



Case report

A rare case of type III popliteal artery entrapment syndrome causing popliteal pseudoaneurysm

Jinshou Yang^a, Peng Liu^c, Yingxin Xu^b, Yan You^d, Xiao Di^{b,*}, Yuexin Chen^{b,**}^a Department of General Surgery, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences, Beijing, China^b Department of Vascular Surgery, Peking Union Medical College Hospital, Beijing, China^c Department of Vascular Surgery, The First Hospital of Hebei Medical University, Hebei Province, China^d Department of Pathology, Peking Union Medical College Hospital, Beijing, China

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ABSTRACT

Here, we report the case of a 30-year-old male who presented with acute right calf muscle pain. Computed tomography angiography revealed a popliteal artery aneurysm in the midsection. Popliteal fossa muscle tissue evaluation revealed that the popliteal artery aneurysm was located beneath an anomalous muscle bundle. Thus, the patient was diagnosed with type III popliteal artery entrapment syndrome and treated surgically. The patient was asymptomatic at rest and during physical activity 4 months after surgery with unobstructed bloodstream in the right popliteal artery. Popliteal artery entrapment syndrome should be considered in young male patients with popliteal artery pseudoaneurysms without atherosclerosis, hereditary diseases, or infections and treated surgically.

1. Introduction

The popliteal artery (PA) is located in the popliteal fossa, adjacent to the femoral condyle posterior aspect and the knee joint capsule posterior. Popliteal artery aneurysm (PAA) is one of the most common PA pathologies (70–80 %) [1]. Approximately 90 % of PAAs are attributed to atherosclerosis, particularly in middle-aged to older patients with a history of long-term smoking, hyperlipidemia, and hypertension, with an incidence rate of 1 % in males aged 65–80 years. [2,3] The PA provides blood supply to the posterior leg muscles. Untreated PAA can lead to reduced blood flow and ischemia in the leg muscles leading to amputation in severe cases [4].

In rare cases, PAA can be induced by popliteal artery entrapment syndrome (PAES), characterized by an abnormal course of the medial head or part of the gastrocnemius muscle compressing the popliteal artery laterally. It was reported that 6 % of PAAs patients were associated with PAES. [5,6] PAES was first reported formally in 1965 with its high morbidity in young males, often associated with thrombus formation [7]. In PAES limbs, 9.1 % (34/374) presented aneurysms or ectasia [6]. PAES predominantly affects young individuals without significant cardiovascular risk factors, and PA compression often results in lower limb ischemia [8]. Mechanically, PAES frequently causes damage to the PA by repeated compression between the bone and contracting muscles, resulting in vascular degeneration ultimately leading to aneurysmal dilatation, plaque formation, thrombosis, and occlusion [9]. Currently, the revised

* Corresponding author. Department of Vascular Surgery, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences, Shuaifuyuan 1st, Dongcheng District, Beijing, 100730, China.

** Corresponding author. Department of Vascular Surgery, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences, Shuaifuyuan 1st, Dongcheng District, Beijing, 100730, China. cyuexin2007@163.com

E-mail address: dixiao56@sina.com (X. Di).

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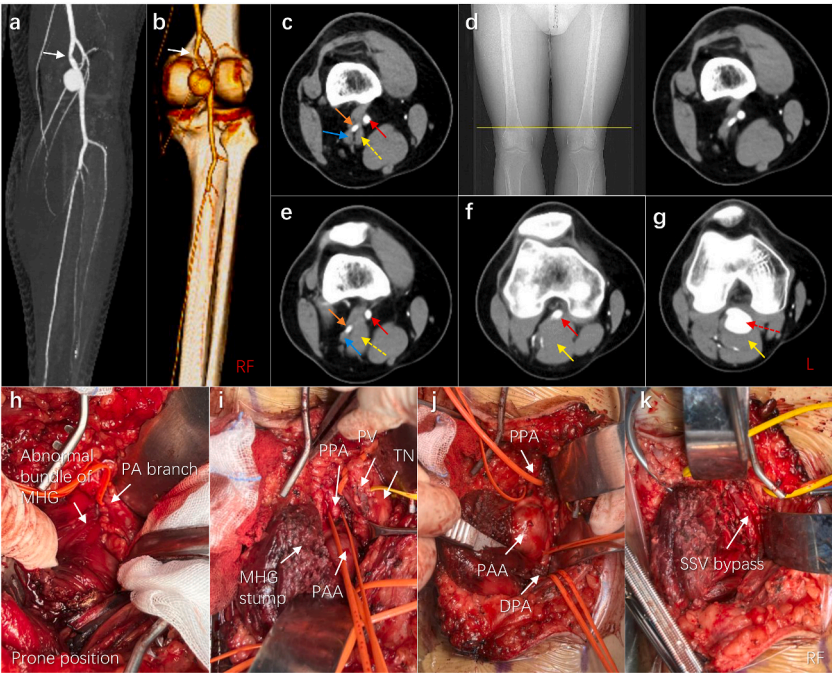


Fig. 1. Images of the PAES patient during diagnosis and treatment procedure. (a, b) Aneurysm of the right popliteal artery (PA) on computed tomography angiography (CTA). The stenosis of PA was shown by white arrows. (c–g) The axial images showed the abnormal muscle bundle and popliteal artery aneurysm (PAA). The yellow line in d show the position of this muscle bundle originating above the medial condyle of the femur. Red arrows show PA. Yellow arrows show the medial head of the gastrocnemius muscle (MHGM), the dotted ones of which shows the abnormal muscle bundle. The orange arrow shows the branch of PA. The blue arrows showed popliteal vein (PV). (h–k) Intraoperative photographs showing the abnormal bundle of MHG, proximal PA (PPA), distal PA (DPA), PV, tibial nerve (TN), and small saphenous vein (SSV).

Table 1
The molecular genetic test report for aneurysm-associated diseases (blood sample).

(Pathogenic/suspected pathogenic) variation that explains target phenotypes	
Nuclear genome SNV/small fragment Indel	Negative
Nuclear genome large fragment CNV/AOH	Negative
Mitochondrial DNA SNV and small fragment Indel	Negative
Variation consistent with target phenotypes but not clearly associated with the onset or progression	
Nuclear genome SNV/small fragment Indel	Negative
Nuclear genome large fragment CNV/AOH	Negative
Mitochondrial DNA SNV and small fragment Indel	Negative
Other variation that partially explains target phenotypes	
Negative	
(ACMG) Variation that is not associated with target phenotypes but involved in severe genetic diseases	
Negative	

^aSNV: single nucleotide variation; CNV: copy number variation; Indel: insert deletion variation; AOH: absence of heterozygosity; ACMG: American College of Medical Genetics and Genomics.

classification of PAES by Rich et al. has been used extensively [10]. Herein, we report a rare case of a patient with PA pseudoaneurysm caused by PAES. Written informed consent for publication was obtained from the patient.

2. Case presentation

A 30-year-old male presented to the vascular surgery department of a local hospital with sudden-onset right calf posterior muscle pain that had occurred after exercise six months prior to presentation. The patient did not have fever, intermittent right lower limb claudication, or the “6P” symptoms [11]. He had a history of hyperlipidemia for many years without regular treatment with grade 2 hypertension. He had no history of smoking, atherosclerosis, hereditary connective tissue diseases, vasculitis, or infectious diseases (syphilis, typhoid fever, tuberculosis etc). The patient was obese (BMI 29.38 kg/m²) with no signs of Marfan or Loeys-Dietz syndromes. The right lower limb skin temperature was normal with no muscle atrophy. A pulsatile mass was palpated in the right popliteal fossa with no palpable arterial pulses detected in the right foot dorsal or posterior tibial arteries. Lower limb arterial Doppler ultrasound

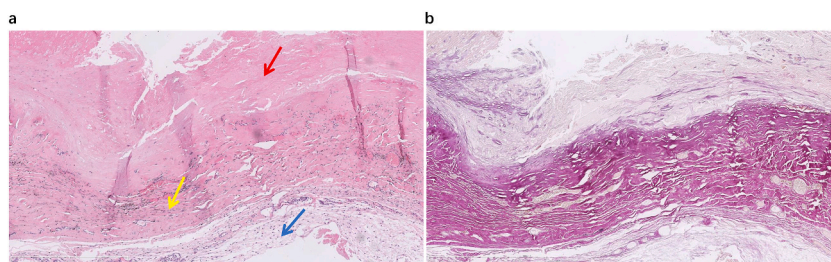


Fig. 2. Representative images of the pathological examination result. (a) Hematoxylin and eosin stain of the vascular wall of popliteal artery pseudoaneurysm; the red arrow refers to mural thrombus, the yellow arrow refers to fibrous connective tissue with hyaline change and hemosiderosis, the blue arrow refers to areolar tissue. (b) Elastic fibers staining by Victoria blue method showing negative result.

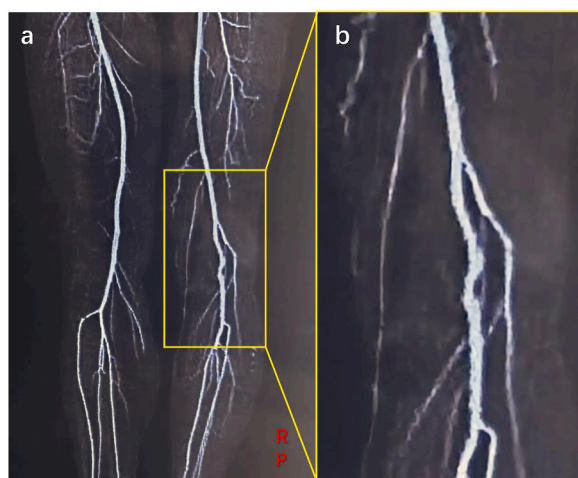


Fig. 3. Images of CTA 4 months after surgery. (a–b) The angiography results showing unobstructed bloodstream in the right popliteal artery.

revealed a right PAA, and the patient was referred to Peking Union Medical College Hospital.

We admitted the patient and performed enhanced computed tomography angiography (CTA) of both lower limbs, revealing a PAA ($3.2 \times 2.9 \times 2.5$ cm) in the mid-segment (P2) of right popliteal artery. Moreover, small mural thrombi were observed at the right PA proximal segment and the right tibiofibular trunk distal bifurcation with segmental occlusion of the proximal right peroneal artery (Fig. 1a and b). Furthermore, aortic and branch artery CTA revealed no other arterial aneurysms or atherosclerosis. The patient had a family history of aneurysms (grandmother and uncle); therefore, genetic testing was conducted, revealing no genetic mutations (Table 1), thus excluding hereditary diseases. After admission, we assessed the erythrocyte sedimentation rate and hypercoagulability marker (including protein C, protein S, antithrombin III, activated protein C resistance, lupus anticoagulant, antiphospholipid antibody panel, and tumor markers) levels, and conducted Widal and Weil-Felix tests for ruling out pseudoaneurysm resulting from rare *Salmonella typhi* infection; all these tests yielded normal results. In addition, we ruled out deep vein thrombosis though routine duplex ultrasound examination.

PAES was considered as an etiology, and enhanced CT images were reevaluated, revealing partial muscle bundles from the medial head of the gastrocnemius muscle (MHGM) passing through the PA lateral side between the PA and popliteal vein (PV) (Fig. 1c–e-g). This abnormal muscle bundle originated above the femur medial condyle, which was higher than the normal MHGM origin (Fig. 1d). Thus, this was a typical case of type III PAES [10]. We speculated that the abnormal MHGM induced prolonged PA compression, inducing PA wall microtrauma, subsequently leading to pseudoaneurysm formation.

We decided to treat the patient surgically. After the patient is generally anesthetized and placed in the prone position, we made an S-shaped incision in the right popliteal fossa and performed layered dissection to expose the tibial nerve (TN), PV, and PA. Partial MHGM muscle bundles ran below a PA branch and terminated at the femur medial condyle (Fig. 1h). We completely severed the MHGM and exposed the PAA (diameter, approximately 3.5 cm) to completely expose the proximal segment of the PA and the aneurysm (Fig. 1i and j). Upon exposing the aneurysm, we observed a significant number of old mural thrombi. Aneurysm tissues were collected for pathological examination. Due to poor retrograde blood flow in the PA distal segment, we performed thrombectomy twice using a 3 French thrombectomy catheter. Finally, bypass with reversed SSV (small saphenous vein) was successfully performed with end-to-end anastomosis of the healthy artery (Fig. 1k).

After surgery, the patient received an intravenous infusion of sodium heparin (activated partial thromboplastin time, 35–40 s) and 100 mg aspirin once daily, without any discomfort (swelling, numbness, or pain in the right lower limb). The pathological report

Table 2

Case reports about PAES resulting in PAA.

Author	PMID	Journal	Age (Years)/ Gender	Classification	Examination	Treatment	Recovery
Hyeon Ju Kim	37885149	<i>Vascular Specialist international</i>	36/Male	Type 5	Ultrasound, CT, MRI	Surgery, GSV graft	Yes
Xitao Song	31448662	<i>The Journal of International Medical Research</i>	60/Male	Mixed type (Type 3 and 4)	CTA, MRI	Surgery, SSV graft	Yes
Jesse Chait	31382002	<i>Annals of Vascular Surgery</i>	47/Male	Type 3	Ultrasound, CTA	Surgery, SSV graft	Yes
Bulent Karaman	23154022	<i>Clinical Imaging</i>	45/Male	Type 3	Ultrasound, CTA	NA	NA
Hiroto Iwasaki	20338137	<i>Vascular</i>	73/Male	Type 3	CTA	Surgery, SSV graft	Yes
Hiroichiro Yamaguchi	23555401	<i>Annals of Vascular Disease</i>	48/Female	Type 3	CTA, MRI	Surgery, GSV graft	Yes
Diego Lopez Garcia	17980291	<i>Journal of Vascular Surgery</i>	31/Male	Left: Type 3 or 4 Right: Type 4	Ultrasound, CTA	Left: Surgery, GSV graft; Right: Surgery, SSV graft	Yes
Salwa Haidar	15798926	<i>Pediatric Radiology</i>	11.5/Female	Right: Type 1	Ultrasound, MRI	Intravenous heparin	NA
Joshua	15192578	<i>Journal of Vascular Surgery</i>	7/Male	Right: Type 1 Left: Type 1 (no aneurysm)	Emergent angiography, MRI	Right: Surgery, end-to-end anastomosis Left: Surgery, entrapment release	Yes
J Pfister	1428917	<i>Helvetica Chirurgica acta</i>	23–43/Female	NA	Angiography	Surgery, popliteal-crural vein graft	Yes
M Delemont	1758643	<i>Minerva Chirurgica</i>	NA	Type 3	NA	Surgery, SV graft	NA

indicated fibrous tissue with hyalinization and mural thrombus formation without a clear elastic membrane (Fig. 2a and b). Therefore, we concluded that PAES induced vascular wall tissue injury with elastic membrane rupture, gradually leading to PA pseudoaneurysm formation under arterial blood pressure. The patient was discharged on the seventh day postoperatively. After discharge, the patient was administered 10 mg rivaroxaban, 100mg aspirin and proton pump inhibitor orally once daily. During the 4-month follow-up, the patient adhered to the medication regimen and reported no recurrence of symptoms, including pain or numbness, in the right calf after prolonged walking. The follow-up CTA of the local hospital showed that the right PA bloodstream was unobstructed (Fig. 3a and b).

3. Discussion

PAES is a very rare disease among the general population. A European study ever reported 0.165 % (33/20000) of the incidence of PAES at the Athens Military Hospital [12]. In Japan, the incidence of PAES was 0.12 % of all the cases with peripheral arterial disease received revascularization therapy [13]. Comprehensive approaches can be used for the diagnosis of PAES including the exercise ankle-brachial index (ABI), duplex ultrasound, CTA and magnetic resonance imaging (MRI). In recent years, several studies have reported the occurrence of PAES leading to pseudoaneurysms of the popliteal artery (Table 2). The male patients had a higher morbidity of 70 % (7/10) than the female patients. Almost all patients underwent surgery with GSV/SSV bypass grafting and had good prognosis. However, the distinctive aspect of our case report was the comprehensive screening of the causes of aneurysm formation, particularly including the results of genetic testing and pathological findings. The evidence in pathological level supported the formation of pseudoaneurysms due to PAES, rather than a pure true aneurysm.

In this case, the aberrant MHGM path was easily discernible on enhanced CT. However, we have reported a case of a pseudoaneurysm of the PA with endovascular repair performed with inadequate CTA evaluation results in 2019 [3]. The patient was re-admitted because of recurrence after a long-distance bicycle 2 weeks after discharge. Thus, we recommend careful and comprehensive CTA assessment for treating patients with PAAs based on the 100 % positive rate for PAES diagnosis using enhanced CT [14]. Nevertheless, the potential for misdiagnosis of PAES remains.[15,16] For equivocal PAES cases identified by CTA, MRI can be used for a definitive diagnosis. MRI, with its superior soft tissue resolution, aids identification of abnormal musculotendinous structures, crucial for accurate PAES diagnosis and classification [17].

Surgical treatment is recommended internationally for PAES patients, achieving a high rate of relief [14] [18–20]. Endovascular repair without PAES decompression extremely likely leads to graft failure manifesting as artery occlusion [21]. Furthermore, the distal runoff status may be compromised by thrombosis or embolism after failed endovascular treatment.[3,22] Additionally, we used the SSV as conduit due to the following reasons. The SSV is located near the popliteal artery and its diameter is sufficient; therefore harvesting SSV is convenient and associated with minimal trauma. In this case, the patient was placed in the prone position during surgery, which makes it more challenging to harvest the GSV. Several studies have also reported SSV bypass grafting in treating PA resulting from PAES (Table 2). In general, surgical intervention for popliteal artery release and autogenous vein grafting should be prioritized for patients with PAES and concurrent PAAs.

CRedit authorship contribution statement

Jinshou Yang: Writing – review & editing, Writing – original draft, Data curation, Conceptualization. **Peng Liu:** Data curation. **Yingxin Xu:** Data curation. **Yan You:** Data curation. **Xiao Di:** Writing – review & editing, Funding acquisition, Conceptualization. **Yuexin Chen:** Writing – review & editing, Funding acquisition, Conceptualization.

Patient perspective

The patient expressed a high degree of satisfaction with the therapeutic approach and its outcomes.

Ethics statement

This study obtained the informed consent of the patient, and the paper did not disclose any identity information of the patient. Medical case reports are exempt from ethics committee review at PUMCH. The relevant declaration form is referred to “*Case_Report_Declaration_Form*”.

Data availability statement

Data included in the article or supplementary materials is referenced in the article.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Not applicable.

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