American Journal of Case Reports

e-ISSN 1941-5923
© Am J Case Rep, 2023; 24: e937113
DOI: 10.12659/AJCR.937113

Received: 2022.05.01
Accepted: 2022.10.13
Available online: 2023.01.16
Published: 2023.02.07

A 54-Year-Old Man Who Developed a Femoral Pathologic Fracture from a Giant Popliteal Artery Pseudoaneurysm 7 Years After Ligation and Bypass of a Popliteal Artery Aneurysm: A Case Report and Literature Review

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Financial support: None declared
Conflict of interest: None declared

Patient: Male, 54-year-old
Final Diagnosis: Femoral fracture due to gigantic pseudoaneurysm secondary to the rupture of a previously ligated and bypassed popliteal artery aneurysm
Symptoms: Swelling and pain
Clinical Procedure: —
Specialty: Orthopedics and Traumatology • Surgery

Objective: Unusual clinical course

Background: This case report describes a giant pseudoaneurysm that grew in size during the years following surgical treatment of a popliteal artery aneurysm, eventually causing a femoral fracture. Bone fractures secondary to vascular injuries are rarely described in the literature.

Case Report: A 54-year-old man underwent surgical ligation and bypass for left popliteal artery aneurysm. Seven years later, he suffered a left distal femur pathologic fracture surrounded by a giant soft-tissue mass. The patient came to us with a diagnostic hypothesis of angiosarcoma from another hospital at imaging evaluation. After computed tomography angiography (CTA) and angio-magnetic resonance imaging (MRI), we made a diagnosis of femoral pathologic fracture caused by a giant pseudoaneurysm of a treated popliteal artery aneurysm refilled by an aberrant anterior tibial artery (IIA2, Kim classification). We performed excision of the mass and open reduction and internal fixation, with anatomic plate, of the fracture. Fracture healing and good functional outcome were observed at follow-up.

Conclusions: A possible complication of surgical treatment of popliteal artery aneurysms is refilling of the excluded aneurysm due to collateral blood flow or, such as in the present case, aberrant vessels. Therefore, the knowledge of anatomical variants of the vessels is important in surgery. Follow-up evaluation after surgery is advisable and a growing mass should be further investigated with an angio-CT scan. In case of a non-pulsating soft-tissue mass causing pathologic bone fracture, a biopsy is mandatory to exclude malignancy.

Keywords: Anatomic Variation • Aneurysm, False • Femoral Fractures
Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/937113

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Background

Aneurysmal or pseudoaneurysmal vascular lesions after traumatic events or orthopedic surgical procedures are commonly described in the literature [1-4], but pathologic fractures secondary to aneurysms or pseudoaneurysms are less frequently reported [5]. Occasionally, the clinical course and pattern of the vascular lesion may be confused with a soft-tissue sarcoma [5-7]. A true aneurysm is defined as a focal dilation, greater than 50% of the normal caliber of the vessel with involvement of all 3 layers of the vessel wall [8,9].

The femoral artery continues in the popliteal fossa as the popliteal artery; the latter runs in an oblique direction, from proximal to distal and from medial to lateral, and in 94.7% of cases it ends at the lower margin of the popliteal muscle and branches into the anterior tibial artery and tibio-peroneal trunk, which are the 2 terminal branches of the popliteal artery. Anatomical variation of the popliteal artery's branches has been previously described [10,11].

Popliteal artery aneurysm represents 70% of all peripheral arterial aneurysm, and the most common surgical treatment is ligation of the popliteal aneurysms and femoro-popliteal bypass with the great saphenous vein [12,13]. To promote aneurysm thrombosis, the aneurysm should always be ligated both proximally and distally as close as possible to the sac [13]. Despite this procedure, 30% of aneurysms continue to enlarge if collateral blood flow into the aneurysm sac persists, in a scenario comparable to a type II endoleak with endovascular aneurysm repair [14-16].

The aim of this report is to describe an exceptional case of a giant pseudoaneurysm that increased in size during the 7 years following surgical treatment of a popliteal artery aneurysm, eventually causing a femoral fracture. We also highlight the importance of differential diagnosis and knowledge of vascular anatomy and its variants.

Case Report

A 54-years-old man was admitted to our Orthopedic Oncology Unit for a pathologic left femoral metaphyseal fracture, with a mass on the left thigh. The swelling of thigh first appeared 4 years after ligation and bypass surgery of a popliteal aneurysm and grew slowly for 3 years before the fracture occurred.

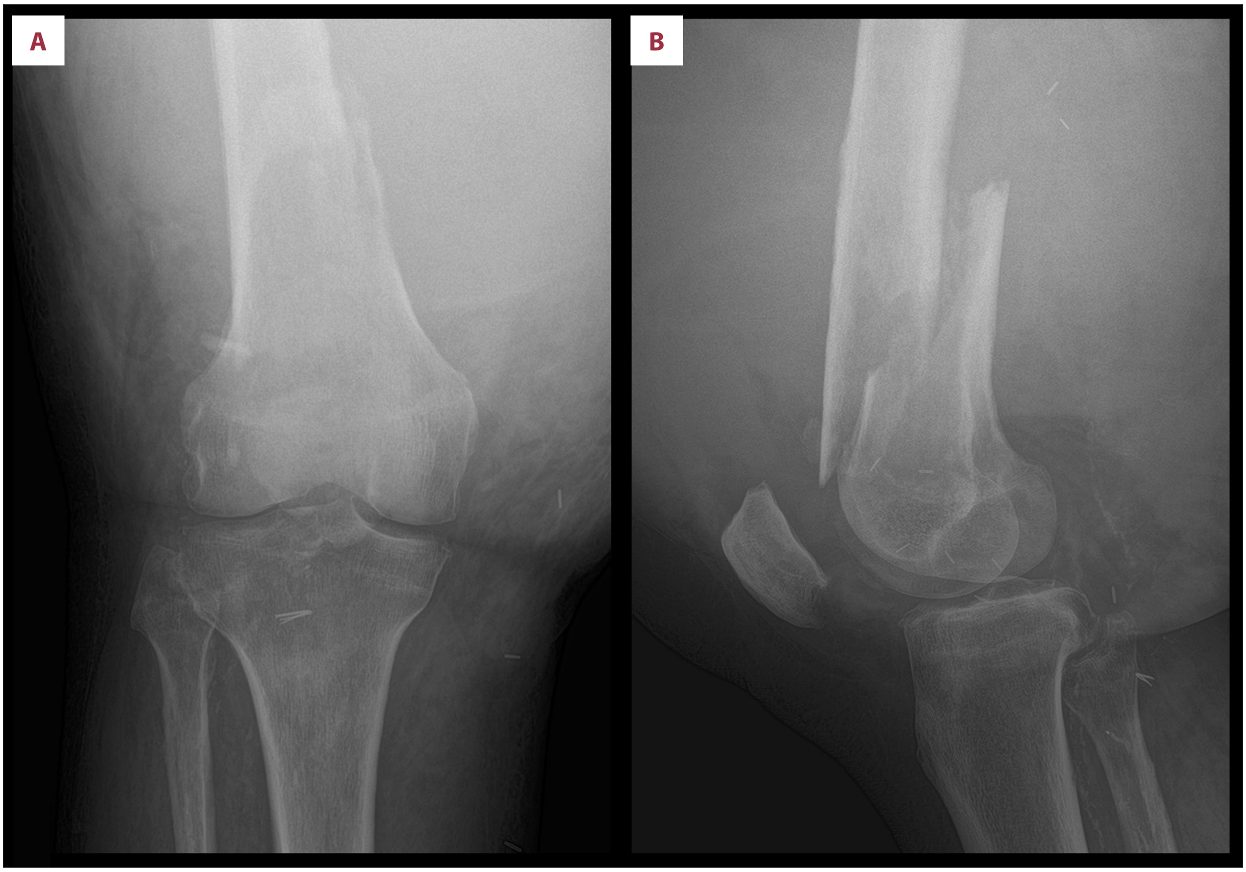

Figure 1. Preoperative X-ray of the distal femur: pathologic fracture (AO 33A2.2). Cortical bone scalloping is evident. (**A**) Antero posterior view; (**B**) Lateral view.

Figure 2. Preoperative clinical image. The giant soft-tissue mass (92 cm maximum circumference) is visible with the lower limb completely extra-rotated. Ulceration is visible on the antero-medial side of the thigh.

He was referred from another hospital, where primary evaluation with plain X-rays (**Figure 1**) and computed tomography (CT) scan had been performed. Additionally, ultrasound-guided imaging and histology were negative for malignancy. On the X-ray and CT images, the cortex of the 2 fracture abutments, surrounded by the mass, appeared to be thinned, with scalloping, pressure erosion, and sclerotic margins. These radiographic signs were all suggestive of a pathological fracture with extensive periosteal reaction.

However, based on the size of the mass and the pathologic fracture, a provisional diagnosis of angiosarcoma was made, and after staging with total body CT scan the patient was referred to our center. His medical history included smoking (75 packs/year), arterial hypertension, gout, and a bilateral popliteal aneurysm.

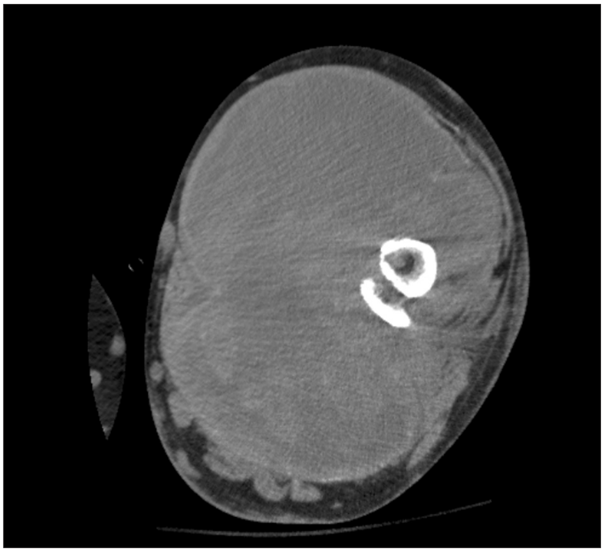

Figure 3. Axial computed tomography (CT) scan showing the soft-tissue mass almost circumferentially surrounding the fractured bone and extending into the anterior, posterior, and medial compartments.

Left femoro-popliteal in situ internal saphenous vein (ISV) bypass with medial approach was performed in 2011 for asymptomatic left popliteal artery aneurysm. The medical record reported a bypass revision for acute ISV thrombosis occurred the second day after the bypass procedure. The presence of a painless swelling of the left thigh was observed during the duplex ultrasound exams performed 4 years later; the first diagnostic interpretation of the mass was a lymphatic dysfunction and no surgical treatment was offered.

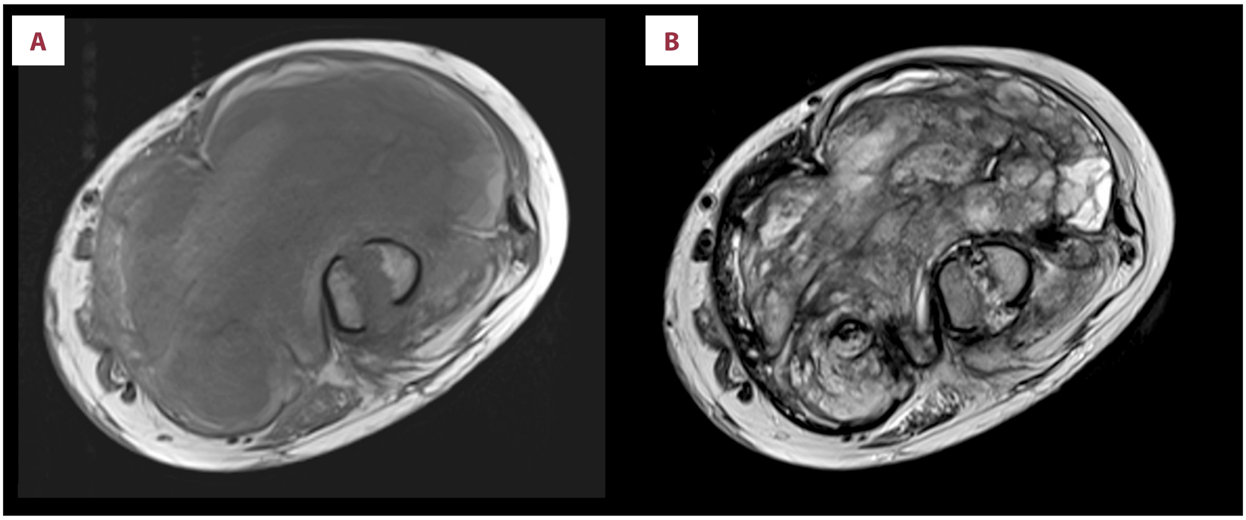

Figure 4. (**A**) T1-weighted magnetic resonance imaging (MRI) on axial plane showing the mass with an isointense aspect to the muscle, visible at the ipsilateral limb with a hyperintense ring. (**B**) T2-weighted MRI image on axial plane showing greater structural inhomogeneity.

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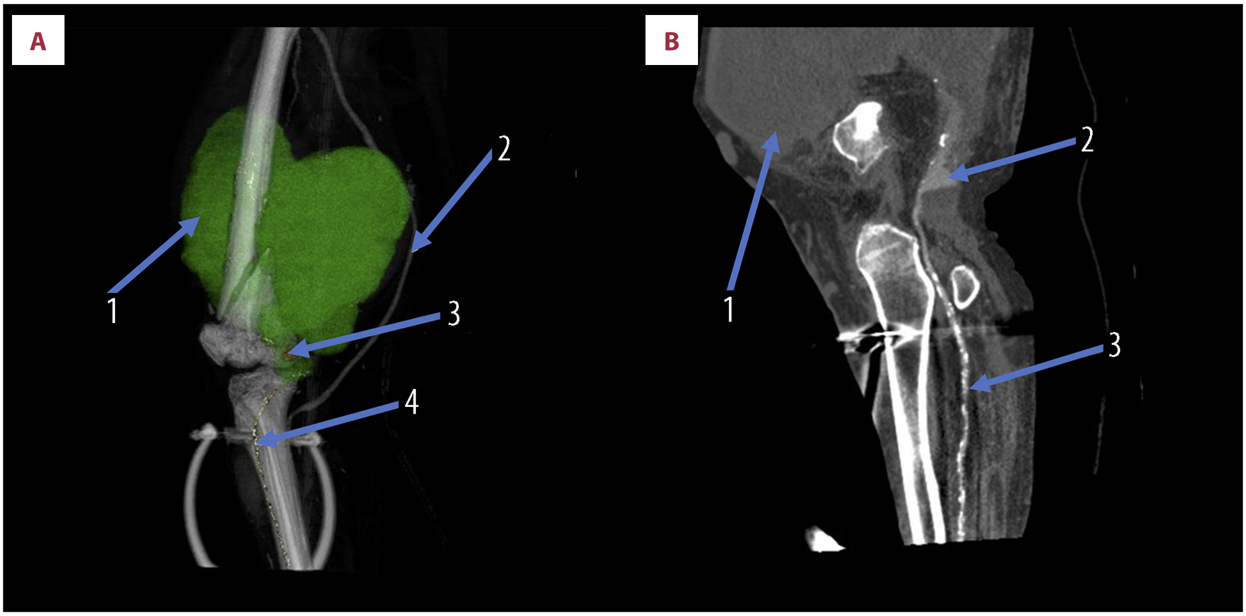

Figure 5. (**A**) 3D volume-rendering CT scan of the left tight, showing: (1) Giant pseudoaneurysm (green color). (2) Great saphenous vein bypass graft (GSV bypass). (3) Popliteal artery aneurysm (PAA). (4) Anterior tibial artery (ATA). (**B**) Sagittal CT scan view showing: (1) Giant pseudoaneurysm. (2) Popliteal artery aneurysm (PAA). (3) Anterior tibial artery (ATA).

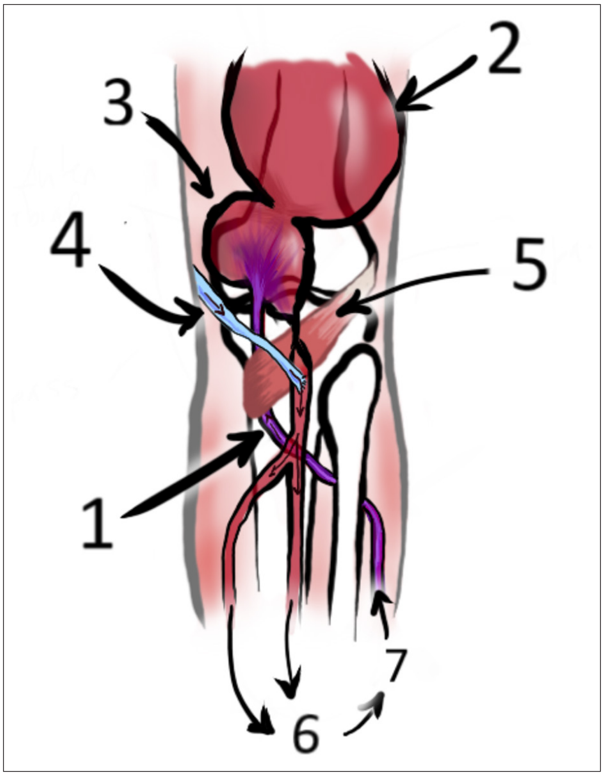

Figure 6. Descriptive and schematic drawing of the refueling mechanism of popliteal aneurism by ATA. (Drawn by Dr. Melani and used with permission). (1) Anterior tibial artery arising above the joint line, taking a medial path anterior to the popliteal muscle (Type 2A-2 of Kim Classification). (2) Pseudoaneurysm. (3) Legated popliteal aneurysm. (4) Femoro-popliteal bypass. (5) Popliteal muscle. (6) Foot/ankle circulation. (7) Retrograde blood flow reperfusion of the ligated aneurysm trough the anterior tibial artery.

When the patient came to us, the first clinical evaluation revealed a huge swelling of the left thigh (92 cm maximum circumference). The overlying skin was tense and a small ulceration was present on the anterior distal part of the thigh (**Figure 2**). The mass was firm on deep and superficial planes, with uniform solid consistency and no transmitted pulsatility was present. Both the ISV graft and the posterior tibial artery were pulsatile. No signs of sciatic nerve compression were detectable.

CT angiography (CTA) and contrast-enhanced magnetic resonance imaging (MRI) were performed (**Figures 3, 4**) at the Radiology Department of our hospital. The imaging showed a giant mass surrounding the femur for 270°, in continuity with a partially thrombosed aneurysm of the popliteal artery. The popliteal artery aneurysm had a diameter of 4 cm proximal and 3 cm distal to the knee joint, with clear patency of the ISV bypass graft.

Further evaluation of the CTA exam, performed and processed using dedicated software (Tararecon), showed the retrograde reperfusion of the aneurysmal sac from the anterior tibial artery (ATA) with type II-A2 anatomical variation (Kim et al classification) (**Figures 5, 6**) [17]. What was previously described as an undefined mass was eventually identified as a giant thrombosed popliteal aneurysm (estimated

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Figure 7. Intraoperative image, showing the organized hematoma removed from the pseudoaneurysm (3.6 kg) on a Mayo table.

volume 5923 cm^3), grown in the thigh due to the slow blood reperfusion from the ATA.

The patient underwent surgery and the vascular surgeon was involved to ensure the integrity and patency of the femoro-popliteal bypass. A medial incision based on the previous one was performed. A bulky mass appeared immediately beneath the quadriceps muscle. The macroscopic aspect of the mass suggested an organized hematoma. After removal, the tissue was weighted (total 3.6 kg) (**Figure 7**) and sent for histologic examination. Distally, in the popliteal region, the mass presented continuity with the popliteal aneurysm, which was promptly removed, and the resulting bleeding was controlled with transfixion suture of the ATA ostium.

A second lateral approach to the thigh was performed to proceed with open reduction and internal fixation of the femoral fracture. The cortical bone presented extended periosteal reaction, and progressive growth of granulation tissue with cortical scalloping was the cause of the pathologic fracture. Periosteal and intramedullary samples were sent for histology.

Reduction and internal fixation were done with two 3.5-mm lag screws and anatomic LISS plate with 4.5-mm angular stability screws (Synthes, De-Puy).

A vacuum-assisted closure (VAC) device was placed in the medial wound of the thigh, partially replacing the defect previously containing the mass. The lateral incision was closed on suction drains. A week later, the VAC device was removed and the medial wound was sutured.

Preoperative laboratory data showed normal indices of renal function with a slight hyponatremia of 132 mEq/l and hemoglobin values of 8.4 g/dL. Given the duration of the surgery and the risk of bleeding, it was considered useful to transfuse the patient preoperatively with 2 units of whole blood, achieving a hemoglobin value of 10.4 g/dL.

However, post-surgical anemia occurred (7.3 g/dL), which required the transfusion of 4 units of concentrated red blood cells, with stabilization of hemoglobin values above 8 g/dL.

The second day after VAC removal, the patient started rehabilitation treatment with continuous passive mobilization of the knee and walking with toe-touch bearing on the operated limb.

Thirty-days after discharge, the wounds were completely healed without complications. The hardware was in place, the articular range of movement was complete, the circumference of the thigh was 68 cm, and the pathology report confirmed the vascular origin of the lesion.

Five months after discharge, the fracture was completely healed, radiographically and clinically. The patient could walk without crutches, going back to his normal activities. One-year radiographic follow-up showed complete bone remodeling (**Figure 8**).

The follow-up with duplex ultrasound of femoro-popliteal bypass showed good patency of the graft, excellent perfusion of leg's vessels, and no sign of bleeding or pseudoaneurysms in the popliteal region.

Discussion

This case report is an example of an arterial anatomical variant influencing the outcome of the femoro-popliteal bypass procedure. As shown in the 3D volume-rendering reconstruction obtained from CTA (**Figure 4**), the origin of the type II-A2 ATA was further dislocated in the anterior region of the popliteal artery aneurysm. This challenging anatomy and the highly calcified aspect of the ATA might have led the vascular surgeons who performed the femoro-popliteal bypass not to ligate the ATA, expecting its occlusion after the popliteal artery aneurysm ligation. Nevertheless, the ATA remained patent and, due to the atypical anatomy, the calcification, and the slow blood flow, it is also reasonable to assume that the patency of the ATA remained unseen at the Doppler ultrasound (DUS) follow-up, and the mass was misdiagnosed as the result of a lymphatic disorder.

Considering the intraoperative findings, it is reasonable to assume that the popliteal artery aneurysm underwent asymptomatic rupture in its superior region and the inextensible layer offered by the muscular fascia prevented massive haemorrhagia, leading to formation of a giant pseudoaneurysm.

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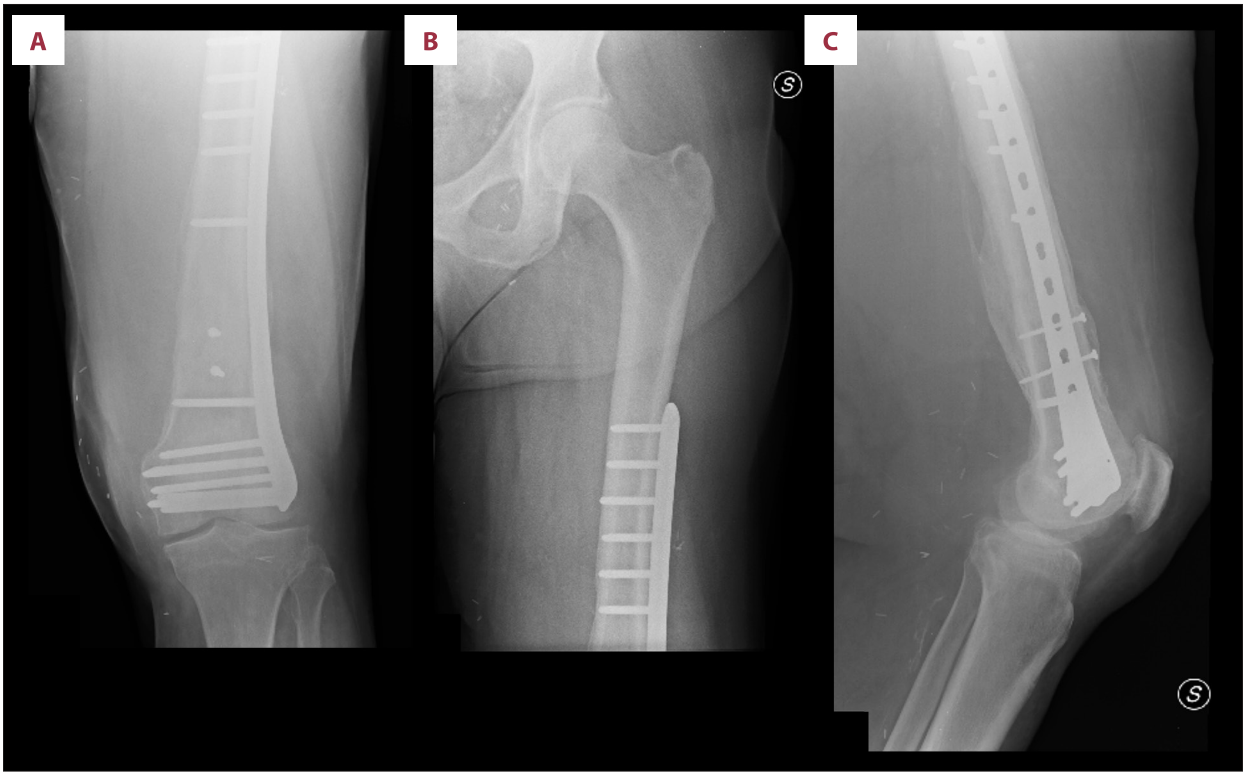

Figure 8. Radiographic control at 1 year, demonstrating complete fracture healing and remodeling. (**A**) Antero posterior (AP) distal view; (**B**) AP proximal view; (**C**) Lateral view.

Bone erosion and spontaneous pathologic fractures associated to extrinsic compression of pseudoaneurysms has been described in literature for aortic and post-traumatic peripheral vessels pseudoaneurysms [5,18,19].

Sing et al presented a case of a 30-year-old man who sustained a penetrating trauma of the lateral aspect of the proximal right leg; 5 months later, swelling and pain appeared over the proximal leg. An X-ray investigation was performed in a tertiary hospital with a provisional diagnosis of soft-tissue neoplasm causing a pathological fracture of the proximal fibula with signs of erosion and scalloping of the margins of the fracture. Further evaluation with a selective angiogram confirmed a pseudoaneurysm of the posterior tibial artery as the cause of pain and bone erosion [5]. This case in literature is the one closest to our report – the fracture was due to a pseudoaneurysm, initially diagnosed as soft-tissue cancer, but the site, timing of the fracture, pseudoaneurysm's cause, and dimension are different. The pseudoaneurysm may have grown more in the thigh than in the proximal tibia due to the greater dimension of the muscular compartment, giving less pain and bone erosion only when it had already reached gigantic size.

About 20 years before the report by Singh et al, Gantz et al described a case a non-penetrating trauma that caused a pseudoaneurysm of the anterior tibialis artery, that did not produce a fracture but caused erosion of the corticals of the tibia and fibula [6].

Prete et al described a 76-year-old man with progressive, disabling lower-back pain. Twenty-two months before, he underwent an elective aneurysmectomy and aortic bifurcation graft replacement for a large abdominal aneurysm. CT scanning of the abdomen and lumbar spine revealed a destructive process involving the centrum of the third lumbar vertebra with marked anterior erosion of the cortex and cancellous bone. After intravenous contrast medium was injected, marked thrombosis was noted around the true lumen of the aorta, indicative of a large false aneurysm extending into the left psoas muscle anteriorly. At surgical exploration, the aortic graft was found to have separated from the wall of the aorta at the site of its proximal anastomosis, and a large false aneurysm filled with thrombus had formed and eroded the third lumbar vertebra. That paper reported a rare complication caused by an anastomotic pseudoaneurysm of a previous vascular intervention with no pathological fracture but with similar bone erosion process [18].

Akiyama et al described a case of a lumbar vertebrae fracture due to an inflammatory abdominal aortic aneurysm, in which the fracture occurred because of occlusion of the lumbar arteries due to vasculitis [19].

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In our case, considering the clinical presentation, a differential diagnosis with soft-tissue sarcoma was necessary. In particular, chronic lymphedema and aneurysm surgery have been associated with angiosarcoma [20,21]. The popliteal artery is the site of 70% of all peripheral arterial aneurysms. The latter affects women less frequently than men, (women-to-men ratio=20: 1) reaching a maximum prevalence peak of 1% in men aged 60-80 years [13]. The natural course of popliteal artery aneurysm includes a high incidence of acute and chronic thromboembolic complications. Due to chronic thromboembolism, around 50% of patients with popliteal artery aneurysm have intermittent claudication, rest pain, blue toe syndrome, or acral necrosis [22].

Approximately 30% of untreated patients with popliteal artery aneurysm present acute thrombosis and distal embolization, with amputation rates of up to 20%. Another limb-threatening complication of popliteal artery aneurysm is rupture, occurring in approximately 2% of patients with this condition. In contrast, early elective treatment of popliteal artery aneurysm with open surgery is associated with limb salvage rates of 86-99% and primary patency rates of 66-86% at 5 years [13,23,24].

Elective treatment consists of aneurysm ligation and subsequent bypass with an autologous vessel, which is more frequently performed than autologous IVS [25]. However, this procedure does not always completely exclude the blood flow and the maintained supplied aneurysm occasionally enlarges until it ruptures [26].

A false aneurysm (FA) or pseudoaneurysm is defined as an encapsulated pulsatile hematoma in communication with a perforated artery [2]. They are commonly a consequence of vascular lesions in traumatic bone fractures, tumoral lesions, or iatrogenic surgical damage. A recent review of the literature identified the most frequent causes of popliteal aneurysm as follows, in order of decreasing frequency: femoral exostoses (63%), prosthetic surgery (14%), knee arthroscopy (11%), direct knee trauma (10%), and distal femoral osteotomy (2%) [2].

The popliteal artery is a continuation of the femoral artery that crosses the popliteal fossa, splitting into the ATA artery and the tibio-peroneal trunk (TPT), which further divides into the posterior tibial (PT) and peroneal (PR) arteries. The current classification of popliteal artery variation is as described by Kim et al in 1989 and consists of 3 primary types, with 10 subtypes [17]. Type I, the most common, encompasses all divisions of the popliteal artery below the level of the knee joint. Type II, the "high divisions," includes all divisions of the popliteal artery at or above the level of the knee joint. In particular, in type II-A2 (prevalence estimated at 0.2-0.6%), the ATA goes inward after its origin, taking a medial course, and subsequently goes lateral posterior to the popliteal muscle, crossing anteriorly the TPT and then continuing in the normal location. Type III includes all cases of hypoplasia or aplasia of any of the branches [27,28].

Conclusions

Vascular lesions that cause pathological fractures are not common and they can clinically and radiographically mimic soft-tissue tumors. For this reason, an accurate imaging study is essential to define the vascular origin. A biopsy and histologic diagnosis are mandatory for differential diagnosis with soft-tissue sarcomas in case of a giant, non-pulsating, soft-tissue mass associated with a pathological fracture.

Post-surgical monitoring of the size of a popliteal artery aneurysm is recommended, as enlargement of the aneurysmal sac due to residual blood supply can occur, eventually leading to the rupture, even when there is no detectable blood flow in the Doppler exam. During follow-up, a growing mass should be further investigated with an angio-CT scan and never underestimated. Arterial anatomy and different types of described vascular anatomical variants should be considered in surgical planning of popliteal artery. Notably, knowledge of these anatomical variants of popliteal artery branches can influence the success of a surgical procedure. This is particularly relevant in the case of the pattern described as 2b. Furthermore, the risk of iatrogenic injuries of the popliteal artery or any of its terminal branches during surgical interventions increases if anatomical variants are present.

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Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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