Case report

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Peripheral embolism as first and only clinical symptom of a true aneurysmal degeneration of the radial artery after ligation of a radiocephalic fistula

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Abstract

True aneurysmal degeneration of the inflow artery after arteriovenous fistula ligation is extremely rare. Pain is the most common symptom and surgical treatment by an autologous venous bypass is considered as the treatment of choice with good long-term results. We present a patient with peripheral embolism as first and only symptom leading to the diagnosis of a true aneurysmal degeneration of the entire left radial artery. It was discovered 5 years after the ligation of his radiocephalic fistula. As illustrated by this case, a conservative treatment by antiplatelet and anticoagulation therapy should be considered a satisfying alternative to the standard bypass surgery in patients with anatomical variations (e.g. an incomplete arterial palmar arch) since the latter include a higher risk of postoperative ischemic complications.

Keywords

True aneurysmal degeneration, arteriovenous fistula ligation, peripheral embolism

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Introduction

An autogenous arteriovenous fistula (AVF) is the preferred vascular access for patients with end-stage kidney disease requiring long-term hemodialysis.[1] After kidney transplantation, no consensus exists regarding routine access ligation because of the need for hemodialysis access in case of graft failure. Delayed AVF ligation may lead to various complications such as embolization due to thrombus formation, venous hypertension, steal syndrome, high-output cardiac failure, overmaturation, infection, nerve compression, arteriomegaly of the inflow artery, venomegaly of the outflow vein, rarely aneurysms, and even more scarcely aneurysmal degeneration of the inflow artery.[2,3] Therefore, a periodic clinical and radiological follow-up of the AVF is recommended after kidney transplant but often not performed in clinical practice.[4] More than one-third of the AVFs close spontaneously after transplantation.

Inflow artery aneurysmal degeneration is rare. It can occur in long-term arteriovenous accesses and is not always prevented by ligation or thrombosis of the fistula.[4–6] In contrast to its classical presentation with pain as main symptom, we hereby present a patient with peripheral embolism as

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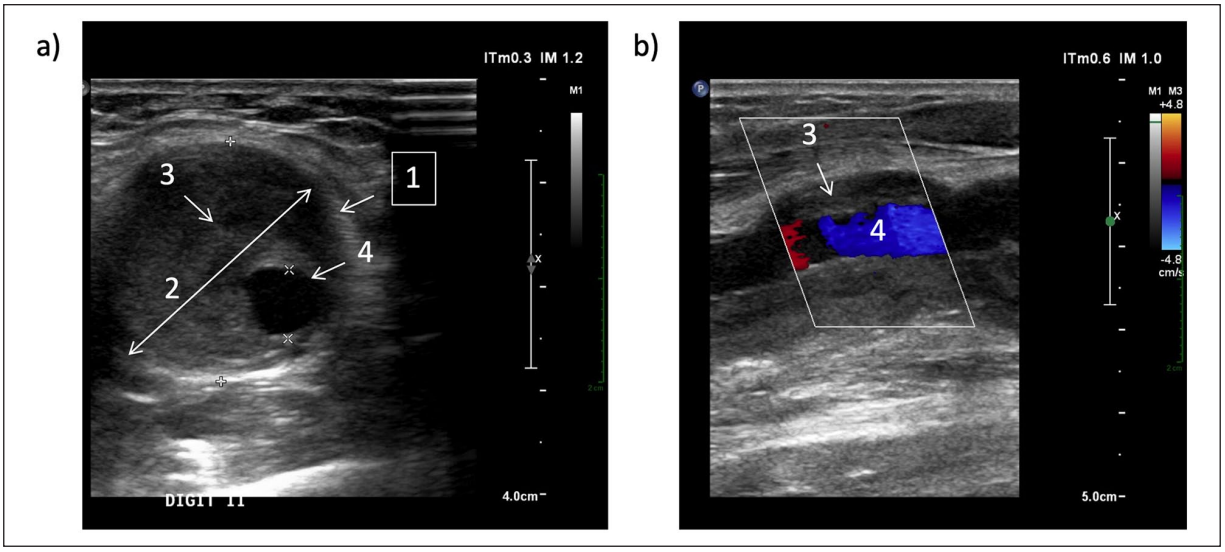

Figure 1. Transverse (a) and longitudinal (b) section by doppler ultrasound illustrating an aneurysmatic dilatation of the radial artery (1) with a diameter of 19 mm (2) including a partial thrombus (3) and a residual patent artery of 3.9 mm (4).

first and only symptom, leading to the diagnosis of a true aneurysmal degeneration of the complete left radial artery 5 years after radiocephalic fistula ligation. Because of the high risk of ischemic complications, we will discuss our treatment preference for a platelet antiaggregant combined with anticoagulation therapy rather than a surgical bypass which is considered as the standard treatment.

Case description

A 50-year-old male patient, suffering from Alport's syndrome, received a first kidney transplant in 1996 with subsequent ligation of a 3-year-old radiocephalic AVF at the left wrist in 1998. Because of chronic rejection, hemodialysis was restarted in 2012 by a newly created radiocephalic AVF at the left wrist. A second kidney transplantation was performed in December 2014 with subsequent uncomplicated ligation of his left, hypertrophic, 4-year-old, radiocephalic AVF in November 2015. In April 2019, the patient went to the emergency department with blue, painless fingertips after sleeping on his left arm in the absence of ischemic symptoms (e.g. pain, paresthesia, cramps). Since the ligation of the AVF, he had noticed a slight loss of strength in his left hand. Clinical examination demonstrated good pulses of the radial and ulnar arteries at the wrist. An ultrasound demonstrated a cephalic vein thrombosis of the left forearm, post-ligation of the AVF. On the middle third of the forearm, an intraluminal non occlusive thrombus was covering left radial arterial wall. The radial and ulnar artery demonstrated a satisfying blood flow and a preserved triphasic component. A conservative treatment was proposed to the patient, and because of complete absence of any symptoms at the vascular access consultation in June 2019, no other technical investigations or treatments were performed.

In August 2020, the patient presented with millimetric blue lesions on the left hand's fingers, disappearing when pressure was applied. Further clinical examination revealed a warm left hand with regular distal radial and ulnar pulses, and no palpable thrill. Graft function was excellent with an estimated glomerular filtration rate of 49 ml/min/1.73 m^2 under standard immunosuppressive therapy. The doppler ultrasound demonstrated a thrombus within the former draining cephalic vein of the forearm with a low flow in the cephalic vein. In the absence of a clear etiology of these lesions, no specific treatment was proposed and the patient was re-evaluated 2 weeks later. This time, despite the absence of ischemic symptoms and the disappearance of all the millimetric blue lesions on his fingers, the doppler ultrasound control showed an aneurysmatic dilatation of the radial artery, starting 5 cm distally from the elbow fold and ending 4 cm proximally to the wrist, with diverse parietal thrombi (Figure 1). The radial and ulnar arteries were patent with prograde flows. The largest diameter of the aneurysm was 19 mm, but due to multiple the intraluminal thrombi, the patent diameter was limited to 3.8 mm. An additional digital subtraction angiography (DSA) revealed an incomplete arterial palmar arch (Figure 2). The ulnar artery with normal anatomy furnished digital arteries for the fifth and fourth finger and the ulnar side of the third finger. The blood supply of the other digital arteries for finger I–III was exclusively provided by the radial artery. The small collateral arteries visible in the mid-portion of the forearm arose from an interosseous artery didn't provide any collateral flow to the distal radial artery at the wrist or palmar arch. The Allen's test has not been performed mainly because of the information by the DSA angiography, but also to avoid compression of the aneurysmal radial artery (risk of thrombus migration, clot formation, . . .).

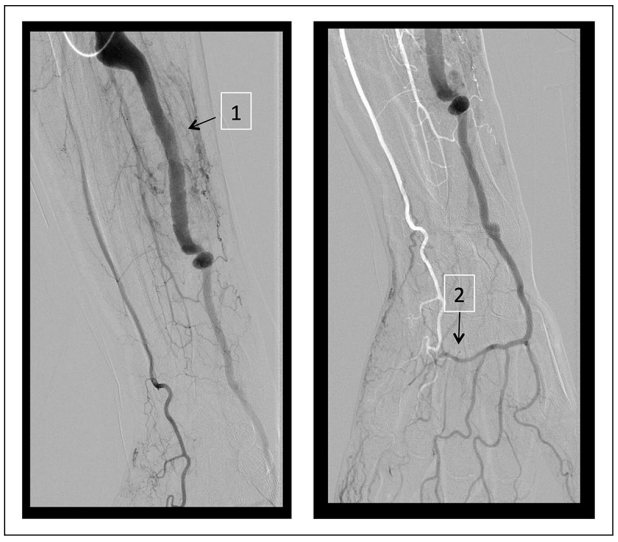

Figure 2. Digital subtraction angiography (DSA) of the left forearm illustrating the long aneurysmatic dilatation of the radial artery (1) and revealing an incomplete arterial palmar arch (2). The ulnar artery with normal anatomy furnished digital arteries for the fifth and fourth finger and the ulnar side of the third finger. The blood supply of the other digital arteries for finger I–III was exclusively provided by the radial artery. The small collateral arteries visible in the mid-portion of the forearm arose from an interosseous artery didn't provide any collateral flow to the distal radial artery at the wrist or palmar arch.

The patient was anticoagulated with a dose of nadroparine, 5700 IU/J (Fraxiparine®, Mylan EPD, Belgium) as soon as the ultrasound revealed this true aneurysmal degeneration in August 2020. After multidisciplinary discussion, medical treatment was preferred over surgical bypass because of the high risk of ischemic complications in case of thrombosis of the surgical bypass in the presence of an incomplete arterial palmar arch. The medical treatment consisted of a platelet antiaggregant (acetylsalicylic acid 100 mg 1x/day (Cardioaspirine®, Bayer, Belgium)) combined with an anticoagulant (apixaban 2.5 mg 2x/day (Eliquis®, Bristol Myers Squibb, Belgium) (off-label indication)). Six months after the start of this medical treatment, the patient was re-evaluated. He was completely asymptomatic and no progression of the aneurysmal degeneration of the radial artery was observed by ultrasound. Medical treatment was continued, and a clinical and radiological follow-up is foreseen every 6 months. Informed consent for patient information and images to be published was provided by the patient.

Discussion

Peripheral embolism as first and only symptom of a true aneurysmal degeneration of the complete left radial artery after radiocephalic AVF ligation is extremely rare, and therefore often late diagnosed. In this particular case, a medical treatment combining antiplatelet and anticoagulation therapy was preferred over the classical surgical bypass. The main reason for this choice was the existence of an incomplete arterial palmar arch resulting in a higher risk of ischemic lesions in case of complicated surgical bypass surgery (e.g. stenosis, thrombosis).

True aneurysmal degeneration of the inflow artery affects 4.5% of all AVF, generally 7–19 years after vascular access creation, even after fistula ligation, making it an unusual and late complication. Unlike our case, its preferred site of expansion is the distal part of the brachial artery, in the cubital fossa, probably due to higher turbulence proximal to the bifurcation.[6] One third of the patients have as clinical presentation an asymptomatic pulsatile mass, whereas others show symptoms due to medial nerve compression such as neuropathic pain and/or paresthesia, edema, ischemia, or compression pain from surrounding structures.[4,6] Distal embolization has been described in 30% of the cases, caused by a thrombus accompanying 83% of these aneurysms, but this remains very rare as first and only presenting symptom such as described in our case.[6] Rupture is extremely rare.[4,6]

The mechanism is still unclear but the increase in arterial flow following AVF creation probably produces an increase in wall stress and a decrease in wall thickness.[6,7] High blood flow could be a trigger to nitric oxide (NO) release from the endothelial cells, which can potentially lead to dilatation of the artery with high flow. This could injure internal and external elastic laminae fibers and lead to greater dilatation of the proximal arterial site of the AVF caused by reactive oxygen species liberated under stress from the wall, resulting in an overproduction of matrix metalloproteinases production (MMP-2 and MMP-9), which can degrade type IV and type V reticular collagen.[8] In addition, life-long immunosuppressive treatment with corticosteroids following kidney transplantation could be associated with late aneurysm degeneration, because of vascular remodeling affecting each layer of the arterial wall.[6,8] Furthermore, the luminal thrombus itself could entertain an inflammatory state and cause the fragmentation of elastic tissue.[8]

The diagnosis of true aneurysmal degeneration of the inflow artery in a functional or ligated AVF is based on physical and ultrasound examination, and most often angioCT/angiography imaging is performed during preoperative workup.[9] The definition of an arterial aneurysm is a localized and permanent diameter enlargement of at least 50% of the normal expected vessel diameter and all the three layers of the vessel are concerned by the enlargement.[4,10] Data about normal diameters are sparse.[11–13] Brown[13] reported a normal diameter of the radial artery may range from 2 to 3.5 mm.

Although there is no consensus in the literature, surgical treatment is the preferred treatment option for true aneurysmal degeneration of an inflow artery.[4,6]

Table 1. Literature overview of the surgical treatments of aneurysmal degenerations of inflow arteries in arteriovenous fistulas for hemodialysis.

Author	AVF type	Aneurysm location	Treatment	Surgical complications	Follow-up after surgery
Nguyen et al., 2001	RC	ABA (after fistula ligation)	Distal ligation of axillar artery, proximal end-to-side anastomosis with autologous GSV graft in subcutaneous route then radial artery anastomosis	No	NA
Schunn et al., 2002	RC	BA (after fistula ligation)	Aneurysmectomy and autologous bypass using a forearm median vein graft	No	24 months
Murphy et al., 2009	RC	BA (after fistula ligation)	Aneurysmectomy and autologous bypass using dilated (due to previous AVF) cephalic vein graft	No	12 months
Lam et al., 2009	BC	BC (still patent)	Fistula ligation, aneurysmectomy and brachial artery reconstruction with end-to-end arterial anastomosis	No	NA
Basile et al., 2010	RC	BA (after fistula ligation)	Aneurysmectomy and autologous bypass using GSV graft	NA	NA
Fejioo Cano et al., 2012	RC	RA (after fistula ligation)	Aneurysmectomy and ligation of RA	No	12 months
Dinoto et al.[14]	NA	BA	Aneurysmectomy and prosthetic bypass using PTFE graft	No	6 months
Garza et al., 2013	BC	BA (after fistula ligation)	Aneurysmectomy and autologous bypass with GSV graft	No	NA
Ferrara et al.[15]	RC	BA (after fistula ligation)	Aneurysmectomy and autologous bypass using ipsilateral basilic vein graft	No	12 months
Fernández et al., 2017	RC	BA (after fistula ligation)	Aneurysmectomy and autologous bypass using homolateral basal vein graft	NA	NA
Anastasiadou et al.[8]	RC	BA (after fistula ligation)	Aneurysmectomy and autologous bypass using ipsilateral branch of cephalic vein graft	No	24 months
Lee et al.[5]	RC	BA and RA (after fistula ligation)	Aneurysmectomy and autologous bypass with GSV graft	No	6 months
Salerno et al., 2020	RC	BA (after fistula ligation)	Aneurysmectomy and autologous bypass using GSV graft	No	6 months

ABA: axillo-brachial artery; BA: brachial artery; BC: brachiocephalic; GSV: great saphenous vein; NA: not available; PTFE: polytetrafluoroethylene; MRA: magnetic resonance angiography; RA: radial artery; RC: radiocephalic.

Indications for surgery are poorly defined: (1) an arterial diameter over 30 mm with a lower threshold of 20 mm acceptable for radial aneurysms, (2) significant complications (embolization, pain, neurological symptoms), and (3) patency with inflow and outflow arteries not being overly dilated, for it to be technically possible.[6,14] Surgery consists in resection of the aneurysm and reconstruction of the arterial continuity. This can be performed by direct suture between the proximal and distal artery segment ends if technically feasible or by the interposition of an autologous or prosthetic graft. An autologous graft (ipsi/contralateral arm vein or saphenous vein) is preferred over a prosthetic graft, because the latter is associated with a higher risk of infection and lower patency rates.[15] As illustrated in Table 1, the surgical treatment of aneurysmal degenerations of inflow arteries results in complete symptom relief. Mestres et al.[6] proposed a simple aneurysm excision for radial forearm aneurysms without vascular reconstruction if the following condition can be met: the ulnar artery should guarantee sufficient blood supply to the lower arm and hand, hence there must be a patent arterial palmar arch. Unfortunately for our patient, although he definitely met the criteria for a vascular bypass, this was not a viable option because of his incomplete arterial palmar arch (huge risk of distal ischemia in case of thrombosis after bypass surgery), so we opted for a medical treatment. The therapy consisted of a platelet antiaggregant combined to an anticoagulant, complemented by a clinical and ultrasound evaluation after 6 months and yearly afterwards.

In conclusion, peripheral embolism as first and only symptom of true aneurysmal degeneration of the arterial inflow vessel after AVF ligation is extremely rare, and therefore often late diagnosed. Surgery by vascular reconstruction is the standard treatment. However, this case revealed an aberrant anatomy (e.g. incomplete arterial palmar arch), so medical treatment by platelet antiaggregant combined with anticoagulant was preferred over surgery because of the higher risk of ischemic complications.

Author contributions

CM and TD wrote this work. JMP, LL, NK, AD, CL, MM, MS reviewed the manuscript.

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Informed consent

The patient has given written consent for the use of patient information in this report and its publication.

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