ELSEVIER

Case Report

A Rare Case of Dysplastic Axillary Artery Aneurysm Associated with Arteriovenous Malformation

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Background: Axillary artery aneurysms are rare conditions, and their causes are various. They can determine severe complications, so the treatment is extremely important.
Methods: We report the case of a young man affected by a saccular axillary artery aneurysm associated with intramuscular arteriovenous malformation, without symptoms except for the presence of a pulsatile mass. Duplex scan and computed tomography scan have been essential for a correct diagnosis and planning of the treatment. At first, the patient was submitted to coil embolization of an efferent vessel, and then he was treated surgically through ligation and detachment of the aneurysm and replacement of part of the axillary artery with a Dacron graft (Vascutek, Inchinnan, Renfrewshire, Scotland, UK).
Results: Follow-up at 1 and 6 months revealed normal patency of the axillary arterty and the prosthetic graft with complete exclusion and thrombosis of the aneurysm sac.No sensitive nor motor deficit were observed.
Conclusions: Aneurysms of the axillary artery associated with intramuscular arteriovenous malformations are very rare, but have to be suspected. The treatment is challenging and can be surgical, endovascular, or hybrid, based on the patient's conditions and aneurysm's anatomical features.

INTRODUCTION

Aneurysms involving the axillary artery are rare findings in vascular surgery practice. They are mostly associated with 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 embolization, which can lead to the ischemia of the upper limb and may require urgent surgical intervention.[1] Less frequent complications are rupture and nervous compression.[2]

Case Report

We observed the case of a young man who was incidentally diagnosed with a true saccular aneurysm of the axillary artery, associated with intramuscular arteriovenous malformation (IMAV-M) of the shoulder and the arm. He was a 49-year-old carpenter that presented to our observation with an asymptomatic but bulky pulsatile mass in the left armpit. The upper limb was not edematous, and distal pulses were valid. The patient had no history of trauma, infections, atherosclerosis, or other

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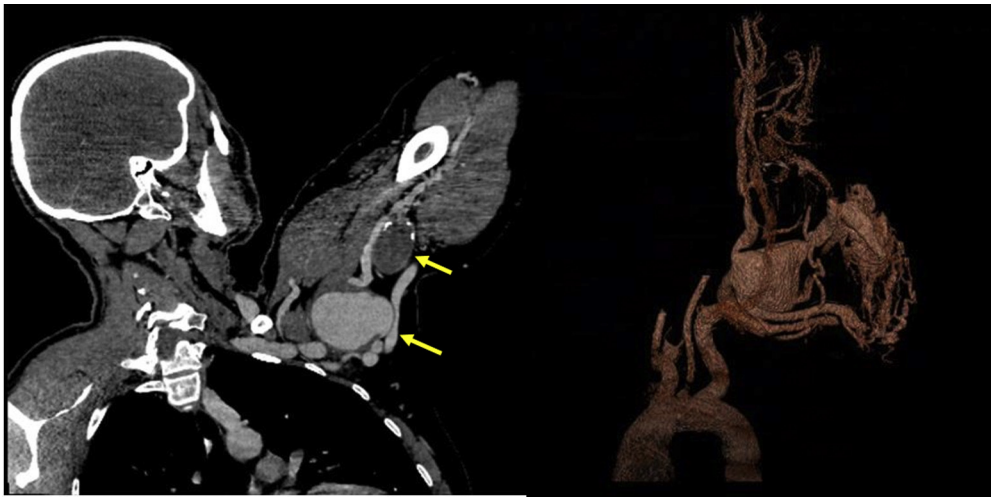

Fig. 1. The CT angiography with multiplanar and volume rendering reconstructions. The *arrows* show the first big patent saccular aneurysm and the second saccular spontaneously occluded aneurysm. The whole arterial axis is patent.

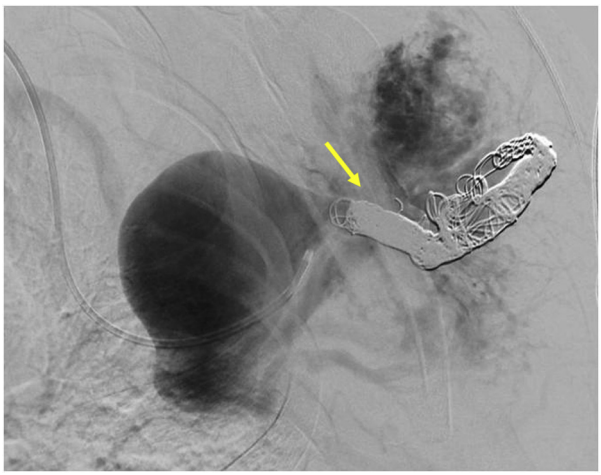

Fig. 2. Embolization of the efferent vessel (*arrow*).

aneurysms. He had not been experiencing pain or sensitive and motor deficit.

First, we performed a Duplex scan that revealed an ectasia (14 mm) and low resistance flow patterns of the first part of the left axillary artery and turbulent and accelerated flow in the axillary vein. Moreover, there was a patent saccular aneurysm of the second and third part of the artery (50 mm in diameter deep in the armpit) with large neck and another very saccular but totally occluded aneurysm (30 mm in diameter) with narrow neck likely originating from a side branch. The axillary artery trunk close to the latter aneurysm was patent. Inside the proximal bigger aneurysm, there was high and swirling flow with one big efferent branch. For this reason, we suspected an arteriovenous communication and we decided to comply with a computed tomography (CT) scan.

The CT angiography (Fig. 1) revealed multiple dilated and twisted arteriovenous nest formations within the subscapularis, supraspinatus, and infraspinatus muscle, showing arterial afferences from different axillary artery branches. The big saccular axillary aneurysm, measuring 58 mm × 41 mm in diameter, originated from the posterior wall of the second/third part of the artery; an efferent branch, taking origin from the deep wall of the aneurysm, was linked to the IMAV-M. Caudally, the CT scan confirmed another totally occluded aneurysm (30 mm diameter) and other little arteriovenous fistulas from the other branches of the humeral artery. Therefore, the hypothetic diagnosis of a dysplastic axillary artery aneurysm associated with congenital IMAV-M was confirmed.

Because of the patient's young age, job, particular anatomical site (subjected to flexion movements), and absence of symptoms related to IMAV-M, we chose to treat only the bigger patent aneurysm in 2 steps. First, the efferent vessel of the aneurysm directed to the IMAV-M was embolized using 4 Interlock Coils (Boston Scientific, Natick, MA) and 12 Penumbra Coils (Penumbra Inc., Alameda, CA) (caliber ranging from 8 mm to 32 mm) to reduce the subsequent risk of intraoperative bleeding

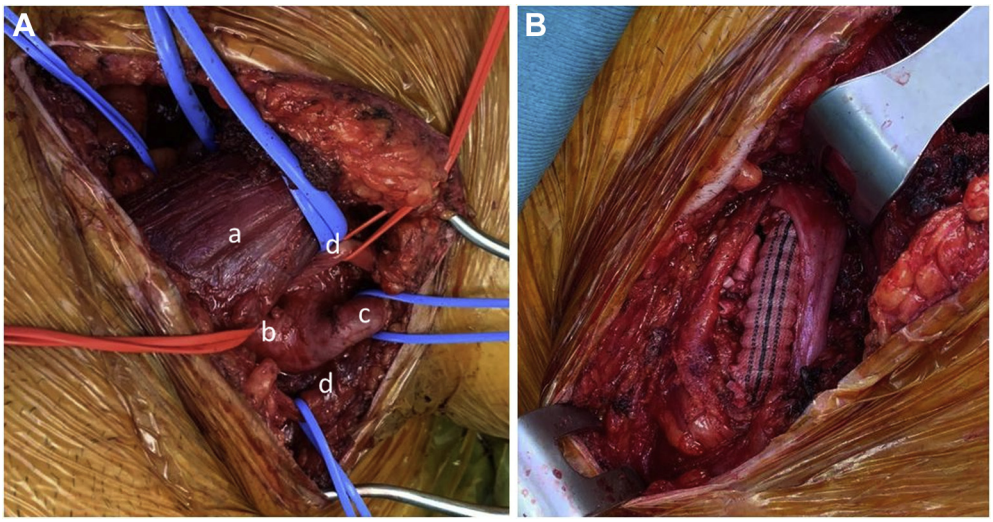

Fig. 3. (**A**) The pectoralis major muscle, (**B**) proximal axillary artery, (**C**) distal axillary artery, (**D**) brachial plexus cords. **(B)**: Replacement with a Dacron graft.

(Fig. 2). For this reason, we did not use any liquid agent like cyanoacrylate or alcohol.

Second, we proceed to the surgical repair. After incision in the pectoral-deltoid groove, we cut the pectoralis major muscle, preserving its tendon and the brachial plexus cords. Isolated the proximal and distal segment of the para-aneurysmatic axillary artery (Fig. 3A), we ligated 2 branches originating from the large neck of the aneurysm which developed deeply and posteriorly in the armpit. After heparinization and clamping, to avoid the residual back-bleeding from the sac, before opening the aneurysm, we performed a continuous double suture leaning on Teflon stripes on the aneurysm's neck to detach the aneurysm from the rest of the artery, excluding it from the direct arterial flow. The dysplastic quality and thinness of the arterial wall were microscopically clear. After resecting a short part of the axillary artery, we replaced it through 8 mm Dacron graft (Vascutek, Inchinnan, Renfrewshire, Scotland, UK), performing an end-to-end anastomosis (Fig. 3B).

The postoperative period was uneventful, left upper limb pulses were valid, pain was well controlled, and the patient did not report any sensitive or motor deficit. He was discharged on the third postoperative day. The patient received preoperative antibiotic coverage and postoperative prophylactic anticoagulation (enoxaparin 4000 UI daily for 3 days) and aspirin for three months.

The first clinical and ultrasound follow-up was performed after 1 month from the procedure. The patient was fine, with no sensitive or motor deficits and able to perform every movement of the shoulder. The Duplex scan revealed normal patency of the axillary artery and the prosthetic graft with complete exclusion and thrombosis of the aneurysm sac (Fig. 4A, B). The distal artery was patent with accelerated and low-resistance flow because of the origin of other branches afferent to IMAV-M (Fig. 4C). The ulnar and radial arteries were normally patent. The axillary vein was patent as well, with persistent accelerated and turbulent flow due to a residual arteriovenous shunting.

At 6 months, we confirmed these findings and, in addition, we observed the shrinkage of the treated aneurysm (Fig. 5). The diameter of the second saccular aneurysm was stable (30 mm).

DISCUSSION

Peripheral aneurysms of the subclavian-axillary segment account for less than 1 % of all aneurysms,[3] of which 88% occur in the subclavian artery. The main cause of axillary artery aneurysm is accidental or iatrogenic trauma; other causes may be represented by infections, thoracic outlet syndrome, arteriovenous malformations (AVMs), connective tissue disorders, and, very rarely, atherosclerosis.[1] Patients with axillary aneurysms are often asymptomatic, except for the presence of a pulsatile mass; symptoms may appear as the aneurysm grows, causing compression of the brachial plexus, with consequent neurological defects. Furthermore, the

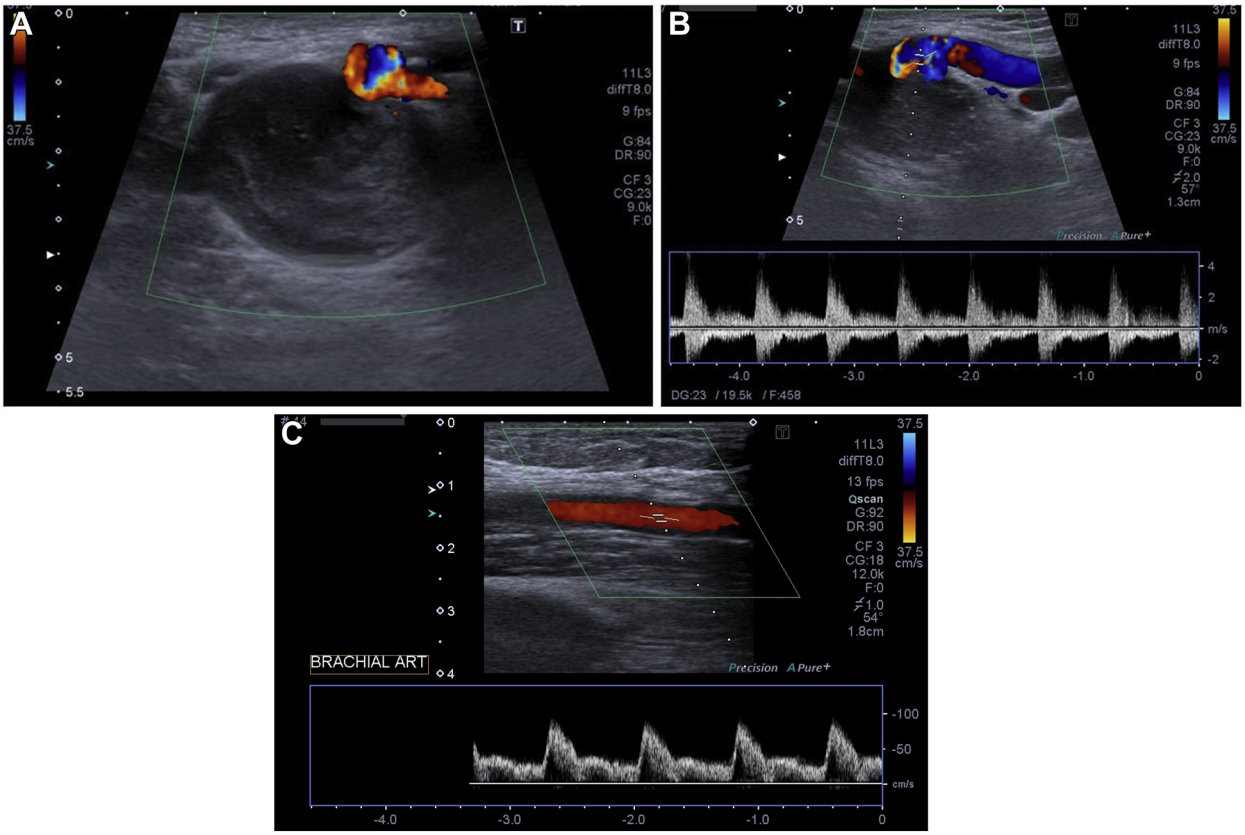

Fig. 4. **(A)**: Thrombosis by exclusion of the axillary artery aneurysm. **(B)**: Patency of the graft. **(C)**: The distal axillary artery with low-resistance flow.

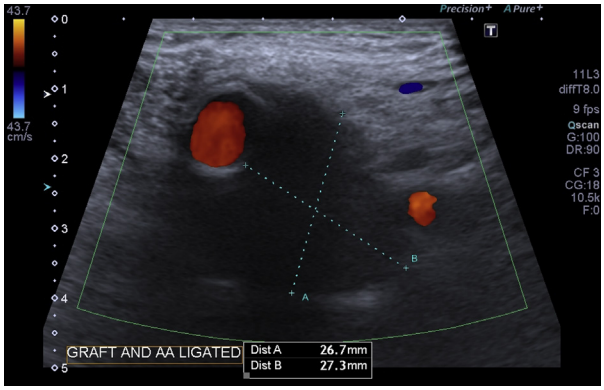

Fig. 5. Shrinkage of the treated aneurysm at 6-month follow-up.

aneurysm can determine thromboembolic events with following ischemia of the upper limb or rupture with severe and life-threatening bleeding.[4]

The most common vascular anomalies are the venous malformations (70%), followed by lymphatic ones (12%), arteriovenous ones (8%), combined malformations (6%), and, finally, capillary malformations (4%).[5] In this case, we observed both the conditions of aneurysm and intramuscular AVM in a patient with no history of connective tissue disease, trauma, signs, or symptoms.

Duplex scan is the first diagnostic line for the diagnosis of axillary artery aneurysms and flow characterization. Vascular malformations, for their part, are a diagnostic and therapeutic challenge, requiring dedicated interdisciplinary management. AVMs are high-flow vascular anomalies: Sadick M et al. affirm that with increasing size, it becomes extremely difficult to locate the area of direct arteriovenous shunting connections (the ''nidus'' of the AVM) and to differentiate between feeding inflow arteries and draining outflow veins.[5] If performed by well-trained staff, Duplex scan gives reliable information on the high-flow arteriovenous shunts.

In our case report, the suspect of an axillary artery aneurysm associated with an AVM was based on the echographic morphological pattern of the swelling and the flow features of afferent and efferent

vessels. In fact, the Duplex scan revealed a big pulsatile axillary artery aneurysm with high and swirling flow, without thrombus and one big efferent branch, dilatation and low resistance flow in the proximal axillary artery associated with high and turbulent flow in the axillary vein, and lower caliber and flow in arteries distal to the aneurysms. For this reason, we suspected an important distal arteriovenous shunting and we decided to perform an angio-CT scan.

The panoramic view of the CT scan confirmed the arterial features, showed large intramuscular arteriovenous nidus (subscapularis, supraspinatus, and infraspinatus muscles) with some arterial afferent branches taking origin close to neck of the aneurysm. The distal presence of another chronically saccular occluded aneurysm supported the hypothesis of an overflow component in the genesis of both aneurysms, of which, the one with efferent branch had remained patent.

The treatment of axillary aneurysms is still a matter of discussion. The main concern is the identification of an adequate procedure with low invasiveness and acceptable morbidity and durability. In accordance with different studies, conservative management may be possible in asymptomatic patients, but complications have been reported in up to 50%.[6,7] Thus, because of the aneurysm diameter, we decided to treat it promptly.

Tham et al. realized a review of the literature and discussed about the main operative options: open surgical repair versus endovascular exclusion.[8] Both are viable options, with no absolute superiority of one treatment above the other. The appropriate treatment depends on the patient's characteristics (operative risks, age, work, comorbidities, and preferences) and aneurysm's factors (size, location, anatomical variation). Open repair is the standard treatment of care for this pathology: it achieves the complete resection of the aneurysm with grafting and consequently the reduction of compression on the surrounding structures and the removal of the risk of rupture and thromboembolism.[9] Nevertheless, it carries greater morbidity risks, such as bleeding, potential brachial plexus injury, and graft (prosthetic or venous) stenosis or occlusion.[6]

The alternative to surgical repair is the endovascular strategy, consisting in the exclusion of the aneurysm through the insertion of a stent graft. The coil embolization has been reported in the emergent management of ruptured axillary aneurysms, only as an interim measure due to the high risk of limb ischemia.[2,10] The endovascular treatment implies lower morbidity and mortality rates, offers benefits regarding patient discomfort, and limits the intraoperative and postoperative risks of the surgical open repair.[11,12] Nevertheless, it presents severe limits as well: first of all, high risk of stent collapse, fracture and compression on near the thoracic outlet, durability, and more extensive collateral branch coverage.

We decided to submit our patient to open surgery before coil embolization for the following reasons: the patient was a young man, with no comorbidities, performing a job requiring wide movements of the shoulder, and the segment involved was the second-third part of the axillary artery, more exposed to shoulder movements; the aneurysm developed in association to a large IMAV-M with multiple inflow and outflow vessels (some of them from the aneurysm), which would have been a potential source of hemorrhage and back-bleeding leakage.

The surgical treatment was performed under general anesthesia and consisted of the section of 2 inflow para-aneurysmatic branches, ligation and resection of the aneurysm with embolized efferent branch to exclude it from the direct arterial flow without opening it, to avoid residual back-bleeding. In the absence of preoperative compression symptoms, we did not consider the problem of the possible mass effect of the excluded residual aneurysmatic sac. Furthermore, we avoided the use of the autologous saphenous vein for immediate diameter mismatch and for the risk of dilatation overtime in a patient with a long life expectancy. The second distal aneurysm was not treated because it was saccular with a very narrow neck and likely involved a side branch and not the main artery wall; moreover, the aneurysmal sac was completely occluded.

CONCLUSION

Aneurysms of the axillary artery associated with intramuscular AVMs are very rare, but have to be suspected. The treatment is challenging and can be surgical, endovascular, or hybrid, based on the patient's conditions and aneurysm's anatomical features.

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