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# Incidental discovery of a long standing arteriovenous fistula after thrombectomy for acute lower limb ischaemia



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## ABSTRACT

**INTRODUCTION:** Arteriovenous fistula (AVF) is the abnormal connection between an artery and vein. Congenital AVF of the popliteal artery is very rare.

**PRESENTATION OF CASE:** 89 year old lady presented with right acute lower limb ischaemia. She had unilateral chronic venous hypertensive change in the right leg. Femoral embolectomy was performed. Backflow was achieved. Arteriotomy was closed. The patient's leg continued to deteriorate. She returned to theatre. On-table angiogram showed an occluded SFA. Thrombectomy was completed. SFA was patent but no blood flowed into the distal popliteal artery. A second on table angiogram revealed AVF between popliteal artery and vein. Dissection to the posterior aspect of the knee revealed the fistula. The vein was arterialized and enlarged. The AVF was ligated. Normal distal blood flow was achieved. Retrospectively we measured the leg lengths. Right leg was 3 cm longer than the left. The right leg circumference was 7 cm greater than the left. She reported chronic venous change from a young age. She did not report any history of trauma to the limb.

**DISCUSSION:** Popliteal artery to popliteal vein fistula is a rare. Trauma is the most common cause of popliteal AVF. Should the condition develop before closure of the epiphyses, there may be an increase in leg measurements.

**CONCLUSION:** We postulate that this case of AV fistula may be congenital due to discrepancy in leg measurements and unilateral chronic venous hypertensive change. Rarely persistent remnants of the embryonic sciatic artery can lead to arteriovenous anastomoses, which may be a possible aetiology.

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## 1. Introduction

An arteriovenous fistula (AVF) is the abnormal connection between an artery and vein that bypasses the capillary bed. They can be congenital or secondary to trauma or inflammation.<sup>1</sup> Congenital AVF of the popliteal artery is very rare, with few case reports in the literature.

## 2. Presentation of case

An 89-year-old lady was transferred to our hospital with progressive pain in her right calf radiating to her toes over the course of a few days. On presentation her right leg was acutely ischaemic. The patient reported paraesthesia and decreased power. The patient had a background history of Type 2 diabetes mellitus, hypertension and atrial fibrillation. She was not on therapeutic anticoagulation.

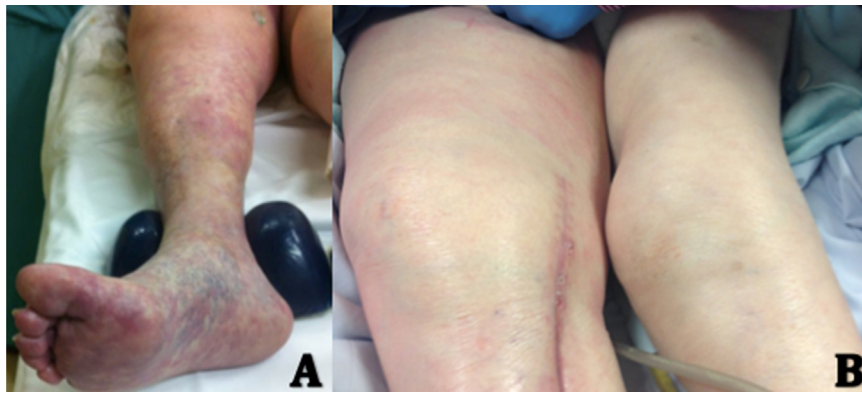
On examination of the right lower limb there was a blue discoloration of the foot. She had signs of chronic venous hypertension in the right leg (Fig. 1a). Her right leg appeared to have a greater diameter when compared to the left leg (Fig. 1b). Her right leg was cold on palpation. Capillary refill was delayed. No pulse or thrill was palpable at the site of the popliteal artery, nor were the distal pulses palpable. Sensation in the limb was intact, however power was reduced. The clinical impression was that of acute limb ischaemia secondary to lower limb embolism. As she had an acute presentation, she did not undergo any pre-operative radiology or vascular investigations.

The patient was started therapeutically on a heparin infusion and brought emergently to theatre for a femoral embolectomy. A small embolus was retrieved, however the Foley catheter could not pass beyond the level of the popliteal artery. As there was reasonable backflow, the arteriotomy was closed.

The patient's leg continued to deteriorate post-operatively, with loss of sensation and further loss of power over 48 hours. CT peripheral angiography revealed an occlusion in the distal Superficial Femoral Artery (SFA). There was a discrepancy between the leg diameters. The right femoral vein was 4.2 mm greater in diameter

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**Fig. 1.** (a) Signs of chronic venous hypertension. (b) Enlarged right leg compared to the left.

compared to the same point in the left leg (Fig. 2). The imaging also revealed signs of chronic arterial disease with vessel calcification.

The patient returned to theatre where an on-table angiogram showed an occluded SFA in the mid part. Below knee popliteal exposure was performed. Retrograde thrombectomy was completed through this incision to the SFA with subsequent angioplasty and stenting. Though the SFA was patent, no blood flowed into the distal popliteal artery. A second on table angiogram was performed through the groin incision revealed an unusual finding – a connecting fistula from the popliteal artery, filling the femoral vein, with lack of flow to the distal patent popliteal artery (Fig. 3).

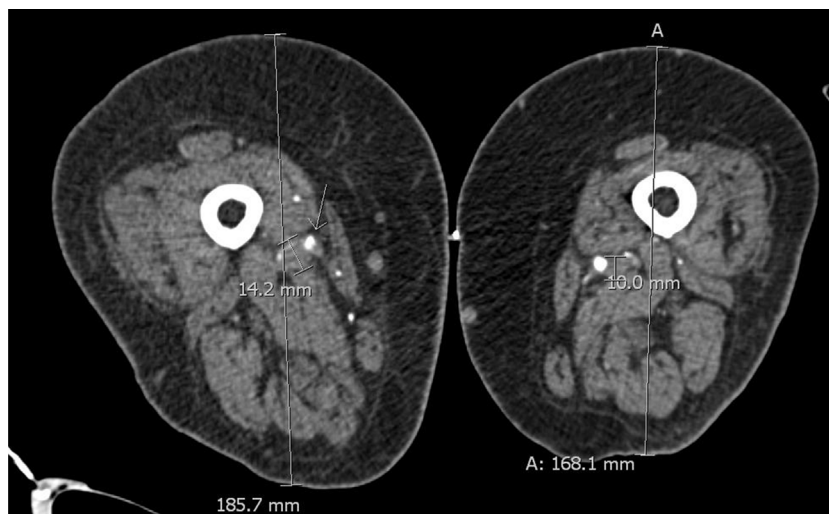
Further dissection to the posterior aspect of the knee revealed a popliteal arteriovenous fistula, with all the blood from the artery draining directly into the vein. The vein appeared arterialized and enlarged. There was no evidence of dissection or extravasation of blood in this area. There was a continuous machinery signal on Doppler assessment. The fistula underwent open ligation. Endovascular stenting was not performed as the fistula was at the level of the joint. Following closure of the fistula, with repair of the popliteal artery and vein, normal flow was achieved into the anterior tibial artery, which was the only available outflow vessel. Medial and lateral fasciotomies were performed.

As this patient had prolonged ischaemia, she was left with significant motor deficit in the limb. At six weeks follow-up, the fasciotomy sites were healing well with the aid of vacuum assisted closure. Sensation was intact in her right lower limb, as were her peripheral pulses. The motor function in the leg was improving.

### 3. Discussion

Popliteal artery to popliteal vein fistula is a rare condition. Causes include trauma, from injury to the knee,<sup>2</sup> or iatrogenically after surgery.<sup>3</sup> Our patient did not report any history of trauma. Infection and malignancy can also predispose to fistula formation. A congenital arteriovenous fistula of the lower limb can also be seen. It can occur in syndromes such as Klippel-Trenaunay syndrome, or spontaneously, as is the apparent case with our patient. Spontaneous fistula may occur at sites of chronic atheromatous arterial disease.<sup>4</sup>

Arteriovenous fistulae can present with discrepancy in leg diameter. Should the condition develop before closure of the epiphyses, there may be an increase in leg length.<sup>5</sup> In retrospective measurement of the lower limbs six weeks post-operatively, her right leg was 3 cm longer than the left leg. There was also a discrepancy in the



**Fig. 2.** CT peripheral angiogram prior to re-intervention shows discrepancy between diameters of the femoral veins in the right and left leg. It also showed a difference in radial leg diameter. There was significant calcification in the SFA (arrow).



**Fig. 3.** Intraoperative angiogram – retrograde flow of contrast from the popliteal artery to the popliteal vein.

leg circumference, with the right leg measuring 7 cm greater than the left. Asymmetric varicosities are often present due to venous hypertension. Ulceration may also be present. It can also present with cardiac decompensation. Our patient reported chronic venous hypertensive change in her right calf over her lifetime, with an enlarged right calf compared to the left side. Other general physical findings that may be present include a palpable thrill and increased skin temperature. There may be a continuous machinery murmur on auscultation. Distal pulses may be diminished. An interesting sign to look for includes “Bragman’s Sign”, whereby the heart rate drops after occluding the artery proximal to the fistula.<sup>6</sup>

The investigation of choice for diagnosis of arteriovenous fistula is duplex ultrasonography. The presence of an AVF is demonstrated by low-resistance flow in the enlarged afferent artery, high velocity flow spectrum at the fistula site and a high velocity, arterialized waveform in dilated, thick-walled draining veins.<sup>7</sup> Although not 1st line, CT angiography may also be used to diagnose AVFs.<sup>8</sup> Catheter angiography can also show the precise anatomy of AVFs. However, given the acute presentation of lower limb ischaemia, the patient did have a pre-operative angiogram, as urgent intervention was indicated. If indeed a pre-operative angiogram had been performed prior to initial operative management, there may have been a more favourable outcome, as the fistula may then have been detected on the primary intervention, which would have reduced the ischaemic injury to the limb.

Treatment of the AVF is indicated in patients who become symptomatic.<sup>9</sup> Those who are asymptomatic may be managed conservatively as spontaneous resolution is possible.<sup>10</sup> Surgical repair has long been the mainstay of treatment, however there are other methods including ultrasound-guided compression that may be useful in the treatment of early fistulas. Surgical approaches for repair of the fistula site can be open or endovascular. Endovascular approach may be suitable for patients with a high anaesthetic risk. Our patient was not an ideal candidate for endovascular repair as the fistula was located across a joint. Ultrasound guided compression therapy was also inappropriate as the fistula in this case was longstanding.<sup>11</sup> Newer techniques of repair include percutaneous coil of the AVF<sup>12</sup> or embolization with N-butyl-cyanoacrylate.<sup>13</sup>

#### 4. Conclusion

We postulate that this case of AVF may be congenital. The patient did not report any history of trauma to the knee. It is evident that the AVF is longstanding due to discrepancy in leg measurements. The aetiology of congenital popliteal AVF is unknown. Rarely persistent remnants of the embryonic sciatic artery can lead to arteriovenous anastomoses.<sup>14</sup> There are a few case reports of similar conditions in the literature. One case report described an incidental finding of a popliteal arteriovenous fistula, which appeared to be spontaneous, however that case the patient had bilateral venous hypertensive change.<sup>15</sup> More evidence is needed to ascertain the underlying aetiology of spontaneous arteriovenous fistulae.

Should a patient present with acute lower limb ischaemia and limb discrepancy or signs of asymmetrical venous hypertension, then routine intra-operative on-table angiogram should be performed after successful thrombectomy to exclude AV fistula that may be managed with immediately endovascular intervention, to avoid further intervention. Clinical awareness of this condition is required to anticipate the need for this.

#### Conflict of interest

None.

#### Funding

None.

#### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Author’s contribution

S. Beecher – writing the paper.  
M. Alawy – Paper review.  
M. Elbakar – Data collection.  
M. Tubassam – Data collection.

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