EJVES Vascular Forum (2024) 61, 85–88

CASE REPORT

Surgical Treatment of Multiple Venous and Arterial Aneurysms Due to Arteriovenous Malformations of the Arm

Killian Fontaine [*], **Louis Magnus, Gwenaël John, Tristan Leterrier, Mathilde Burgaud, Olivier Rouyer, Fabien Thaveau**

Department of Vascular and Endovascular Surgery, Gabriel Montpied Hospital, Clermont-Ferrand, France

Introduction: Arteriovenous malformations (AVMs) are rare, especially in elderly patients. Occasionally, AVM can produce aneurysmal degenerations, which can lead to bleeding or rupture. The aim of this case report was to describe the surgical treatment of large arterial and venous aneurysms in the arm associated with an AVM.
Report: An 83 year old woman of White ethnicity who was a non-smoker presented with a large pulsatile aneurysm at the left elbow with paresis of the first three fingers. The diagnosis was made by duplex ultrasonography (DUS), computed tomography angiography (CTA), and arteriography. Additional tests confirmed aneurysms of the brachial artery and the outflow veins, with the largest more than 7 cm in diameter. A very proximal brachial artery bifurcation and increased venous flow were noted. DUS confirmed the AVM by showing continuous flow in the axillary vein. The decision for surgical resection involved vascular surgeons, radiologists, angiologists, and anaesthetists. Treatment involved opening and excision of multiple venous aneurysms and AVMs. A short segment of the aneurysmal brachial artery was also resected and repaired with end to end anastomosis. The deep brachial artery which supplied AVMs and venous aneurysms was ligated and excision of these lesions was performed. At one year follow up, there were no complications and the revascularisation was patent.
Discussion: Arterial and venous aneurysms occurring together with AVMs are rare and not well documented in the medical literature. In this case, surgical intervention, including resection with direct anastomosis of the arterial aneurysm coupled with excision of venous aneurysms and AVM, proved to be effective, as evidenced by stable post-operative outcomes after one year.
© 2024 The Authors. Published by Elsevier Ltd on behalf of European Society for Vascular Surgery. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).
Article history: Received 18 September 2023, Revised 18 January 2024, Accepted 7 February 2024,
Keywords: Aneurysms, Arteriovenous malformation, Upper limb, Brachial aneurysm

INTRODUCTION

Aneurysms involving the brachial artery are rare. They most commonly arise from blunt trauma or infections (mycotic aneurysms), thoracic outlet syndrome, arteriovenous fistula, connective tissue disorders, and, very rarely, atherosclerosis.[1] Their main complications are thrombosis and embolisation, which can lead to ischaemia, and may require urgent surgical intervention.[1] Other complications include rupture and nerve compression.[2]

There are few published reports of co-existing arteriovenous malformations (AVMs) and associated arterial and venous aneurysms. Here, the complete management of a patient with upper extremity arterial and venous aneurysms with multiple associated AVMs is described.

CASE REPORT

The case of an elderly woman with pulsatile swelling of the left upper limb is reported. She was a non-smoking, 83 year old woman of White ethnicity; she was a retired hairdresser and right handed. She presented with several bulky pulsatile masses of the left arm, which had been present since 2009 and had shown significant growth in the recent weeks. The patient had no history of upper limb intervention, infections, atherosclerosis, or other aneurysms. She had a left elbow flexion deformity resulting from a childhood elbow dislocation. No family history of cardiovascular diseases or genetic mutations was found.

Clinical examination revealed a large pulsatile aneurysm at the left elbow with signs of skin damage associated with several smaller adjacent aneurysmal formations (Fig. 1). The upper limb was not oedematous.

* Corresponding author. Department of Vascular and Endovascular Surgery, Gabriel Montpied Hospital, 58 rue Montalembert, 68000 Clermont-Ferrand, France.
E-mail addresses: fontaine.killian@gmail.com (Killian Fontaine); louis.magnus@gmail.com (Louis Magnus); gjohn@chu-clermontferrand.fr (Gwenaël John); tleterrier@chu-clermontferrand.fr (Tristan Leterrier); mburgaud@chu-clermontferrand.fr (Mathilde Burgaud); orouyer@chu-clermontferrand.fr (Olivier Rouyer); fthaveau@chu-clermontferrand.fr (Fabien Thaveau).
2666-688X/© 2024 The Authors. Published by Elsevier Ltd on behalf of European Society for Vascular Surgery. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).
https://doi.org/10.1016/j.ejvsvf.2024.02.002

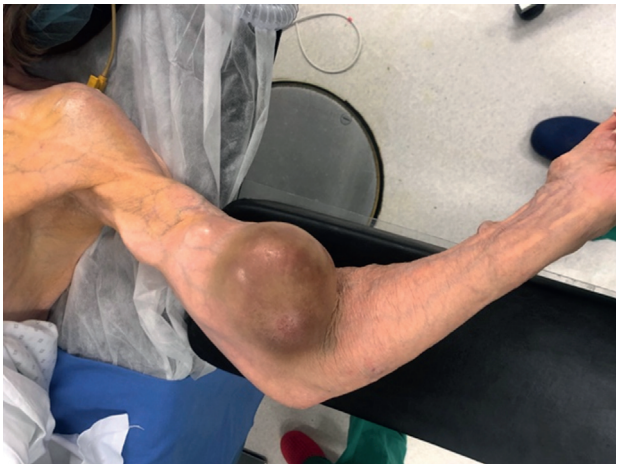

Figure 1. Voluminous pulsatile aneurysm at the left elbow with visible dilation of the superficial veins.

There was significant dilation of the superficial veins with a palpable thrill. Also observed were angiomatous lesions of the second and third fingers associated with paresis of the lateral three fingers, suggesting the presence of a distal AVM in the hand. Unlike the ulnar artery, the radial pulse was palpable with a radial aneurysm above the left wrist.

Duplex ultrasound (DUS) revealed accelerated blood flow in the subclavian artery, reaching 270 cm/second, and slight continuous diastolic flow in the subclavian vein. On the arterial side, there was a significant aneurysm with a mosaic appearance on colour Doppler exhibiting high systolic and diastolic flow velocities with low resistance. The brachial artery bifurcation was identified in the axilla followed by a sequence of pulsatile swellings due to dilations. On the venous side, there was dilation of the draining veins with a systo-diastolic flow at lower speeds. The largest aneurysm, partly thrombosed, measured at least 70 mm in diameter with affected discoloured skin. There were also three minimal aneurysmal formations along the radial artery's path in the forearm, which remained patent, in contrast to the occluded ulnar artery. Deeper, along the external surface of the radius, there was a significant venous network compatible with AVMs, with sequelae of superficial venous thrombosis.

To confirm the arteriovenous communication, a computed tomography angiography (CTA) was performed

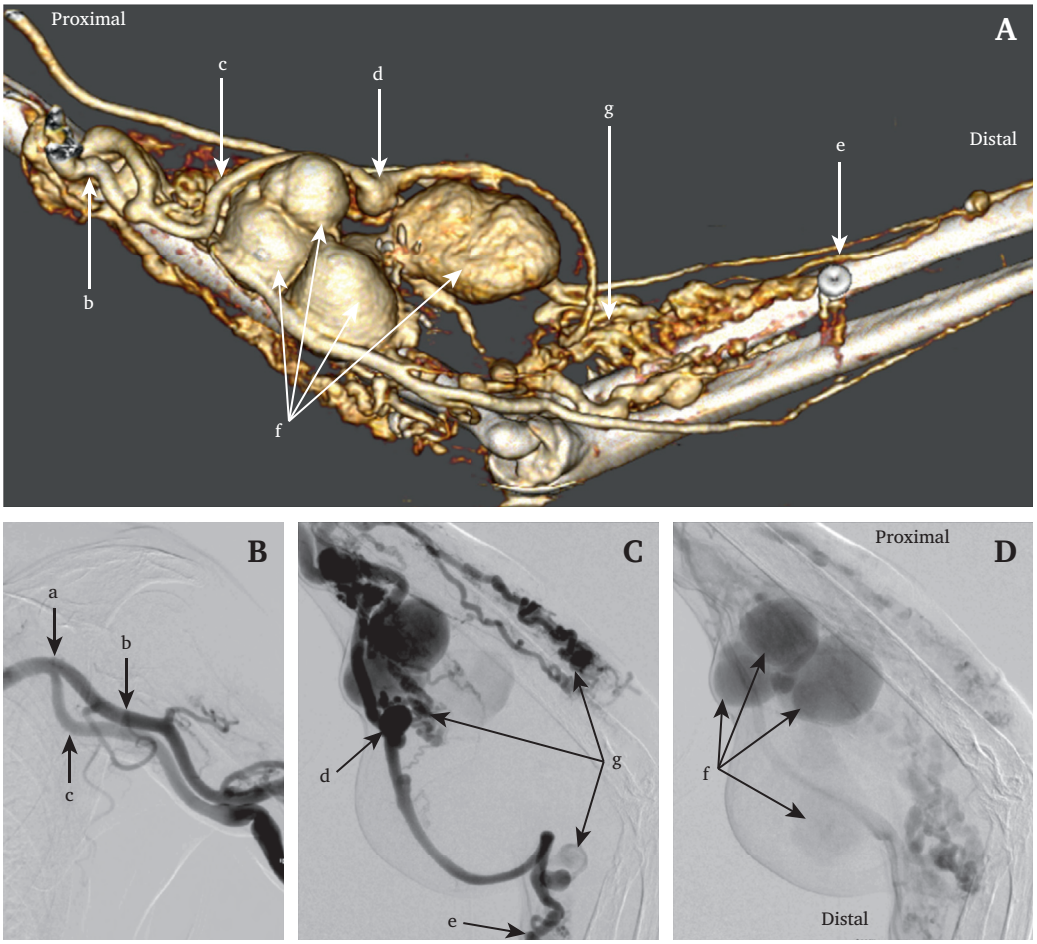

Figure 2. (A) Computed tomography angiography individualising five aneurysmal formations at the portal venous phase. (B) Left subclavian arteriography showing duplication of the brachial network at axillary level. (C) Arteriography of the brachial artery presenting an aneurysm at the level of the elbow with multiple AVMs. (D) Phlebography showing four venous aneurysms, the largest aneurysm was less visible due to the presence of thrombus. (a) brachial artery bifurcation, (b) deep brachial artery, (c) brachial artery, (d) brachial artery aneurysm; (e) radial artery, (f) venous aneurysms, (g) AVM with nidus and dilated outflow veins.

identifying five aneurysmal formations (Fig. 2A). The first four measured 37 × 31 mm, 30 × 28 mm, 19 × 14 mm, and 51 × 43 mm in diameter, without parietal thrombus. The fifth aneurysm, which was partially thrombosed and measured 87 × 74 mm, corresponded to the large subcutaneous mass. CTA revealed abnormally dilated and enhanced outflow veins and tortuous feeding arteries in the arterial phase, which confirmed the diagnosis of AVMs.

Left subclavian arteriography confirmed a duplicated brachial network at axillary level (Fig. 2B). The brachial artery presented an aneurysm in the middle third of the humerus (Fig. 2C). Phlebography showed four venous aneurysms; the largest aneurysm was less visible due to the presence of thrombus (Fig. 2D). There were several AVM niduses extending from the elbow to the distal forearm, and the veins of the arm appeared significantly dilated, probably because of arterialisation due to the high flow through AVM lesions. The complex shunting patterns challenged strict categorisation of the AVM according to Yakes' classification. However, the presence of multiple niduses draining into enlarged outflow veins suggested a Type II AVM.

Due to a significant risk of rupture, surgical exclusion of the main aneurysmal lesion was performed. Under general anaesthesia, a 7 cm axillary incision was carried out, making it possible to control the distal portion of the left axillary artery, the brachial and the deep brachial artery. The incision was then extended along the brachial artery to the mid-forearm to control the radial artery.

Upon opening the largest aneurysmal mass, a contained rupture and evacuated thrombus were observed (Fig. 3A). Along the inflammatory capsule, the brachial artery's wall was discovered, which presented, in the middle third of the arm, an aneurysmal dilation 3 cm in diameter. This aneurysmal segment was excised and a direct end to end anastomosis was performed for reconstruction. The distal portion of the deep brachial artery that was feeding several AVMs was then ligated and the remaining venous aneurysmal lesions were excised (Fig. 3B).

Control angiography was satisfactory, showing a patent brachial artery and its dividing branches supplying the left arm. The proximal deep brachial artery was patent as well, with other AVMs showing no aneurysmal lesions. There was also patency of direct anastomosis and the brachial artery's division into the radial artery without stenosis or haemodynamic abnormalities (Fig. 3C and D).

On DUS, primary patency was confirmed by the presence of arterial Doppler flow with back flow and venous patency by continuous flow and the absence of endoluminal thrombus.

Histological examination of the largest aneurysm revealed a venous like vascular wall.

The patient was discharged seven days after surgery and experienced no complications or adverse events during the one year follow up period.

DISCUSSION

Brachial artery aneurysms occur very rarely, and their surgical management is the subject of few case reports in the literature. These aneurysms can arise as true aneurysms or

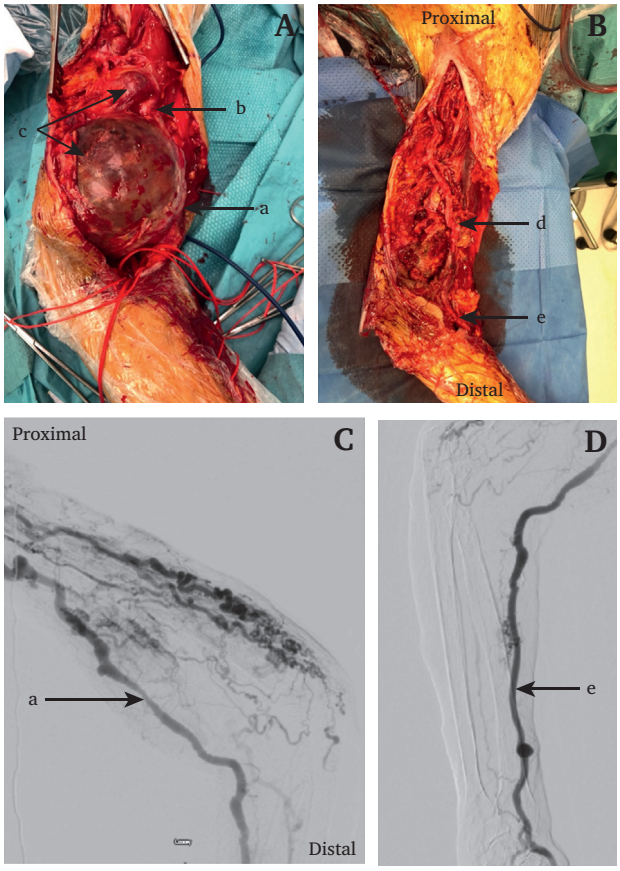

Figure 3. (A) Intra-operative view after control of the multiple venous and arterial aneurysms. (B) Intra-operative view after flattening of the venous and arterial aneurysms and after vascular reconstruction. (C and D) Final arteriography after vascular reconstruction showing good patency of the brachial and radial artery. (a) Brachial artery, (b) brachial artery aneurysm, (c) venous aneurysms, (d) brachial anastomosis, (e) radial artery.

pseudoaneurysms secondary to trauma,[3] iatrogenic aetiology,[4] infection,[5] intravenous drug use, closure of a haemodialysis arteriovenous fistula,[6] connective tissue disease, or degenerative lesions such as neurofibromatosis,[7] with some cases having an unknown aetiology.

The International Society for the Study of Vascular Anomalies classification categorises them into vascular tumours and vascular malformations. Vascular tumours are characterised by the proliferation of endothelial cells. AVMs are classified under vascular malformations as high flow lesions with a nidus of arteriovenous shunts and can be congenital, acquired, or trauma induced. Some remain undetected until childhood, adolescence, or adulthood. AVMs frequently affect limbs, being the second most common site, and, in this case, the elbow AVM could be congenital or the result of a childhood injury. The disease often progresses slowly, categorised by Schobinger's stages: quiescence, expansion, destruction, and decompensation.[8] This patient presented in the destructive stage (stage 3), evident by dystrophic skin changes and the contained rupture of a large venous aneurysm.

In this case, the patient had a deep brachial artery arising from the axillary artery which supplied most of the AVMs, whereas the course and branching of the brachial artery was normal. The variation where the axillary artery bifurcates into a deep brachial artery and superficial brachial artery (brachial artery) is not commonly seen, and it can be the outcome of the natural response to the stimulation by high flow status through the AVM.

This case was unusual because of the co-existence of multiple arterial and venous aneurysms associated with AVMs. Sai Chandran et al. reported a similar case of a woman with multiple serial aneurysms of the brachial artery with an associated AVM of the forearm.[9]

The natural history of brachial aneurysms is not well established but thromboembolic complications from distal embolisation of luminal thrombus are described.[10] The goals of surgery aimed to reduce the risk of limb ischaemia, relieve median nerve compression, and prevent rupture. In this case, these goals were achieved by resecting and reconstructing the aneurysmal portion of the brachial artery. Niduses associated with venous aneurysms were excised as completely as possible to limit recurrence. Due to the advanced age of the patient and the lower risk of progression, a single stage procedure was selected after multidisciplinary discussion. An endovascular approach as stent grafting or embolisation before open surgery was not deemed appropriate in this case due to the urgent nature of the surgery and the significant size of the aneurysms. Generally, surgical considerations included prosthetic interposition bypass grafting, autogenous interposition bypass, aneurysmorrhaphy, or end to end anastomosis. Because the brachial artery was of sufficient length in this case, an end to end anastomosis was performed.

Additionally, the proximal deep brachial artery remained patent, with pathological but non-aneurysmal areas related to other AVMs left untreated due to the low immediate risk. Regular monitoring is essential to promptly identify and manage any new lesions, with endovascular embolisation as the preferred treatment option should they develop.

Conclusion

The co-occurrence of arterial and venous aneurysms with idiopathic and late onset AVMs is rare, and there is a paucity of data in the literature. The treatment is challenging and can be surgical, endovascular, or hybrid, based on the patient's conditions and the anatomical features of the aneurysms. Resection of arterial aneurysms with end to end anastomosis and excision of venous aneurysms and AVM seemed to be a good approach in this case, with a satisfactory result after one year of follow up.

CONFLICT OF INTEREST

None.

FUNDING

None.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

REFERENCES

1 Senarslan DA, Yildirim F, Tetik O. Three cases of large-diameter true brachial and axillary artery aneurysm and a review of the literature. *Ann Vasc Surg* 2019;**57**:273.
2 van der Horst RL, van den Hoven I, Daemen JW. Emergent treatment of ruptured axillary artery aneurysm. *Ann Vasc Surg* 2018;**47**:281.
3 Konishi T, Ohki Si, Saito T, Misawa Y. Crutch-induced bilateral brachial artery aneurysms. *Interact Cardiovasc Thorac Surg* 2009;**9**:1038—9.
4 Wang A, Silberzweig JE. Brachial artery pseudoaneurysms caused by inadvertent hemodialysis access needle punctures. *Am J Kidney Dis* 2009;**53**:351—4.
5 Simson R, Jacobs T, Kulkarni SR. Mycotic aneurysm of brachial artery secondary to infective endocarditis. *EJVES Short Rep* 2020;**46**:9—11.
6 Battaglia L, Bucci F, Battaglia M, Reddler A. Late occurrence of a large brachial artery aneurysm following closure of a hemodialysis arteriovenous fistula. *Ann Vasc Surg* 2006;**20**:533—5.
7 Emori M, Naka N, Takami H, Tanaka TA, Tomita Y, Araki N. Ruptured brachial artery aneurysm in a patient with type 1 neurofibromatosis. *J Vasc Surg* 2010;**51**:1010—3.
8 Gilbert P, Dubois J, Giroux M, Soulez G. New Treatment approaches to arteriovenous malformations. *Semin Interv Radiol* 2017;**34**:258—71.
9 Sai Chandran BV, Nayak D, Badhe B, Subbarao KSVK. Multiple serial aneurysms of brachial artery with multiple arteriovenous malformations of forearm. *Ann Vasc Surg* 2011;**25**:555.
10 Gray RJ, Stone WM, Fowl RJ, Cherry KJ, Bower TC. Management of true aneurysms distal to the axillary artery. *J Vasc Surg* 1998;**28**:606—10.