



Cirroid Aneurysms, the Arteriovenous Fistulae of the Scalp: Case Series and Literature Review

Aneurismas cirsoideos, las fístulas arteriovenosas del cuero cabelludo: serie de casos y revisión de la literatura



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Summary

Cirroid Aneurysms of the scalp are rare malformations with severe cosmetic deformities and possible potentially lethal hemorrhages. Diagnosis and therapy have been a challenge throughout history, but nowadays imaging technologies give us enough data to facilitate diagnosis and proper therapy plan. From four cases featured in our service unit we will do a systematic review about this type of malformations with their respective correlation.

Resumen

Los aneurismas cirsoideos del cuero cabelludo son malformaciones raras, con alteraciones cosméticas graves y posibles hemorragias potencialmente mortales. Históricamente han sido un reto para el diagnóstico y el manejo, pero la tecnología de las imágenes diagnósticas en la actualidad nos ha aportado datos suficientes para facilitar el diagnóstico y la adecuada planeación del manejo. A propósito de cuatro casos presentados en nuestra unidad de servicio se hará una revisión sistemática respecto a este tipo de malformaciones con su respectiva correlación.

1. Introduction

Arteriovenous malformations (AVMs) are abnormal fistulous connections between the arteries that irrigate and the veins that drain without a capillary regulating component of the flow, which causes sudden dilations of the vascular channels involved, with the subsequent apparent physical deformity (1). Cirroid aneurysms (CA) of the scalp are an infrequent form of AVM, they can be of traumatic origin (in their majority they are) or congenital; the lack of the capillary regulatory network of flow in the subcutaneous cellular tissue generates the typical hemodynamic behavior of the fistulas (2,3).

Generally, they are difficult to treat due to their complex vascular anatomy, high flow bypass, intracranial anastomoses and the possible cosmetic complications that they imply, so the therapeutic method must be chosen with caution.

Many options have been proposed for the management of this type of lesions, including endovascular management with intraarterial or transvenous embolization through the application of sclerosing material and / or coils of platinum. Similarly, surgical manage-

ment with resection and ligation of vessels have been proposed. The latter, usually, as a definitive complement to endovascular therapy.

In this article we will share the experience with endovascular management (spirals and / or sclerosing material) and its results, together with a brief review of the literature.

Because it is of low incidence, the largest case series have been those of Fisher-Jeffers, in which 24 cases were exposed, and that of Gurkanlar et al., in which 21 cases were studied. This series of four cases would be, according to our review of the literature, the first in Colombia (4-7).

2. Materials and methods

Four patients diagnosed with CA who were treated endovascularly in a unit of endovascular and interventional procedures in Cartagena, Colombia, were chosen between 2013 and 2016. The data from this study were collected retrospectively and prospectively. In all cases, brain panangiography with digital subtraction was used to confirm the diagnosis. Magnetic resonan-

ce imaging (MRI) and computed tomography (CT) were used as initial diagnostic methods in some patients.

The subjective aesthetic perception of the lesion was an evaluation criterion and only the concept of the patient in the outpatient clinic was taken into account to evaluate this aspect (excellent, good, acceptable, not acceptable).

3. Methodology

3.1 Endovascular management technique

In two of the four cases presented, only embolizing fluid was used and in the two remaining cases the embolizing fluid plus spirals was used. The decision was based on the cost of the fistula, since in very high expenditure fistulas it is convenient to use spirals that reduce the flow and then embolize with embolizing fluid.

General anesthesia was used to facilitate the adequate visualization of structures and avoid patient movement during the procedure, situations that may affect the image quality (8,9).

All cases were performed in a hemodynamics room with Innova IGS 630 equipment from General Electric.

For vascular access, a puncture was performed with the Seldinger technique via the femoral artery. Then, the catheter system with gauge guide was installed according to the characteristics of the patient through the introducer, to perform a selective digital angiography of the external carotid artery in which the lesions were evidenced, their flow characteristics, their branches feeders and venous drainage vasculature. Subsequently, the system of diagnostic catheters was exchanged and a microcatheter was coaxially augmented, with the help of microguides, to superselectively catheter the branches that showed feeding of the fistula.

For cases in which embolization was performed exclusively with embolizing fluid, the sclerosing substance was injected into the branches that nourish the lesion until fluoroscopy visualized the decrease in the vascular flow thereof and the preservation of the healthy arterial branches. In all cases Onyx® was used as sclerosing material, except in two sessions of case 3 in which the sclerosing agent Phill was also used. Onyx® is an embolizing liquid composed of ethylene-vinyl alcohol copolymer (EVOH) suspended in dimethyl sulfoxide (DMSO) and added tantalum powder to provide radiopacity (10). In cases where it was also decided to use spirals,

it was preferred to start with the use of these and then proceed with the application of the embolizing fluid, in order to decrease the flow in cases of high expenditure and improve control over the administration of the liquid. If the embolizing fluid was administered prior to the insertion of the coils, the risk of extracranial complications of the procedure would increase.

The spirals were inserted until as much flow was prevented as possible towards the site of the lesion, which was evidenced by the administration of contrast medium in control images.

On some occasions, the percutaneous approach to the arterial or venous femoral approach was preferred in order to embolize very superficial distal segments in the scalp. In addition, in the particular cases of CA it is not necessary to drill bone structures to achieve access.

The procedure was performed by direct puncture under ultrasound guidance in the scalp to the feeding vessel to be embolized. Then, embolizing fluid or platinum coils were administered.

4. Results

The age range of the patients was from 22 to 48 years, with three male patients and one female patient. Two cases were fronto-parietal, one occipital and the other temporal. In no case was the patient presented with more than one affected area. Two of the four patients had a strong traumatic history. In one the lesion was of spontaneous appearance and in another one after surgical intervention of hemangioma. In three of the four cases (cases 1, 3 and 4) the AVM extended beyond the midline (Table 1).

The most common clinical symptoms were the sensation of throbbing mass, associated with thrill and global headache. In two cases, retroocular pain was present (Table 2).

In three of the four cases (cases 1, 2 and 4) the main artery involved was the superficial temporal artery (TSA), while in the remaining case (case 3), the left occipital artery was, although after the multiple endovascular sessions that this patient received, new afferents were generated from the left vertebral and ascending cervical arteries. In one of the four cases (case 3), communication with the vertebral artery after several embolization sessions was evidenced in cerebral angiography. In none of the three remaining cases were anastomoses between AVM and intracranial circulation (Table 3).

There were no cases of perioperative bleeding.

Table 1. General information of patients in the study

	Age	Sex	Location	Feeding artery	Aesthetic appearance	Management	Reason for intervention	Complication
P1	27	M	Fronto-parietal	Temporal superficial	Not acceptable	Endovascular	Appearance/risks/clinical	None
P2	40	M	Temporal	Temporal superficial	Acceptable	Endovascular	Risks/clinical	None
P2	23	F	Occipital	Left occipital	Excellent	Endovascular/surgical	Hemorrhages	None
P4	48	M	Fronto-parietal	Temporal superficial	Good	Endovascular	Risks/clinical	None

Table 2. Clinical presentation

	Headache	Thrill	Palpable	Pulsatile	Hemorrhage	Retroocular pain	Tinnitus
P1	X	X	x	X		X	
P2			X	X			X
P3	X		X	X	X		
P4	X	X	X	X		X	

Table 3. Characteristics of procedures

	Access path	Material used	Sessions	Surgical
P1	Intraarterial	Onyx®	3	Not yet
P2	Intrararterial/ percutaneous	Onyx®/ espirals	2	Not yet
P3	Intrararterial/ percutaneous	Onyx®/ Phill	9	Yes
P4	Intraarterial	Onyx®/ espirals	1	Not yet

5. Cases

5.1 Case 1

A 22-year-old male patient who presented with violaceous discoloration of the skin in the right fronto-parietal region in his childhood. At the age of 14 years, when perceiving changes, such as an increase in size associated with headache, thrill and palpitation, he consulted general medicine, but this time it was not interpreted as a pathological finding. Three years later, due to the persistence and worsening of symptoms, he returned to consult and was referred to neurosurgery. He reported not having presented hemorrhagic episodes. The lesion did not deform the face in any way or show another mass on the scalp. Cerebral panangiography was requested in which a right parasagittal frontal arteriovenous fistula with afferent temporal superficial artery and reflux to cortical veins was evidenced, for which reason it was decided to program for endovascular management by sessions. In the three sessions performed, Onyx® and spirals were embolized by endo-arterial intravascular injection. There were no perioperative complications. The patient was assessed one month after the last session with improvement of all clinical symptoms and considerable reduction in mass size; however, he said that on the previous two occasions the behavior had been the same and after a few months the mass began to grow again. Currently, it is awaiting assessment by Tumor Neurosurgery to perform the resection of the lesion as the next therapeutic step.

5.2 Case 2

A 40-year-old male patient who suffered blunt trauma in the left temporo-occipital region. Five days after the trauma, he began to feel a pulsating, depressed mass sensation associated with tinnitus and

violaceous discoloration of the surrounding skin without headache or thrill. The patient reported not having presented hemorrhagic episodes. Initially, he considered that the injury would cease with time, but four months later, due to the persistence of the condition, he decided to consult. The lesion did not deform the face in any way or show another mass on the scalp. Cerebral panangiography was requested in which a left temporal arteriovenous fistula was evidenced with affection of the superficial temporal artery and reflux to cortical veins, for which reason it was programmed for endovascular management by sessions. The first one was embolized with Onyx® and spirals, by endovascular intra-arterial route; in the second one, combined treatment was performed: Onyx® was administered intravascularly and percutaneously, by puncture of a fistula drainage vein under ultrasound guidance in the left temporal region, platinum spirals were placed. There were no perioperative complications. The patient was assessed one month later, with improvement of all clinical symptoms and reduction of a high percentage of the mass size. At 10 months, a control panangiography was performed, which showed complete resolution of the fistula (Figures 1, 2 and 3).

5.3 Case 3

Female patient of 27 years of age who was operated on with a diagnosis of hemangioma in the occipital region, at the age of 13 years. Approximately one year later consult again for hemorrhage. The patient referred occasional headaches, although she did not perceive that they were associated with her underlying problem. In this particular case, the patient had multiple hemorrhagic episodes that caused her anemia and in some cases hypovolemic shock. The deforming vascular lesion was located in the occipital region, but it did not compromise the face or cervical region.

Cerebral panangiography was performed to determine the cause of the hemorrhage, with the finding of a left occipital arteriovenous fistula with an appendix of the left occipital artery and drainage to the right and left jugular veins. She was scheduled for endovascular management by sessions. All together, nine embolizations were performed with Onyx®; in one of them, combined treatment was performed: intravascularly and percutaneously, by puncture of a drainage vein of the fistula under ultrasound guidance in the occipital region, Phill was given embolizing fluid. In no session there were perioperative complications. After the last session, she was surgically treated successfully and even today she reports disappearance of the symptoms, although she reports photophobia with sunlight (Figures 4 and 5).

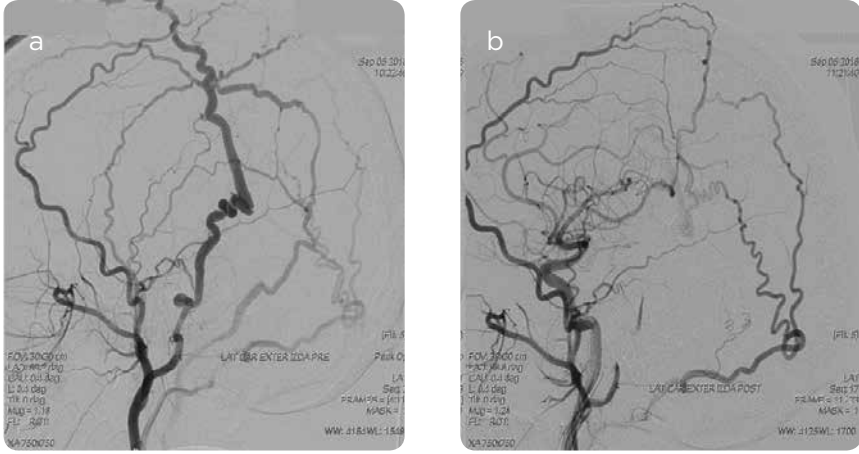


Figure 1. a) Selective catheterization of the left CEA: AVF (CA) is observed with the main afferent of the left STA and drainage vein of the scalp dilated with reflux towards multiple collaterals. b) Lateral CEA lateral projection angiography: spiral occlusion of the main afferent of the left superficial temporal artery with remnant of the fistula due to multiple collaterals of the occipital and middle meningeal arteries to a drainage vein in the scalp.

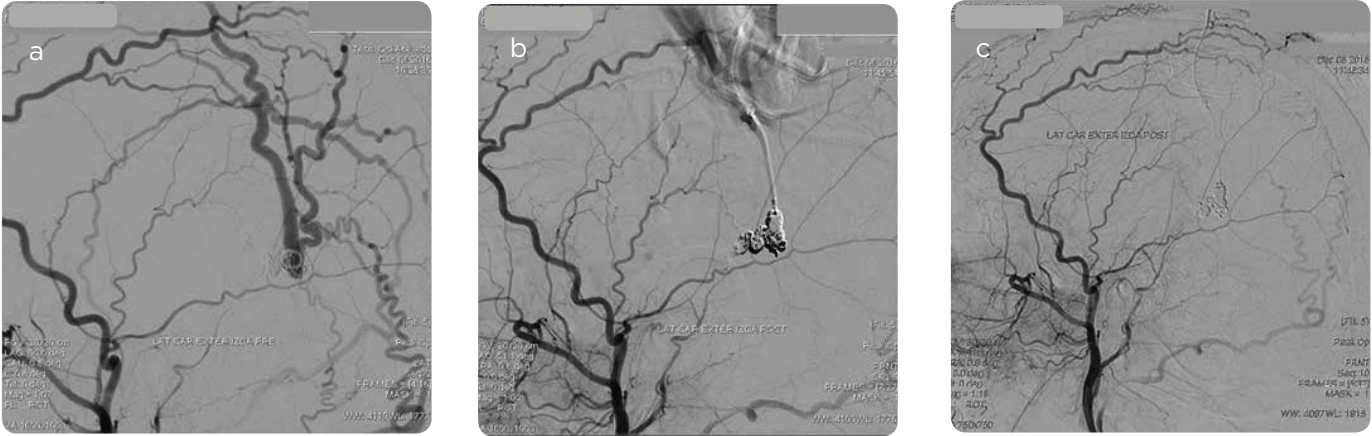


Figure 2. a) Angiography of the second treatment session in lateral projection of the left CEA: multiple afferents are observed to the fistula by branches of the occipital and middle meningeal arteries. b) Percutaneous approach with selective catheterization of the vein of the scalp where the multiple afferents of the fistula were found. c) Lateral angiography of the CEA left after treatment with occlusion by spirals of the cortical vein and healing of the AVF of the scalp (CA).

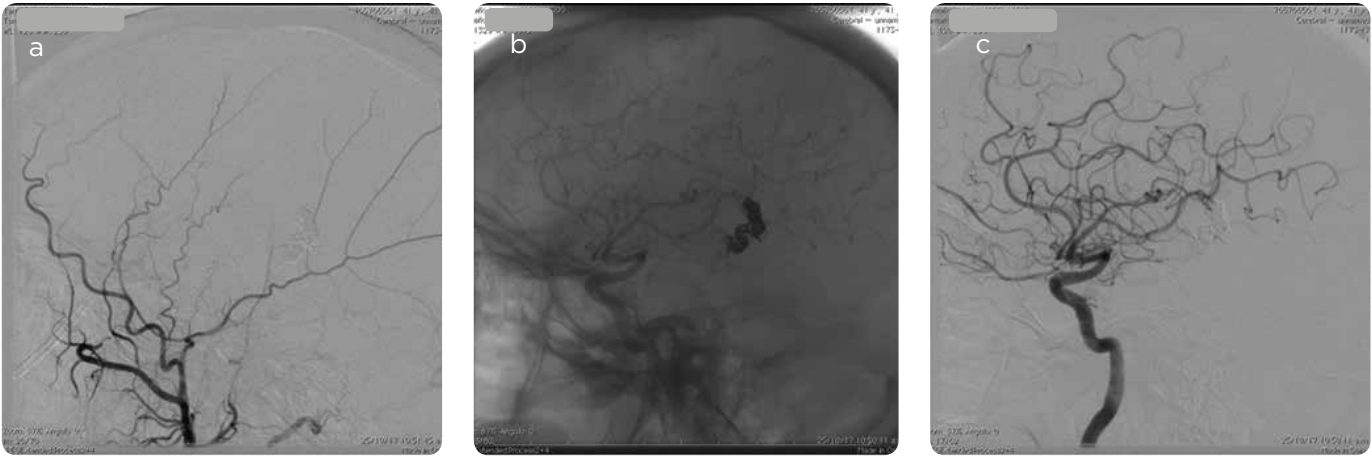


Figure 3. a) Control angiography at 6 months. Lateral view of the internal carotid artery with total occlusion of afferents. b) Spiral mold and ethylene vinyl alcohol copolymer are observed in venous dilatation occluded percutaneously. c) Lateral view of the internal carotid artery with total occlusion of afferents.

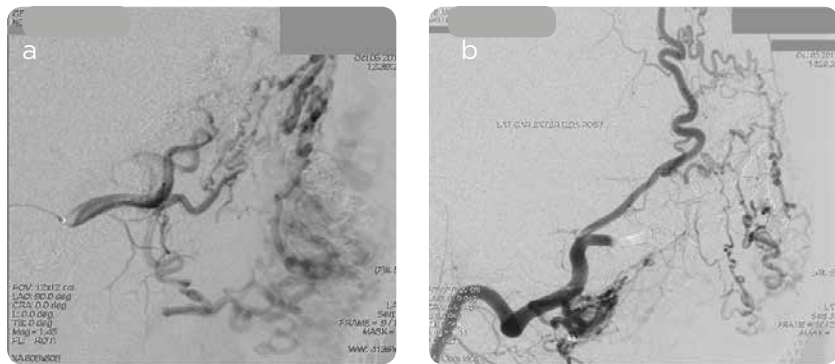


Figure 4. a) Angiography in lateral projection of the left occipital artery: AVF (CA) is observed in the occipital region with multiple afferents of the occipital artery. b) Post-treatment angiography with ethylene vinyl alcohol copolymer by left occipital branch of CA, with multiple remnants by occipital branches.

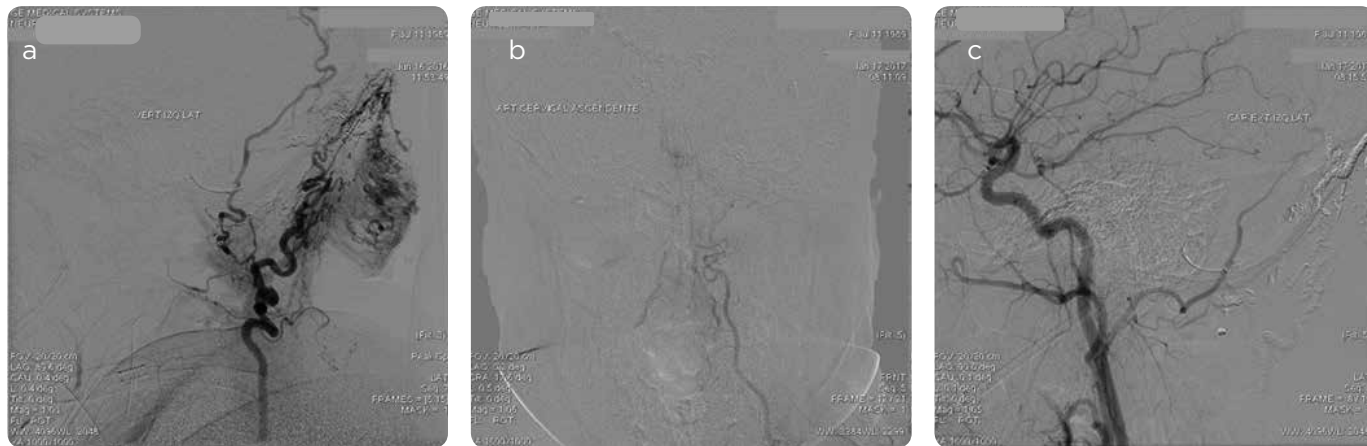


Figure 5. a) Selective catheterization in another treatment session, in this case of the ascending cervical artery. FAV (CA) is seen with multiple afferents of the ascending cervical artery (extracranial resection of the AVF of the scalp 7 months later), satisfactory exclusion of the fistula was observed. c) Angiographic control of the left CEA in lateral projection after surgery with satisfactory exclusion of the fistula.

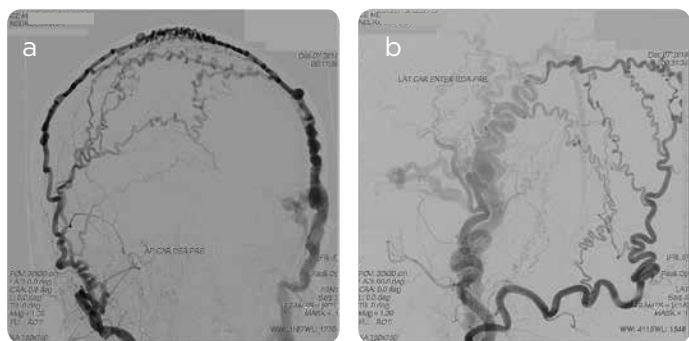


Figure 6. a) Angiography in AP projection of the right CEA: left temporal AVF is observed with multiple afferences of the right and left side (crosses the midline). b) Lateral angiography of the left CEA: AVF was observed with the main reference of the left ATS and the left occipital artery.

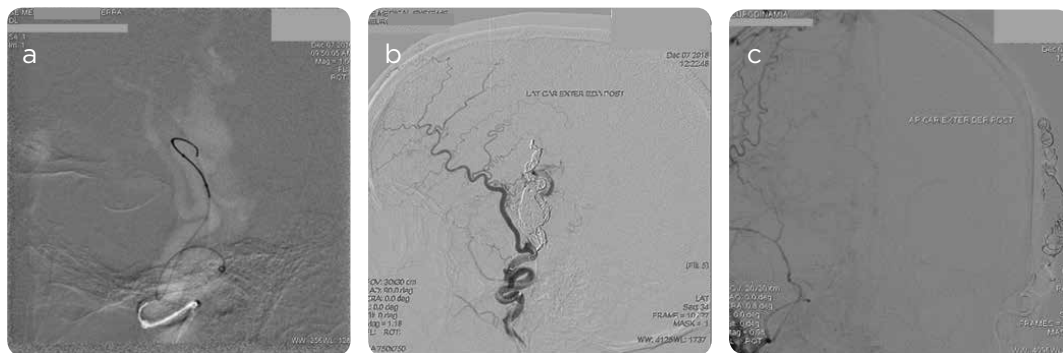


Figure 7. a) Selective catheterization with venography in the lateral view of the left STA. b) Angiographic control in the lateral projection of the left CEA after occlusion with spirals of the main afference of the fistula. c) Selective angiography in AP projection of the right CEA: occlusion of the afferents towards the fistula of the right STA crossed the midline in Figure 6.

5.4 Case 4

49-year-old male patient who suffered blunt trauma in the left temporal region. Two months after the trauma, he began to feel a bulging sensation in this same region, which evolved to become throbbing, depressed, with thrill, accompanied by global headache, retroocular pain and discrete proptosis. The lesion did not deform the face in any way, nor did another mass appear on the scalp. CT, MRI, and MR angiography (MRA) imaging studies were not performed in this case. Cerebral panangiography was performed with the finding of a fistulous lesion with primary afferent of the superficial temporal artery. Therefore, endovascular management with embolizing fluid and spirals was performed, without perioperative complications. The patient was assessed one month after management with improvement of all clinical symptoms, although occasional mild headaches persisted (Figures 6 and 7).

6. Discussion

The nomenclature used to describe cirroid aneurysms has been complex and inconsistent and many alternate names have been raised throughout history. Names such as *cluster aneurysm*, *varicose aneurysm*, *arteriovenous aneurysm*, *aneurysmal variceal*, *plexiform angioma*, *pulsatile angioma*, *abnormal arteriovenous communication*, *arteriovenous malformation*, among others, appear throughout the literature (3,8). The authors of this article prefer to refer to this type of lesions as AVF of the scalp -although in some studies it has also been found as AVM of the scalp, and cirroid aneurysm is a term of global acceptance-, with a defined clinical presentation and complications common and recognized.

The word *cirroid* comes from the Greek *kirsos*, which means varice, and began to appear in literature in the 19th century, when Brecht in 1833 proposed the term. William Hunter's report in 1757 was the first record of arteriovenous fistula in the literature, although some authors mention it in 1764 (7,11-13).

The etiology has generated controversy between traumatic and spontaneous. Some authors affirm that those of spontaneous appearance are more frequent while others affirm the opposite. In approximately half of the cases reported in the literature, there is no history of trauma, and even in a series of cases genetic influence has been suggested (3,14,15).

The superficial temporal artery has been described as the most frequent in cases of post-traumatic cirrhosis aneurysms, because the course of the superficial temporal artery is long and usually exposed, after crossing the zygomatic arch, accompanied by its vein, it courses between the external table of the skull and the subcutaneous cellular tissue, cushioned only by the superficial temporal muscle in a part of its trajectory and is particularly vulnerable when crossing the sharp edge of the superior temporal line, where there are no muscular fibers that cushion the impact. For this reason, some authors have come to affirm that the superficial temporal artery is involved in 75% of traumatic cases, and others, in 90% of cases (8,16). In our review, in three of the four cases, this vessel was one or the most affected (13,17).

The classic pathological description of these traumatic arteriovenous fistulas is of a single vascular channel, well formed between

an artery and a vein given by channeling a thrombus or through an aneurysmal sac; however, there may be multiple channels lined by endothelium suggesting that the pathogenesis consists of an artery and a vein with disruption of its vasa vasorum adjacent to a hematoma, allowing the formation of an endothelial button and its proliferation, to create multiple channels vascular (4,13,18).

Lesions of traumatic origin may be after direct blunt force blows to the scalp or surgical procedures (13). These so-called "cirroid aneurysms" are, in fact, arteriovenous fistulas, the vast majority of which are traumatic, secondary to hematomas that are recanalized to the venous side. However, some authors have even stated that there could be a functional fistula not patent base, and clinically not identifiable, before the trauma, that this would be responsible for activating the injury and, once the short circuit was established, it would receive more flow to through the abundant collateral circulation of the scalp; this theory remains controversial (19). In our study, two of four patients (cases 2 and 4) had a strong traumatic history and one (case 3) was postoperative trauma.

The lesion usually begins as a small subcutaneous lump in the head that evolves into a large deforming mass. The murmur or throbbing headache is usually the initial complaint of patients. In other cases, tinnitus may appear or, rarely, hemorrhage (8). In general terms, the symptoms are well described and vary from a simple deformity, such as a pulsating mass, headache, local pain, tinnitus, tinnitus or thrill to life threatening hemorrhages and skin necrosis (15).

In our cases, we agreed with the literature regarding the most frequent clinical manifestation, with the exception of case 3, in which the patient mainly presented frequent hemorrhages that made her consult and the other symptoms were secondary.

The diagnosis of CA can be easily made with the clinical examination, since its physical characteristics are typical.

Angiography is the gold standard for delineating the lesion and excluding the intracranial component. Some authors, however, have proposed multidetector computed tomography (MDCT) as the preferred method for its diagnosis (20). MRI or angio-resonance may also be helpful for the diagnosis (2,14,21). Some studies report that angiotomography (CTA) can make great contributions by providing the exact location and extent of the lesion, in addition to its relationship with adjacent cranial structures and the possibility of following the contributing arteries and their venous drainage, which is key in the planning of an appropriate treatment (20).

In the cases of this article, angiography was used as the main diagnostic method and, due to the advantages it offers, we believe it should continue to be the case for cases of AVM of the scalp.

7. Management

In 1989 Stanley and colleagues published an article on the endovascular management of scalp AVF associated with large varicose veins, and mentioned that in the past the treatment of this entity was exclusively surgical with the ligation of the contributing arteries, procedures with excessive bleeding and need for postoperative reconstruction; it also referred to endovascular management as a more recent procedure, as well as the frequent recurrence of lesions due to the recruitment of new collateral vessels (16). With the evident

improvement of endovascular techniques, the use of this technique became more frequent and the option of offering preoperative embolization was proposed (due to the high risk of intraoperative bleeding), as well as being an option for definitive management.

Endovascular methods include the application of embolizing fluids, thrombogenic spirals or radiopaque gel particles through the contributing arteries or directly into the percutaneous fistula. It has been reported in the literature that in cases where there are many small afferent arteries, the transvenous approach is recommended, whereas in cases of multivessel or plexiform fistulas, embolization followed by a surgical approach is appropriate (8,16,22,23).

In the endovascular therapy equipment we find: intraleisional injection of sclerosing material, such as sodium tetradecyl sulfate, absolute alcohol, thrombogenic coils, EVOH (Onyx®, the most used by the authors of this article) (8,22,24). In some cases, this method has totally obliterated the lesion without the need for reoperation (8, 24), but in the majority the recruitment of new collateral vessels makes reoperation or definitive surgical management necessary. In our cases, only one of the four patients (case 4) did not have the need to be reoperated.

There are complications with these methods, such as sensitivity and hyperemia, necrosis of the skin over the lesion, permanent alopecia, pain or escape of embolizing fluid into the systemic circulation. These types of complications can occur both endovascularly and percutaneously (8,14,16,22,24). In our study none of the complications mentioned were presented.

There are studies in which surgery without embolization has shown good results and others in which endovascular therapy is recommended to reduce the risks of bleeding and intraoperative times (17,19,25). The main advantage of surgical resection in these cases is the prevention of recurrences. Some authors have proposed that surgical excision can be performed in a single block (26,27).

The endovascular technique offered in our service is very similar to that found in the literature by consensus. Of the embolizing liquids we prefer the Onyx® for its characteristics of results and safety.

8. Conclusion

Cirsoidal aneurysms are rare lesions, but with well defined clinical symptoms and high risk, which can be definitively managed in some cases with endovascular procedure; however, most will require complete surgical excision for this purpose. Endovascular therapy has brought great advances in the management of this type of injuries and its important role in pre-surgical interventions is undeniable.

Study limitations

The discrete number of cases for this article is the main limitation, although in the context of the incidence of this pathology can be a significant amount.

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