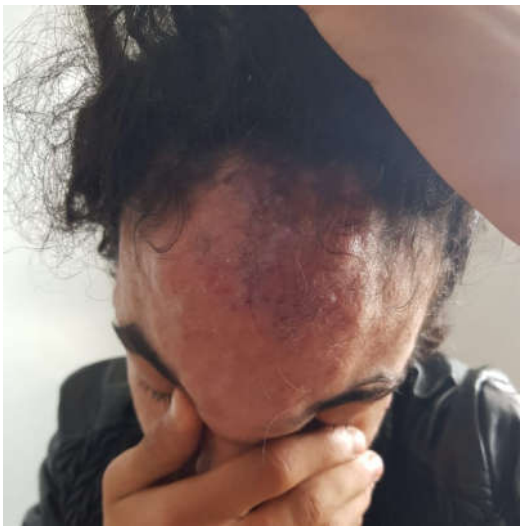

Figure 1 frontal region dystrophy

The first-line angiogram shows: a huge AVM of the frontonasal region supplied by the two superficial temporal arteries.

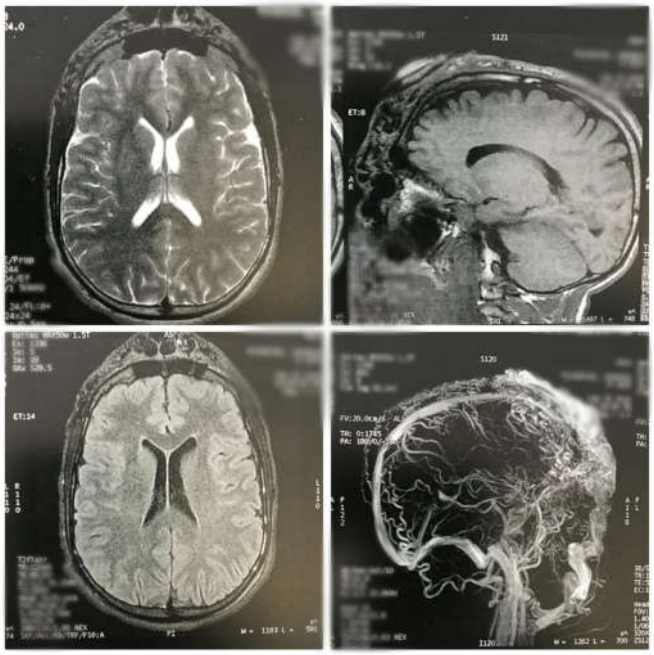

Figure 2 angio IRM pictures

Carotid angiogram performed by puncture of the right femoral objective: a bilateral superior fronto-palpebral AVM, predominantly on the left, with a spreading nidus fine mesh, supplied by sinuous branches of the superficial temporal artery, dilated recurrent branches of the ophthalmic arteries and secondarily by trans-ethmoidal branches of the internal maxillary arteries. Venous drainage was through dilated and sinuous frontal para sagittal veins joining the facial veins. There were no intracranial abnormalities identified during this examination.

Following a pluridisciplinary consultation with the radiology team, we opted for embolization of the two superficial temporal arteries followed by surgical treatment of this malformation due to the bleeding which was worsening at the time of admission, and therefore the risk of a possible dramatic hemorrhagic development. This surgery consisted of a complete resection of the AVM and a microsurgical reconstruction of the defect with a left anterolateral thigh flap.
The outline takes off the scalp from the vertex, descending slightly towards the parietal regions, to end on the face, laterally taking the internal ends 2 eyebrows, the two internal ocular canthi, it widens in the cheek region to finally take the nasal region into its upper 2/3. This resection will be done step by step with progressive hemostasis and ligation after exposure of the 2 superficial temporal arteries and the 2 angular arteries in order to control and properly reduce the bleeding during the procedure.

The AVM will therefore be radically excised, taking away its arteries, its nidus and its drainage veins. The loss of substance was covered by a free left muscular anterolateral thigh flap.

The postoperative period was marked by a suffering of the flap which has become venous within 24 hours and necrosis which sets in after 3 days. This complication was surely due to the arterial micro-anastomosis on the superficial temporal that was dystrophic with a very significant blood rate of flow causing the inefficient venous return and clogging the flap to necrosis. A new operation has been performed to remove the flap.

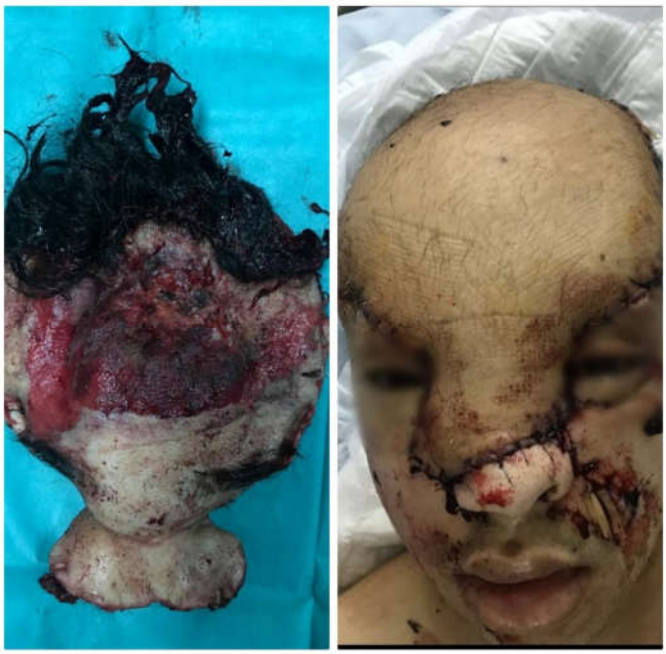

Figure 3 Resection and reconstruction with an anterolateral thigh flap.

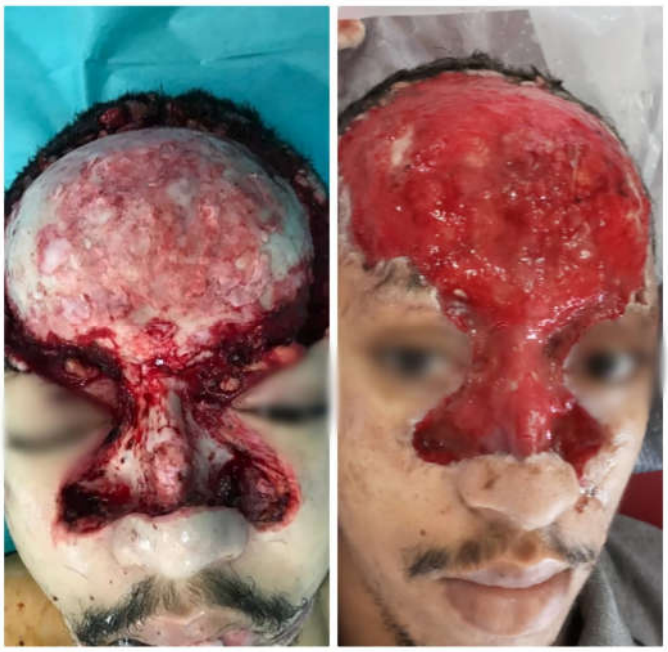

Figure 4 Removal of the anterolateral thigh flap at D5 and evolution after fatty dressings. Evolution after guided cicatrization.

Coverage with thin skin grafts will be done after 3 weeks of fatty dressings. The immediate operative consequences were simple and healing has well progressed

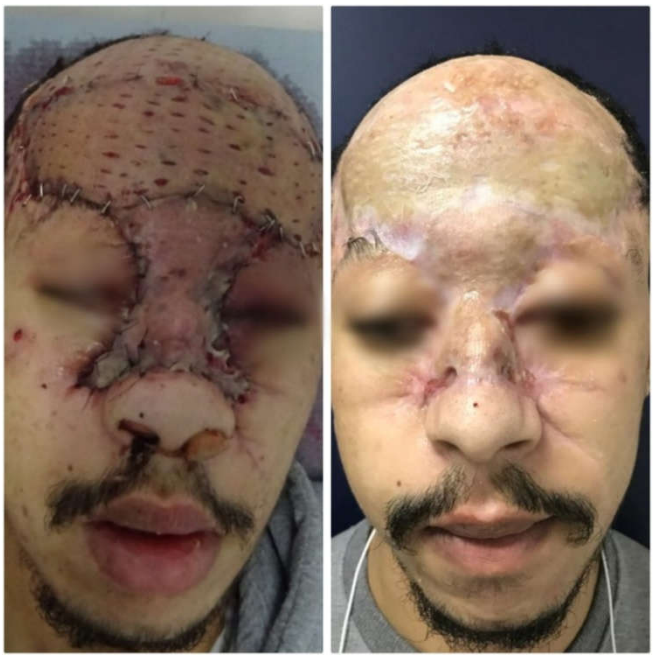

Figure 5 Thin skin grafts and patient outcome at 7 months

DISCUSSION

Vascular malformations have a prevalence of 1.5% of the general population and arteriovenous attacks are rare forms which are present from birth. These anomalies can be found in all organs but have a high prevalence in the face and the ENT sphere. Frontal damage is rare, and a few cases have been published in the literature. The first consultations are made at the end of adolescence or young adulthood, on the occasion of the first morphological disturbances mentioned by the patient or minor trauma with hemorrhagic consequences.[1,2]

The diagnosis of arteriovenous malformations is essentially clinical. On examination, there is a mass that is pulsating hot, with a local increase in the venous drainage network. There is sometimes a local thrill on palpation and the appearance of systolo-diastolic puff on auscultation.

In advanced stages, we notice the appearance of skin lesions such as necrosis, ulcerations and intermittent bleedings. It is especially important, during clinical examination, to remember to look for signs of heart failure, especially when we are in the presence of a massive AVM with a very high rate of flow.[2,3] From a radiological standpoint, this pathology now benefits from developments in imaging techniques, which allows to have a precise diagnosis and sometimes to directly perform certain therapeutic procedures.

Doppler ultrasound is the first-line examination in the presence of suspected arteriovenous malformation. This examination allows us to globally differentiate vascular tumors from malformations and above all to define the anatomical situation of the anomaly. In Doppler mode, fast-flow malformations are differentiated with an arteriovenous shunt (AVM) from other types of slow-flow malformations.

Sometimes the main vessels are brought out with objective blood flow measurements. Thus the Doppler echo is a very practical examination for a precise diagnosis and regular monitoring in less serious forms, and suitable for very young children because it is non-invasive and does not require special sedation. The only drawback of this exam is that he is operator-dependent and the objectivity of his result is subject to the qualification and experience of the practitioner. [3,4,5]

Computed tomography (CT) and magnetic resonance imaging (MRI) are complementary examinations that are useful in the precise diagnosis of AVM. MRI allows above all with a much higher yield to have a more precise estimate of the morphology of the malformation, its extension and its impact on neighboring structures. The different arteriovenous shunts are highlighted during the hemodynamic sequences.[4,6]

Arteriography has long been considered a means of diagnosis, but it is used much more nowadays when a therapeutic gesture such as embolization is opted for. In addition, this technique gives us a better idea of the prognosis of AVMs based on the rate of total obstructions obtained on the lesions.**[2,7]**

Finally, as part of the impact assessment, a cardiac ultrasound will be systematically requested to search for cardiac insufficiency in the presence of high-flow or massive AVMs.**[2]**

At the end of the various explorations, 4 stages of increasing severity of this pathology are booked according to the Schobinger classification. This will then make it possible to define the best therapeutic management to adopt once the precise diagnosis is made.

Schobinger classification of arterio-venous malformations	
Stage	**Clinicalsigns**
I (quiescence)	Hot lesion, bluish pink, Doppler shunt
II(expansion)	Enlargement of the lesion, pulsatile lesion, thrill, tortuous and dilated vein
III(destruction)	Skin dystrophy, ulcerations, bleeding, pains.
IV(decompensation)	Heartfailure

Figure 6 Schobinger classification

It should be noted that AVMs do not typically evolve according to the 4 stages described above. They can sometimes transform into an unpredictable way or conversely remain silent for many years.[2,8]

The treatment of these AVMs, especially at the level of the cephalic end is very complicated due to several factors which are: the high flow rate, the complex vascular anatomy and especially the aesthetic constraints. This therefore requires a broad multidisciplinary consultation (angiology, maxillofacial surgery, interventional radiology, anesthesia, intensive care, psychiatry).[9,1O]

The main goal of the treatment of this pathology is first of all, to make a precise diagnosis, to provide answers to the various questions of the patient and his family, then to ensure a follow-up adapted to the evolutionary stage and to the symptoms of the patient with specific recommendations. This treatment must therefore urgently eliminate any risk of the disease developing which could be life-threatening, ultimately allowing acceptable aesthetic and functional performance, and better social reintegration of the patient.[8,11,12]

We could broadly classify the means available into two groups:

So-called conservative means

Appropriate analgesics if necessary, elastic compressions to reduce edema and local hypertension, appropriate dressings in the event of bleeding, skin care (hydration) and psychological care.

Interventional means:

Mainly two types of interventions are carried out:

- The embolization of the AVM which is done in the afferent arteries by an angiography technique
- Complete surgical excision with ligation of the nutrient vessels.

The attitude to adopt depends mainly on the stage of the disease combined with the profile of the patient. Each case will require customized care depending on the characteristics of the malformation and the functional, somatic and psychological repercussions. Thus, for patients with stage I disease, the intervention will not be indicated. They will benefit from symptomatic and conservative treatment, above all with psychological support which is of paramount importance in view of certain particular locations. Injuries concerning the cephalic extremity, like the case we are treating, have a significant psychological impact and therefore social isolation of patients from adolescence.[8,10,12]

The intervention will be decided in the complicated forms (stages II to IV): severe pain, frequent bleeding, annoying noises, unsightly skin dystrophy, or an impact on the general condition (cardiac, mental). This intervention consists of a first embolization of the main afferent vessels, followed according to the extent of the malformation of an excision and local reconstruction of the loss of secondary substance.

In localized AVMs, the intervention generally includes the aforementioned means: intra-arterial and venous embolization, injection of sclerosing material into the nidus, to end with surgical excision and vascular ligation most of the time. When preoperative embolization is not done, heavy blood loss is almost inevitable. Always be prepared for a transfusion if needed. Although surgery represents the possible solution to eradicating the pathology, its effectiveness depends on the quality of the resection. Any incomplete resection is almost followed by recurrence by recruitment of local vessels.

It is therefore necessary to extend the resection to the pericranial components. Reconstruction of the residual loss is also a challenge for the restorative surgeon. Several means can be employed: thin skin grafting, mobilization of local regional flaps, placement of an expander to obtain sufficient tissue area for secondary coverage or the use of loose flaps. The means of coverage will thus be chosen according to the extent of the loss of substance, its precise location and the underlying structures. [11,12] The extent of the loss of substance in our patient and especially the extension towards the centro-facial part and the nasal pyramid required us to bring more ideally large fascio-cutaneous tissue. We therefore opted for an anterolateral thigh flap.

It is a perforating flap supplied by the descending branch of the lateral circumflex femoral artery which we often use in maxillofacial reconstruction because of the its plastic properties. [13] Our flap unfortunately did not survive due to ischemic suffering from vascular thrombosis. It has been deposited after 72 hours and the coverage has resumed three weeks later with thin skin grafts with satisfactory post-operative consequences. Regular follow-up with clinical checks and psychological support are of capital importance in this care, because the psycho-mental burden to be borne in such mutilation and disfigurement is heavier for the patient than the organic disease itself**.**

We realize that AVM surgery is quite complex and delicate, especially with regard to extensive locations on the cephalic end. [8]

It should be noted, however, that at the same time, our treatment generated a proven mental benefit in our patient who has clearly gained in confidence, but who is still insistent, requesting a reconstruction surgery of the scalp and especially at the level of the nasal area.

CONCLUSION

Cephalic extremity AVMs are rare congenital anomalies with a generally favorable prognosis, but with a course that can sometimes be unpredictable. Doppler ultrasound and MRI are nowadays the first-line examinations carried out for a precise diagnosis of the pathology and its extension to neighboring structures. Embolization remains the therapeutic gold standard used in the event of major aesthetic and functional repercussions or when a complication engages the vital prognosis. However, surgical excision remains a possible solution in certain forms and particular situations despite the high risk of recurrence. It is therefore important to undertake multidisciplinary and personalized care for each case.[12,14]

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The authors declare that there is no conflict of interest

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