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Case report

Scrotal arteriovenous malformation: Case report

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ABSTRACT

Scrotal arteriovenous malformations (AVM) are extremely rare, making them difficult and complex to diagnose, and a therapeutic challenge; few cases of this type have been reported in the literature. This article reports the case of a male patient, 12 years old, with congenital AVM and background of treatment with sclerotherapy and embolization: it was decided to perform a surgical resection, which was successful. Angiography is obligatory in arteriolar cases, and it was backed up with surgical AVM treatment, taking into account the great importance of pre-surgical embolization.

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Introduction

Vascular lesions are rare conditions little described in medical literature, which may mention varicocele, hemangioma, lymphangioma and arteriovenous malformations [1]. Among them all, arteriovenous malformations (AVM) are the least common [2–5]. Their areas of frequent appearance are the

throat, limbs, trunk sites, intracranial and extracranial [6]; therefore, it is said that AVM in the scrotal region are the most rare [1,6–8].

AVM are present from birth, produced by alterations in the morphogenesis of the vessels [9], and grow proportionately to the growth of the child [6,9]. They are characterized by large feeding vessels, hypervascularity and excess of arteriovenous connections at the nidus level [10]; they may be asymptomatic

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or present growth in size, bleeding, pain or azoospermia [1,8], and cause infertility, heart failure, and even fatal hemorrhages [4,11].

Case presentation

Male patient 12-years-old, Hispanoamerican, weighing 33 kg, height 150 cm, secondary school student, who since birth had presented a congenital violet-colored mass at the scrotal region; at 4 years of age he was referred to a pediatric hospital, where he was diagnosed with scrotal hemangioma, which was treated with propranolol 10 mg every 24 hours for 3 years. Later, in 2017, he was referred to a third-level hospital, where angiography and phlebography were performed, and he received multiple sclerotherapy every month for 9 years, with partial improvement. Monthly sclerotherapy was initiated (10cc of glucose solution at 50% and 5cc of sclerol at 0.75%), achieving reduction in the volume of the vascular malformation; with hormonal changes and the onset of puberty, hospitalizations were added secondary to scrotal bleeding (approximately 5 or 6 times a year), and embolizations were performed to control the bleeding.

Upon physical examination of the patient, the genitals showed increased volume; at palpitation they were compressible, without pain, with slow filling and violet-colored circumscribed lesions covering the entire scrotum, the skin at the base and posterior face of the penis, as well as the foreskin. For adequate diagnosis, an angiogram was performed of the internal iliac arteries, detecting part of the scrotal malformation and predominant arteriovenous micro-fistulas on the left side (Fig. 1), and MR angiogram of the abdominal and iliac arteries revealed the location of the anomaly in the inguinal-scrotal, perineal, left ischioanal fossa, base of the penis and pelvic floor regions (Fig. 2). Arteriovenous malformation of the scrotum was diagnosed.

Due to the profuse bleeding in the scrotal area and secondary acute anemia, a multidisciplinary team (angiologist, pediatric urologist, pediatric surgeon, interventionist and plastic surgeon) agreed on surgical treatment and pre-surgical embolization to contain bleeding during resection of the scrotal arteriocapillary vascular anomaly.

Patient received multiple sessions of sclerotherapy and angio-embolization. Due to the recurrence of hospitalizations secondary to profuse bleeding of the arteriocapillary anomaly of the scrotal area, it was decided to perform complete surgical resection and secondary healing reconstruction. On November 30, 2021, the patient entered interventionist radiology, where his clinical record was evaluated and laboratory reports of coagulation time and platelets were found to be within normal parameters, without finding counter-indications for the procedure, since he had been transfused 1 week earlier to compensate for acute anemia secondary to hemorrhage. The patient was placed in dorsal decubitus position after general anesthesia by the anesthesiology service; asepsis and antisepsis were performed in the groin and right cervical areas with placement of sterile fields and puncture was done with 18G needle and, later, placement of vascular introducer with hemostatic valve caliber 5 Fr in the common right



Fig. 1 – Angiogram of the left internal iliac artery.

femoral artery. For the hemostatic valve, a Cobra 4 catheter was used, and the left internal iliac artery catheterized, where the greatest portion of the scrotal vascular anomaly was observed, from the internal pudendal artery to the left obturator (Figs. 3A and B); this anomaly was classified as arteriocapillary vascular malformation. Approximately 3cc of embolic agents (700-900 particles) was applied. Later, fluoroscopic control was done and demonstrated a lack of irrigation to the scrotal vascular anomaly, with reduction of vascular stain of close to 70%. In retrograde phlebography covering the right hemicneck, 5 Fr vascular introducer was placed, where a 5 Fr head hunter catheter advanced, which catheterized the sacrovenous plexus and which with a pass of contrast medium visualized the base of the cavernous bodies, since it was decided not to place closing devices due to the great risk of causing priapism (Fig. 3C). It was decided to use direct puncture embolization of venous anomalies guided by ultrasound and phlebogram.

The percutaneous embolization was performed with gelfoam paste (with the addition of 2cc of lauromacrogol at 0.5% plus 3cc of lipiodol 10%), administered in the venous lacus for a total of 10cc in the left scrotal region, 16cc on the right side and 8cc perineal (Fig. 4). The total amount administered by non-ionic contrast medium, in the angiogram, phlebogram and sclerosis was 100cc.

Then, the vascular introducers were withdrawn and hemostatic control was done, finishing the procedure without inci-

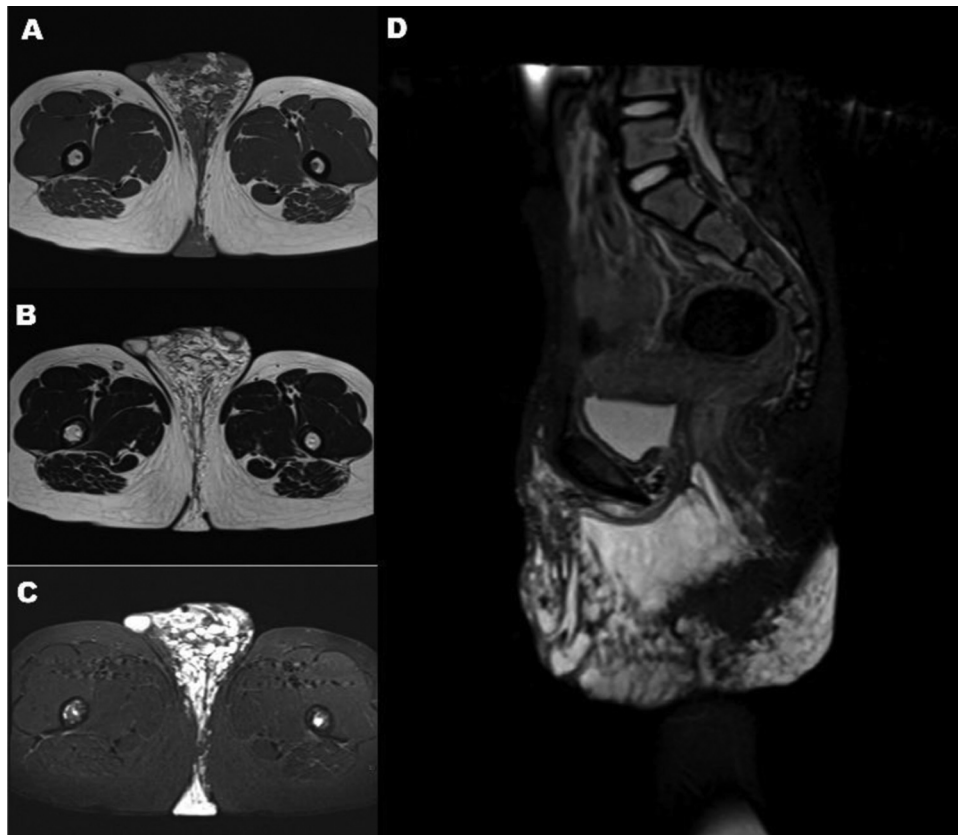


Fig. 2 – MR angiogram of the abdominal aorta: weighted images on the T1 axial (A), T2 axial (B), T2 axial trufi (C) and T2 sagittal trufi (D).

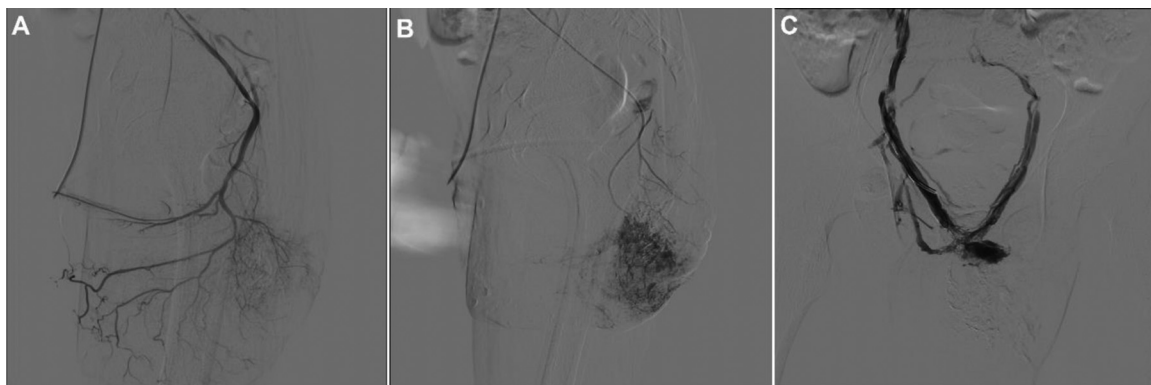


Fig. 3 – Embolization: (A, B) catheterization of the left internal iliac artery; (C) retrograde phlebogram showing sacro-venous system.

dents or accidents of comment. Surgical approach was performed 24 hours after embolization, with resection of the vascular anomaly, where multiple tortuous bleeding veins were observed; the surgical event permitted preservation of part of the scrotal skin, and the vascular surgery service closed the skin while noting residual perineal bleeding, so that a small

plaque of gelfoam was applied and hemorrhaging controlled, thus ending the surgical procedure (Fig. 5).

Histopathological study of the dried lesions showed arteriovenous malformation of the scrotal area.

During days and weeks after the surgical event, infection was found in the small plaque of gelfoam, so it was removed,



Fig. 4 – Percutaneous sclerotherapy of scrotal venous lacus (left venous lacus).

together with start of antibiotic administration, which controlled the infectious process and allowed a favorable scarring of the scrotal incision and ostensible reduction in lesion size.

Discussion

Scrotal AVM are extremely rare, representing less than 1% of vascular neoforations, making them difficult to diagnose and to find adequate treatment [4,6]. For their diagnosis, the physical exam should be taken into account, as well as evaluation of scrotal inflammation, pain, and history of hemorrhages [7]. However, for a precise diagnosis, an angiogram is needed to show the feeding vessels, nidus, and drainage veins, which are essential for embolization, surgical incision or combined treatment [1,2,8,9]. In this case, the angiogram, phlebography and angio-resonance revealed and supported resection. The arteriocapillary malformation favors distension of the veins that carry much of the flow, causing venous hypertension of the area, and this makes the venous capillaries break and causes bleeding, which can become chronic and light or abundant, requiring hospitalization. This bleeding brings the patient to anemia, which should be treated with hemorrhage control and, if necessary, transfusion.

Regarding treatment, sclerotherapy, embolization and surgical resection are recommended as the best option [9,12]. In this case, the patient was submitted to all 3. Sclerotherapy has the function of reducing the size of venous nidus; emboliza-

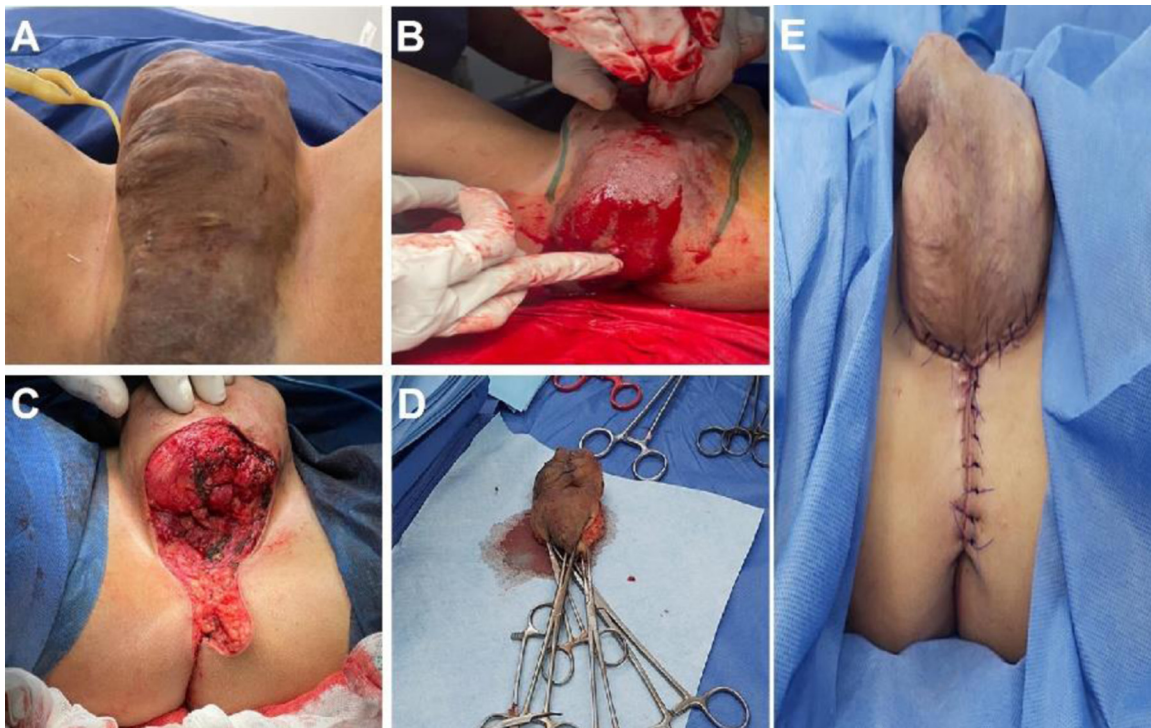


Fig. 5 – Surgical steps: (A) scrotal area before surgery, (B) scrotal hemorrhage at the time of surgical planning, (C) scrotal arteriovenous malformation, (D) surgical resection, (E) surgical closure.

tion is performed before surgical resection to facilitate the procedure with minimum risk of bleeding [2,4], although it is also used as a permanent treatment [8]. Finally, surgery is the definitive, recommended treatment [8]; however, it presents some complications, as malformations tend to be extended and poorly defined [1]. There is a risk of acute hemorrhage [8] and poor management of the procedure can cause impotence and infertility [2]. In the case described, there were no major complications.

Conclusions

Diagnosis and classification help find the adequate, pertinent treatment according to the type of vascular anomaly. It is complex due to its rarity; however, if in agreement in handling, multidisciplinary medical specialists can offer a conservative or invasive management in the case of high-flow lesions or those that compromise the life of the patient, from their location or growth behavior, as well as chronic or profuse bleeding during the life of the patient.

The consensus to decide the fate of the patient in these anomalies requires imaging methods that offer information about the organic implication and extension of the lesion; the vascular phase is highly important, since it gives us the first step in determining if the compromise is arterial or venous. Knowing the classification of the vascular anomalies offers a better dialogue between specialists, since their behavior determines the correct treatment.

Competing Interests

The authors have declared that no competing interests exist.

Patient consent

Informed consent was obtained from the patient's parents.

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REFERENCES

- [1] Jaganathan S, Gamanagatti S, Mukund A, Dhar A. Bleeding Scrotal Vascular Lesions: Interventional Management with Transcatheter Embolization. *CardioVascular and Interventional Radiology* 2011;34(S2):113–16. doi:10.1007/s00270-010-0007-8.
- [2] Konus O, Ilgit E, Yücel C, Özbek E, Önal B. Scrotal arteriovenous malformation and its preoperative embolization. *European Radiology* 1999;9:425–7. doi:10.1007/s003300050686.
- [3] Sule D, Lemmers M, Barry J. Scrotal arteriovenous malformation: case report and literature review. *The Journal of Urology* 1993;150:1917–19. doi:10.1016/s0022-5347(17)35935-9.
- [4] Ascoli Marchetti A, Citoni G, Gandini R, Ippoliti A. Case report of a successful multidisciplinary approach to a giant scrotal malformation. *International Journal of Surgery Case Reports* 2020;69:24–7. doi:10.1016/j.ijscr.2020.03.019.
- [5] Bezirdjian D, Reznikov P, Tisnado J. Transcatheter embolization of an arteriovenous malformation of the scrotum. *CardioVascular and Interventional Radiology* 1989;12:267–9. doi:10.1007/BF02575413.
- [6] Mohammad A, Sahyouni W, Almerce T, Alsaïd B. Angioembolization of scrotal arteriovenous malformations: a case report and literature review. *Case Reports in Vascular Medicine* 2020;2020:1–8. doi:10.1155/2020/8373816.
- [7] Hamid S, Aquillina J, Davidson W, Dhabuwala C. Arteriovenous malformation of scrotum: a case report. *The Journal of Urology* 1992;147:160–2. doi:10.1016/s0022-5347(17)37172-0.
- [8] Zachariah J, Gupta A, Lamba S. Arteriovenous malformation of the scrotum: is preoperative angioembolization a necessity? *Indian Journal of Urology* 2012;28(3):329. doi:10.4103/0970-1591.102716.
- [9] León A, Palacios J, Gutierrez P, Klincovstein J, Gúzman J. Malformación vascular mixta en escroto: informe de un caso y revisión de la literatura. *Acta Pediátrica de México* 2012;33(3):112–19.
- [10] So WLA, Chaganti J, Waugh R, Ferguson RJ. Management of scrotal arteriovenous malformation with transcatheter embolisation coils and percutaneous sclerotherapy under angiographic guidance. *Journal of Medical Imaging and Radiation Oncology* 2015;59(4):468–70. doi:10.1111/1754-9485.12261.
- [11] Danno K, Narushima M, Iida T, Banda C, Todokoro T, Tashiro K, et al. Resection and reconstruction of giant abdominoscrotal arteriovenous malformation. *Plastic and Reconstructive Surgery - Global Open* 2020;8(3):e2725. doi:10.1097/GOX.0000000000002725.
- [12] Sato N, Kuroki T, Tosa Y, Kusano T, Yoshimoto S. A scrotal arteriovenous malformation: a case report. *Modern Plastic Surgery* 2014;04(01):16–19.