

Received: 2016.10.12
Accepted: 2016.11.16
Published: 2017.01.26

ISSN 1941-5923
© Am J Case Rep, 2017; 18: 90-95
DOI: 10.12659/AJCR.901925

Spontaneous Anterior Tibial Artery Avulsion and Tibio-Peroneal Trunk Transection Resulting into a Pseudoaneurysm: A Case Report

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
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Conflict of interest: None declared

Patient: Female, 53
Final Diagnosis: Spontaneous non-traumatic anterior tibial artery avulsion and tibio-peroneal trunk transection
Symptoms: Pain
Medication: —
Clinical Procedure: Autogenous popliteal-tibioperoneal trunk bypass
Specialty: Surgery

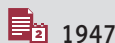
Objective: Rare disease
Background: Popliteal fossa pseudoaneurysms as a result of trauma are not uncommon. However, spontaneous pseudoaneurysms as a result of non-traumatic rupture of posterior tibial artery (PTA), anterior tibial artery (ATA), or tibio-peroneal trunk (TPT) artery segment are extremely rare. We report a case of spontaneous popliteal fossa pseudoaneurysm resulting from spontaneous avulsion of the ATA and transection of the TPT. Despite a thorough workup, no underlying associated disease was found. The extreme rarity of this disease presentation prompted us to report this case.

Case Report: A 53-year-old female patient presented with a 10-day history of sudden onset of non-traumatic left popliteal fossa pain and swelling. A popliteal fossa pseudoaneurysm was diagnosed by duplex ultrasound examination. Computed tomography angiography (CTA) was performed to confirm the diagnosis and to plan treatment. Surgical exploration revealed avulsion of the ATA and transection of the TPT leading to a pseudoaneurysm. Autogenous popliteal-tibioperoneal trunk bypass was performed with uneventful recovery.

Conclusions: A spontaneous popliteal fossa pseudoaneurysm caused by non-traumatic ATA avulsion and complete transection of TPT is extremely rare. Yet, it can be the cause of limb loss if not recognized early and treated promptly. Awareness by the medical community will help reduce the potential morbidity associated with this condition.

MeSH Keywords: Aneurysm, False • Aneurysm, Ruptured • Tibial Arteries

Full-text PDF: <http://www.amjcaserep.com/abstract/index/idArt/901925>



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Background

Popliteal fossa pseudoaneurysms are not uncommon, and are usually related to trauma [1,2]. A rare type of popliteal fossa pseudoaneurysm is spontaneous pseudoaneurysm which occurs in the absence of trauma and can be a diagnostic dilemma, and a possible cause of limb loss. Spontaneous lower limb pseudoaneurysms have been reported to occur with a spontaneous peripheral artery rupture associated with connective tissue disorders, defective collagen formation (as in Ehler-Danlos syndrome), infections, hypertension, and ruptured atheroma (Table 1). An extremely rare subtype of spontaneous pseudoaneurysms is associated with a spontaneous peripheral lower limb artery rupture in the absence of any obvious underlying pathology. In this context, Pollack et al. in 1950 were first to report a spontaneous rupture of the posterior tibial artery (PTA) with no obvious underlying disorder, which was successfully managed by ligation of the involved artery [3].

In this article we describe a case of a popliteal fossa pseudoaneurysm resulting from spontaneous anterior tibial artery (ATA) avulsion and tibioperoneal trunk (TPT) transection that was treated surgically. Despite thorough workup, no underlying associated disease was found. The extreme rarity of this disease presentation prompted us to report this case. Awareness by the medical community will help reduce the potential morbidity associated with this condition.

Case Report

A 53-year-old female patient presented to our vascular outpatient clinic complaining of a 10-day history of sudden onset

of left popliteal fossa pain, paresthesia, and swelling extending from the knee down to the foot. The patient specifically denied any history of trauma or symptoms of peripheral arterial disease. She had a history of rheumatic valvular heart disease for which she underwent mitral and aortic prosthetic valve replacement in January 2011. She was on oral anti-coagulant (warfarin 10 mg once daily). She had no history of diabetes mellitus, hypertension, or connective tissue disease. She was not a smoker. She was afebrile, and her other vital signs were within normal range. Local examination revealed a tense swollen left leg, associated with ecchymosis on the proximal anterolateral aspect of the leg and a tender pulsatile popliteal fossa swelling. Another pulsation was noted on the proximal anterior leg compartment at the site of the ecchymosis. The left foot showed delayed capillary refill. The PTA and dorsalis pedis pulses were not palpable; they had a weak signal on hand-held Doppler. The ankle-brachial-pressure index (ABPI) was 0.5 compared to 1.1 on the right side. The patient was unable to fully extend her left knee and her left foot dorsiflexion was limited due to pain.

An underlying arterial disease was obvious. Immediate duplex ultrasound revealed a pseudoaneurysm in the left popliteal fossa. Computed tomography angiography (CTA) confirmed the presence of a large pseudoaneurysm measuring (51.9×59.7 mm) located in the left popliteal fossa below the articular surface extending from the posterior leg compartment to the anterior compartment. Thrombosis of the proximal ATA and TPT was also noticed (Figure 1). However, there was blood flow to the left foot via arterial collaterals surrounding the knee to the PTA, ATA, and the peroneal artery. Transesophageal echocardiogram revealed absent valve vegetation, no significant valve dysfunction, and no evidence of infective endocarditis.

Table 1. Case reports of spontaneous rupture of peripheral lower limb arteries: summary of literature review.

No.	Author	Age	Gender	Artery involved	Possible cause	Management
1	Shinichi et al. [4]	52	Male	DPA ¹	Uncontrolled hypertension	Surgical repair
2	Salman et al. [1]	54	Female	TPT ² and ATA ³	Post-surgery of FA ⁴ aneurysm	Conservative treatment
3	Albert et al. [3]	38	Male	PTA ⁵	Unknown	Ligation of the artery
4	Akira et al. [8]	38	Female	PTA ⁵	Ehlers-Danlos syndrome	Surgical repair
5	P. Puppink et al. [9]	30	Female	Popliteal	Dislocation of unstable knee	Bypass surgery
6	George et al. [6]	67	Male	Popliteal	Salmonella bacteremia	Endovascular treatment
7	IS Sanhegan et al. [10]	21	Male	Popliteal	Distal femoral osteochondroma	Bypass surgery
8	Fabrizio et al. [11]	72	Male	SFA ⁶	Atherosclerotic plaque	Endovascular treatment
9	Andrea et al. [12]	86	Female	SFA ⁶	Atherosclerotic plaque	Endovascular treatment
10	Naoto et al. [13]	77	Male	SFA ⁶	Atherosclerotic plaque	Surgical repair

¹ DPA – dorsalis pedis artery; ² TPT – tibio-peroneal trunk; ³ ATA – anterior tibial artery; ⁴ FA – femoral artery; ⁵ PTA – posterior tibial artery; ⁶ SFA – superficial femoral artery.

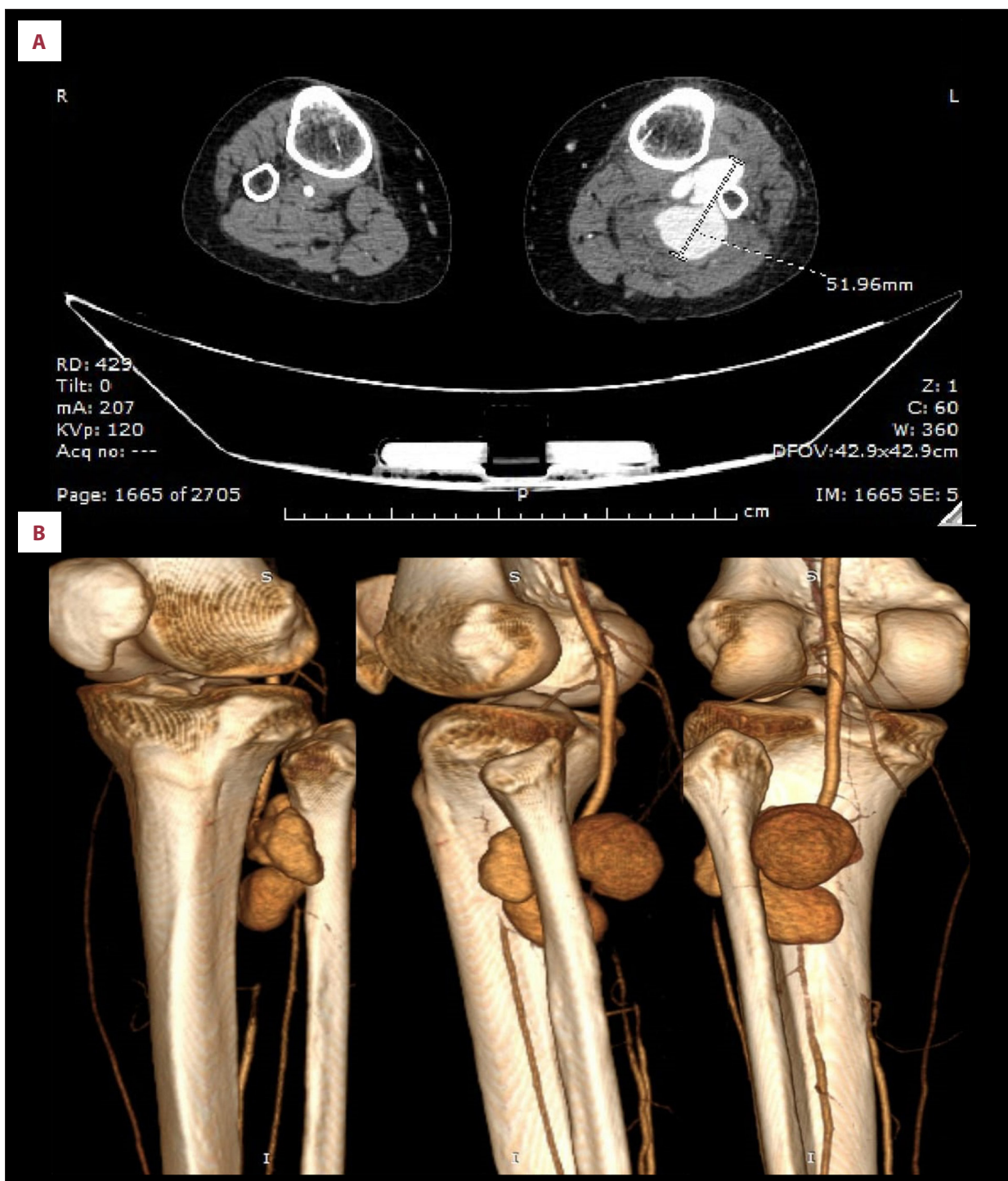


Figure 1. Pre-operative CTA of the left lower extremity: (A) Axial image showing pseudoaneurysm involving posterior and anterior compartment; (B) 3-D images reconstructed with different views.

Laboratory tests showed a white blood cell count of 19,000/mL (mainly neutrophils), a hemoglobin level of 13.8 g/dL, a platelet count of 298,000/mm³, and an international normalized ratio (INR) of 4.5. Blood culture and gram stain were negative. Urine analysis was negative for blood, proteins, and white blood cells.

The oral anticoagulant was withheld, and surgical intervention was delayed for 48 hours until optimization of INR was achieved (INR=1.8). Antibiotics were administered empirically, although no focus of infection was identified at that time. Open surgical repair was decided and an informed written consent

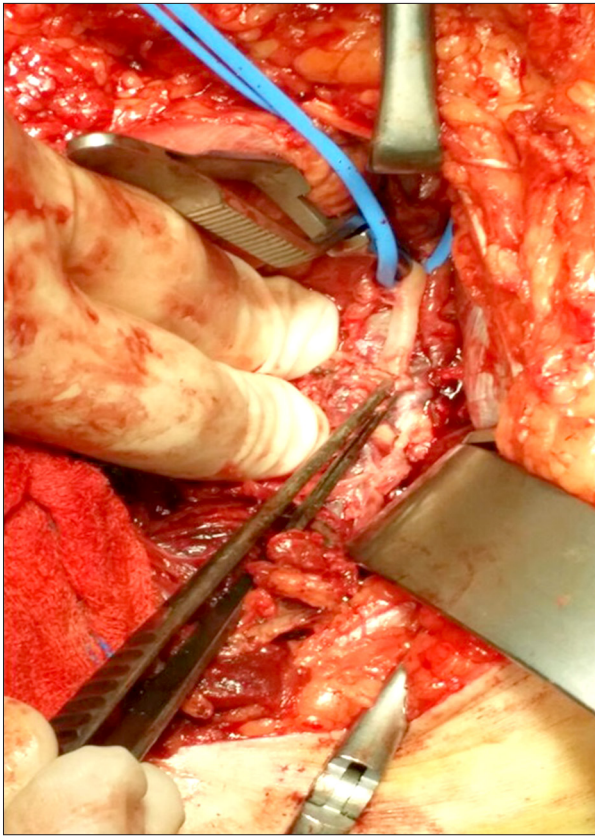


Figure 2. Intra-operative image shows the proximal end (tip of the forceps) of the left popliteal artery where the avulsion of the ATA and transection of the TPT occurred.

was obtained. Under general anesthesia and supine position, proximal control of the common, superficial, and deep femoral arteries was achieved through a groin incision. The popliteal fossa was explored through a medial surgical approach. The popliteal artery was easily controlled and the pseudoaneurysm was meticulously dissected. Evacuation of the pseudoaneurysm revealed avulsion of the ATA from its origin with complete transection of the TPT (Figure 2). The accompanying popliteal vein was thrombosed. The distal end of the avulsed ATA could not be identified as a result of contraction into the anterior compartment. The TPT was identified. It was retracted 4 cm distal to the proximal end and was occluded. The involved arteries did not show any atherosclerotic changes or any changes suggestive of vasculitis (i.e., no weak or fragile arterial walls were grossly recognized). There was no gross evidence of an accompanying extra-vascular bleeding or collection. This made the possibility of a spontaneous calf hematoma from anticoagulants and its subsequent infection as an underlying mechanism of the arterial rupture an unlikely event.

Distal embolectomy of the TPT using a 3F Fogarty catheter revealed a small clot, which was sent for histopathology. The

proximal arterial stump was beveled and a small piece of the artery was also sent for histopathology. A 5 cm segment of the greater saphenous vein from the contralateral limb was harvested and used to construct distal popliteal to TPT reversed interposition graft, using a 6-0 prolene suture.

Revascularization was successful with strong PTA and dorsalis pedis artery (DPA) signals on hand held Doppler. There was no back bleeding from the ATA and the operative field was dry; therefore, identification and revascularization of the ATA was judged to be unnecessary. Closure with surgical drain was done. The patient was put on a heparin infusion pump with partial thromboplastin time (PTT) target of three times the normal value.

Postoperative recovery was uneventful with excellent palpable distal pulses. Heparin infusion was replaced with oral anticoagulation on the second post-operative day. The patient started mobilization and physiotherapy on the third post-operative day. Orthopedic evaluation eliminated any knee instability as a possible underlying cause.

Pathological examination of the segment of the arterial wall showed non-specific inflammatory infiltration, which was not specific for arterial disease. Results of serologic tests for connective tissue disorders, which included anti-nuclear antibodies (ANA), scleroderma (Scl-70), and rheumatoid factor (RF), were negative.

The total hospitalization time was 16 days. A pre-discharge CTA was performed and revealed a patent interposition venous graft. There was good flow to the left foot via the TPT and PTA. The distal ATA was visualized due to intact foot arterial arches (Figure 3). Two months later, the swelling had dramatically improved and the patient's left extremity was fully functional without any sensory or motor deficit. The planned outpatient follow-up was set similar to patients with distal bypass due to occlusive arterial disease.

Discussion

Spontaneous (non-traumatic) rupture of a peripheral lower limb artery is a very rare condition [3]. It may result in the formation of a pseudoaneurysm, which is defined as blood extravasation that is walled off by the surrounding layers of connective tissue, but is still maintaining its communication with the arterial lumen through a neck [1]. Such spontaneous ruptures of a peripheral artery were reported to occur in association with atheromas, severe infections, endocarditis [1], rupture of small true aneurysm [3], defective collagen production in Ehlers-Danlos syndrome and Marfan syndrome, other connective tissue disorders [4], and uncontrolled hypertension.

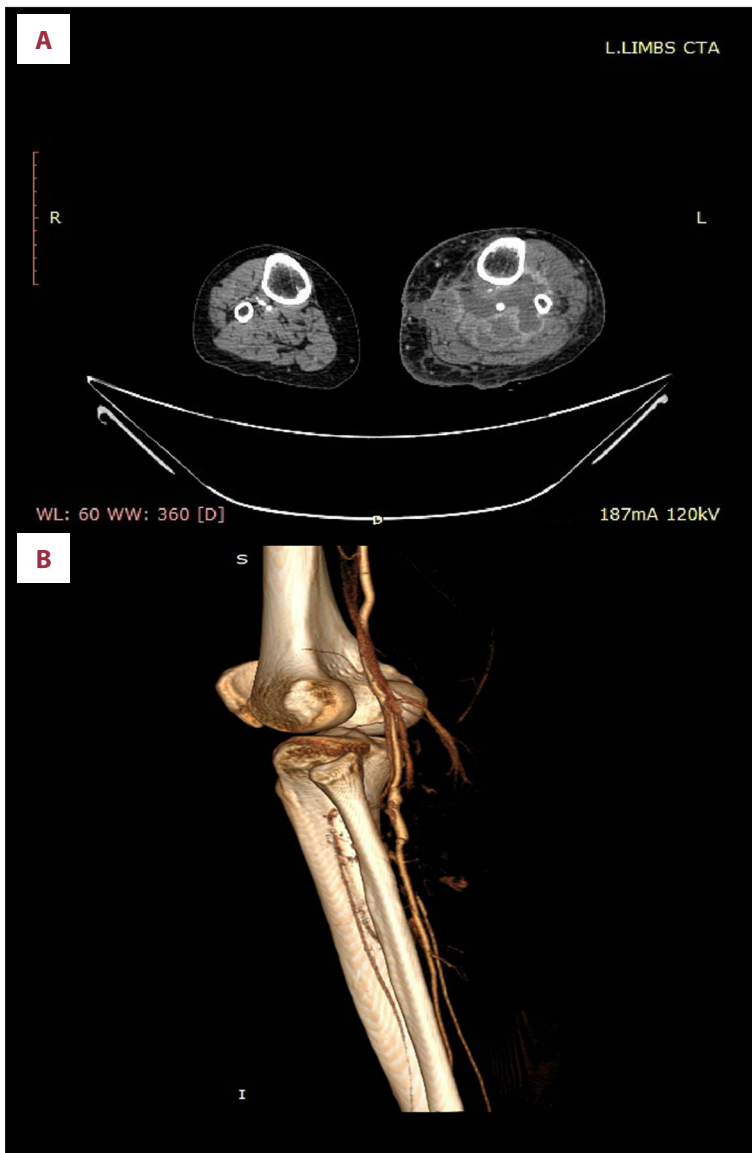


Figure 3. Post-operative CTA of the left lower extremity: (A) Axial image demonstrating patent interposition popliteal-PTT graft; (B) 3-D image reconstruction.

Table 1 summarizes the few case reports of spontaneous (non-traumatic) rupture of peripheral lower limb arteries in the literature. The majority of ruptures affected the superficial femoral artery followed by the popliteal artery. However, involvement of the distal run-off vessels (ATA and TPT) is extremely rare with only four reported cases that we could find in the literature (Table 1).

An exceedingly rare variant of spontaneous rupture of a peripheral lower limb artery is that occurring in the absence of an identifiable associated disease. In this context, Pollack et al. in 1950 were first to report a spontaneous rupture of the PTA due to an unknown cause that was treated by ligation of the affected artery [3]. Our patient had spontaneous avulsion of her ATA with complete transection of the TPT, leading to the formation of a spontaneous non-traumatic pseudoaneurysm.

Despite thorough workup, an underlying mechanism that might explain the avulsion of the ATA and transection of the TPT could not be identified. We could find no previous similar report with a similar anatomical peculiarity. Although this patient had prosthetic mitral and aortic valve replacement, she did not meet the criteria for diagnosis of infective endocarditis [5]. Transesophageal echocardiogram revealed absent valve vegetation with no significant valve dysfunction. The possibility of *Salmonella* endarterial infection was ruled out by negative blood cultures, and the absence of an antecedent clinical picture suggestive of bacteremia [6]. One etiology that has been described as a cause of pseudoaneurysms is connective tissue disorders [4]. However, our patient did not meet the diagnostic criteria for type-IV Ehlers-Danlos syndrome or Marfan syndrome or any other connective tissue disorders. No evidence of a small true arterial aneurysm at the point of rupture was

found during surgery. Histological examination of the affected artery and the thrombi did not suggest any evidence of vasculitis or a specific disease entity.

Pseudoaneurysms can be asymptomatic [1], or can be associated with the characteristic findings of a pulsatile mass, a palpable thrill, and an audible to-and-fro murmur [2]. When left untreated, pseudoaneurysms can be complicated by thrombosis, distal embolization, compression of the nearby vital structures, or rupture [2]. Since morbidity of emergent operations far exceeds that of elective repairs, early diagnosis and treatment are the standards of care [2]. Timely intervention in our case was associated with a preferable outcome leading to limb salvage.

The diagnosis of pseudoaneurysms in the popliteal fossa can be made by duplex ultrasound. However, CTA, magnetic resonance angiography (MRA), or digital subtraction angiography (DSA) are necessary to confirm the diagnosis and to show the anatomy in order to plan for the intervention [1]. In our case, CTA was useful in clarifying the anatomical aspects of the lesion, and in directing the surgical approach.

Regardless of the etiology, the principles for pseudoaneurysm repair are the same. It must be excluded from the circulation, and arterial circulation must be restored [2]. Although simple ligation of a single run-off ruptured vessel is a safe strategy [4], some arterial territories, such as at the popliteal level, might not tolerate the ligation [7]. In our case, involvement of the three run-off vessels mandated restoration of vascular continuity of at least one vessel.

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The majority of lower limb pseudoaneurysms are located in the femoral region, and endovascular treatment could be a suitable option. However, because of the anatomical limitations to the popliteal fossa, unavailability of the proper size Viabahn stent graft in our center, and the advantage of open surgery to perform Fogarty embolectomy to the distal run-off vessels, we decided to perform an open surgical repair. The median surgical approach to the popliteal fossa was chosen because of our familiarity with this approach compared with the posterior midline approach. The proximal femoral control was performed as a safety measure, in case the pseudoaneurysm ruptured during dissection before controlling the popliteal artery and the involved vessels.

The decision not to identify and revascularize the ATA was made because of satisfactory results after successful revascularization of the TPT, the absence of back bleeding from the ATA, and the anticipated difficulty to reach the distal stump of the ATA from the adopted medial surgical approach.

Conclusions

This case study reports on the successful management of a spontaneous popliteal fossa pseudoaneurysm caused by non-traumatic ATA avulsion with complete transection of TPT. Spontaneous pseudoaneurysms as a result of non-traumatic rupture of the infra-popliteal territory arteries are extremely rare. Yet, they can be the cause of limb loss if not recognized early and treated promptly. Awareness by the medical community will help reduce potential morbidity associated with this condition.