e-ISSN 1941-5923 © Am J Case Rep, 2018; 19: 29-34 DOI: 10.12659/AJCR.905170

 Received:
 2017.05.04

 Accepted:
 2017.08.23

 Published:
 2018.01.09

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

Popliteal Artery Entrapment Syndrome: A Case Report and Review of the Literature

- ABEF Francisco Cialdine Frota Carneiro Júnior B Eduardo Nazareno dos Anjos Carrijo
 - в Samuel Tomaz Araújo
 - A Luis Carlos Uta Nakano
 - A Jorge Eduardo de Amorim
 - A Daniel Guimarães Cacione

Service of Vascular and Endovascular Surgery, Federal University of São Paulo, São Paulo, SP, Brazil

Corresponding Authors: Conflict of interest:	Francisco Cialdine Frota Carneiro Júnior, e-mail: cialdine_jr@hotmail.com, Daniel Guimaraes Cacione, e-mail: dancac@gmail.com None declared
Patient:	Female, 47
Final Diagnosis:	Popliteal artery entrapment syndrome
Symptoms:	Thermal gradient • limb pain
Medication:	—
Clinical Procedure:	Supra-genicular popliteal derivation – infragenicular popliteal with inverted parenial saphenous vein graft
Specialty:	Surgery
Objective:	Rare disease
Background: Case Report:	Popliteal artery entrapment syndrome (PAES) results from an anomalous relationship between the popliteal ar- tery and the myofascial structures of the popliteal fossa. The most common presenting symptoms include inter- mittent pain in the feet and calves on exercise, resulting in lameness. PAES can lead to popliteal artery throm- bosis, stenosis, distal arterial thromboembolism, or arterial aneurysm. The treatment of PAES includes surgical exploration with fasciotomy, myotomy, or sectioning of fibrous band formation, to release the popliteal artery. However, in cases with thrombotic occlusion, thromboendarterectomy with venous patch arterioplasty, or ve- nous graft arterial bypass surgery may be required. This report describes the presentation and surgical man- agement of a case of PAES presenting with limb pain and includes a review of the literature on this condition. A previously healthy 47-year-old woman presented with a 20-day history of sudden pain in the left lower limb,
	associated with pallor and a loss of arterial pulses below the knee. Angiography of the affected limb showed occlusion of the left supragenicular popliteal artery, with arterial occlusion, suggestive of arterial thrombus. Imaging of the right popliteal artery, which was not occluded, showed that it was medially deviated. An ipsilateral saphenous vein graft was used to bypass the left supragenicular popliteal artery to the infragenicular popliteal artery, resulting in resolution of the patient's symptoms.
Conclusions:	PAES is rare and can be under-diagnosed, possibly due to lack of knowledge of this condition. However, if the diagnosis is made early, the prognosis is usually favorable, following appropriate surgical treatment.
MeSH Keywords:	Arterial Occlusive Diseases • Lower Extremity • Popliteal Artery
Full-text PDF:	https://www.amjcaserep.com/abstract/index/idArt/905170



Background

Popliteal artery entrapment syndrome (PAES) results from an anomalous relationship between the popliteal artery and the myofascial structures of the popliteal fossa [1–8]. The most common presenting symptoms include intermittent pain in the feet and calves on exercise [1–8]. PAES can lead to popliteal artery thrombosis, stenosis, distal arterial thromboembolism, or arterial aneurysm [1–8].

Although PAES is a rare condition, it is important to consider in the differential diagnosis in young patients, usually under 30 years of age, who present with intermittent claudication, cramping pain, and weakness in the legs, especially the calves, that are arises on exercise and that disappears on rest [9–11]. In 30% of asymptomatic patients, there is contralateral popliteal arterial impairment, which increases to 60% in symptomatic patients [10,12]. This report describes the presentation and surgical management of a case of PAES presenting with limb pain and includes a review of the literature on this condition.

Case Report

A 47-year-old female Caucasian, who was previously asymptomatic and healthy, presented to the Emergency Room (ER)

with a 20-day history of sudden pain in the left lower limb and pallor of the foot. The patient had symptoms of left intermittent claudication that limited her ability to walk to 200 meters. Cardiac abnormalities were not detected. Only the left femoral pulse was palpable, with an ankle–brachial index (ABI) of 0.6 (normal value: 0.9–1.2). Pulses in the right (contralateral) lower limb and the right ABI were normal.

Doppler ultrasound (US) showed a left popliteal artery with no vascular blood flow, and echogenic content, consistent with thrombus (Figure 1). Bilateral lower limb arterial angiography showed occlusion of the left supragenicular popliteal artery with imaging findings suggestive of thrombus in the proximal one-third of the artery, occlusion of the fibular and posterior tibial arteries, and with the vessels of the plantar arch being supplied by the anterior tibial artery. The contralateral (right) popliteal artery appeared to be medially deviated (Figure 2). These presenting symptoms, signs, and imaging findings supported a diagnosis of left PAES, and surgical treatment was undertaken.

Surgical access to the posterior popliteal fossa was achieved using an S-shaped or bayonet-shaped incision, and showed thickening of the popliteal artery intima with content suggestive of recent thrombus (Figure 3). Initially, the patient was treated with popliteal artery balloon-catheter thrombectomy,



Figure 1. Doppler arterial ultrasound (US). Doppler ultrasound (US) of the left popliteal artery (red arrow) with echogenic content and no vascular flow, suggesting thrombus (blue arrow). (A) A transverse view. (B) A longitudinal view.



Figure 2. Popliteal artery angiogram. The angiogram shows some deviation of the right popliteal artery. The left popliteal artery shows occlusion suggesting a proximal thrombus (arrow).

which resulted in good distal arterial perfusion. However, due to the arterial intimal abnormality, the patient underwent popliteal-popliteal artery bypass grafting. An ipsilateral inverted saphenous vein graft was used to bypass the left supragenicular popliteal artery to the infragenicular popliteal artery (Figure 3). Following surgery, the patient's symptoms resolved, which supported the diagnosis of PAES.

At six months following vascular surgery, the bypass graft and distal arteries were patent, her left ABI was 0.92 (normal), and she had no further symptoms of intermittent claudication. The patient is currently under ambulatory follow-up.

Discussion

Popliteal artery entrapment syndrome (PAES) occurs because of the anomalous relationship between the popliteal artery and the myofascial structures of the popliteal fossa or rarely, by fibrotic bands that compress the popliteal artery [1]. PAES can be congenital or acquired [1]. Congenital, or anatomical, PAES is caused by abnormal embryological development of the popliteal artery or of the myofascial structures in the popliteal fossa with muscular hypertrophy causing further arterial compression [2–4]. Acquired PAES has been reported in high-performance athletes, such as cyclists [5,6]. The majority of cases of PAES have been reported in men [5,10,11].

The most common symptoms associated with PAES are intermittent claudication and pain in the feet and calves after exercise. In more severe cases of PAES, the symptoms are caused by complications that include thrombosis, arterial aneurysm, arterial stenosis, or distal embolism [11–15]. On physical examination, there may be decreased or absent pulses during forced dorsal foot flexion, or signs of decreased perfusion,



Figure 3.The left popliteal-popliteal bypass surgical procedure. (A) The final popliteal-popliteal bypass. (B) Thickening of the intima of the popliteal artery with content suggestive of recent thrombus

Туре І	The popliteal artery has an internal deviation and is medial to the internal tendon of the gastrocnemius muscle, inserted in the internal condyle of the femur
Type II	The popliteal artery is normal and anterior to the internal tendon of the gastrocnemius muscle that is inserted more lateral than usual, compressing the artery
Type III	The gastrocnemius muscle has an additional tendon or fibrous band that inserts laterally, compressing the artery
Type IV	In embryologic development, the popliteal artery is initially deep to the popliteal muscle, becoming superficial to it posteriorly. In this type, the popliteal artery remains deep to the muscle causing its compression although normal anatomy of the gastrocnemius muscle
Type V	Types I to IV associated with simultaneously popliteal vein compression
Type VI	Muscular hypertrophy with normal constitution, resulting in functional compression of the popliteal artery and vein

Table 1. Types of popliteal artery entrapment syndrome (adapted from Bettega et al. [1]).

such as pallor, a lower limb thermal gradient, and limb cyanosis in acute cases [12].

Table 1 shows the six types of PAES [1,10,16]. In Type I PAES, although the medial head of the gastrocnemius is normal, the popliteal artery is displaced medial to and beneath the muscle or its tendon. In Type II PAES, the popliteal artery is compressed because the medial gastrocnemius muscle arises from an abnormal lateral position. In Type III PAES, the gastrocnemius muscle has an additional tendon or fibrous band that inserts

laterally, compressing the artery. In Type IV PAES, the popliteal artery is compressed by a fibrous band or by the popliteus muscle. In Type V PAES, compression of both the popliteal artery and the popliteal vein occur. In Type VI PAES, or functional PAES, there is normal anatomy, but popliteal artery muscle hypertrophy may be involved.

In PAES, Doppler ultrasound (US) may show hemodynamic changes resulting from compression, such as arterial stenosis, changes in vascular flow, and increase in systolic peak velocity, especially with dynamic maneuvers involving contraction of gastrocnemius muscle [17]. Magnetic resonance imaging (MRI)has been shown to be the best method for evaluation of the anatomy of the popliteal fossa as this imaging method may accurately show the structures surrounding the popliteal artery in this region [18]. Angiography detects compression and occlusion of the popliteal artery and may show an abnormal route of the artery, with other characteristic imaging findings for PAES [16,19]. If no changes are seen on vascular imaging in patients with a high degree of diagnostic suspicion for PAES, the patient may be asked to perform plantar dorsiflexion until becoming symptomatic, and with repeated angiography [18]. The use of MRI with arteriography has been reported to provide the most accurate diagnostic approach for PAES [19].

For patients with PAES and muscle insertion abnormalities, surgery is indicated even when the patient is asymptomatic [20,21]. Ideally, if the disease is diagnosed early and the artery remains uninjured, simple surgical exploration with fasciotomy, myotomy, or fibrous band sectioning and arterial release may be sufficient [22]. Muscle reconstruction is usually not necessary [23]. In cases with non-occlusive vascular injury, thromboendarterectomy with venous patch arterioplasty is a therapeutic option, although the results for this treatment approach have been reported to be inferior when compared with arterial bypass using a venous graft [9].

There is still no consensus on the best surgical access to the popliteal artery in PAES [24]. In cases with small popliteal artery occlusions, a posterior fossa access approach using an S-shaped or bayonet-shaped incision with the patient in the ventral decubitus position is recommended as it allows better visualization of the structures of the popliteal fossa; in cases with larger arterial occlusion requiring femoropopliteal bypass, medial access is most commonly used, although complete arterial exposure is more difficult [16,22,23].

Although endovascular fibrinolytic treatment for patients with acute PAES (symptoms < two weeks) and angiographic evidence of acute arterial occlusion, for the patient described in this report, more than 20 days had passed since the onset of symptoms. It is also important to note that angioplasty alone

References:

- 1. Bettega M: [Popliteal artery entrapment syndrome: Case report]. J Vasc Bras, 2011; 10: 325–29 [in Portuguese]
- Turnipseed WD: Functional popliteal artery entrapment syndrome: A poorly understood and often missed diagnosis that is frequently mistreated. J Vasc Surg, 2009; 49: 1189–95
- Pillai J, Levien LJ, Haagensen M et al: Assessment of the medial head of the gastrocnemius muscle in functional compression of the popliteal artery. J Vasc Surg, 2008; 48: 1189–96

is not effective in PAES since it does not correct the external causes of arterial compression [23,25].

In cases of functional PAES (Type VI), the pathogenesis and progression remain uncertain, with more gradual and delayed vascular symptoms and lesions [20]. Surgical outcome for patients with Type VI or functional PAES are worse when compared with congenital PAES [11], so surgery should be reserved for confirmed symptomatic cases with structural cause [20,26]. A further reported therapeutic option for PAES is US-guided intramuscular injection of botulinum toxin, which results in paralysis of the compressing gastrocnemius muscle, but there have been no controlled clinical studies to support the efficacy of the use of botulinum toxin for patients with PAES [7].

Following treatment, patients with PAES should undergo longterm outpatient follow-up, with treatment re-interventions if necessary [27]. The prognosis of PAES is usually favorable after surgical treatment, especially if the diagnosis is made early. Review of the literature has shown that, although only a few cases of PAES have been reported, following surgery, more than 90% of patients have a significant improvement in their symptoms and return to normal physical activity within three months [28].

Conclusions

Popliteal artery entrapment syndrome (PAES) is rare and under-diagnosed, mainly because clinicians may be unaware of the condition. However, delay in diagnosis and management may lead to irreversible effects of lower limb ischemia. In the case of PAES described in this report, a previously asymptomatic 47-year-old woman presented sudden onset of pain in the lower left limb, compatible with acute arterial obstruction. The diagnosis of PAES was suspected before surgery and confirmed with imaging studies and intraoperative findings. Due to the degree of arterial occlusion, duration of symptoms, and effects of lower limb ischemia, a popliteal-popliteal bypass was performed using a vein graft and via a posterior surgical approach to the popliteal fossa. Following surgery, there was significant improvement in the patient's condition, but she continues to be followed-up in the ambulatory clinic.

 Alencar TA, Melo DM, Karinna FS, Aguiar BC: [Vascular dysfunctions in lower limbs of cyclists]. J Vasc Bras, 2013; 12(2): 139–50 [in Portuguese]

Rignault DP, Pailler JL, Lunel F: The "functional" popliteal entrapment syndrome. Int Angiol, 1985; 4: 341–43

Hislop M, Kennedy D, Dhupelia S: Functional popliteal artery entrapment syndrome: A review of the anatomy and pathophysiology. J Sports Med Doping Stud, 2014; 4: 140

- Hislop M, Kennedy D, Cramp B, Dhupelia S: Functional popliteal artery entrapment syndrome: Poorly understood and frequently missed: A review of clinical features, appropriate investigations, and treatment options. J Sports Med (Hindawi Publ Corp), 2014; 2014: 105953
- 8. Pillai J: A current interpretation of popliteal vascular entrapment. J Vasc Surg, 2008; 48: 61–65
- 9. Fowl RJ, Kempczinski RF: Popliteal artery entrapment. In: Rutherford RB (ed.), Vascular Surgery. 5th ed. Philadelphia, PA: Saunders, 2000; 1087–93
- Levien LJ, Veller MG: Popliteal artery entrapment syndrome: more common than previously recognized. J Vasc Surg, 1999; 30(4): 587–98
- 11. Sinha S, Houghton J, Holt PJ et al: Popliteal entrapment syndrome. J Vasc Surg, 2012; 55(1): 252–62
- 12. Almeida MJ, Yoshida WB, Melo NR: [Popliteal artery entrapment syndrome]. J Vasc Bras, 2003; 2(3): 211–19
- 13. Schurmann G, Mattfeldt T, Hofmann W et al: The popliteal artery entrapment syndrome: Presentation, morphology and surgical treatment of 13 cases. Eur J Vasc Surg, 1990; 4(3): 223–31
- Bonamigo TP, Becker M, Zylberstein S: [Popliteal artery entrapment syndrome: diagnosis and treatment. Case report]. Rev Bras Ortop, 1996; 31(11): 952–56 [in Portuguese]
- 15. Ximenes JOC: [Popliteal artery entanglement syndrome: Case report and literature review]. Rev Col Bras Cir, 1975; 2: 74–77 [in Portuguese]
- Hoelting T, Schuermann G, Allenberg JR: Entrapment of the popliteal artery and its surgical management in a 20-year period. Br J Surg, 1997; 84: 338–41
- 17. Erdoes LS, Devine JJ, Bernhard VM et al: Popliteal vascular compression in a normal population. J Vasc Surg, 1994; 20: 978–86

- Chun JY, Yella R, Gonzalez-Junyent C et al: Popliteal artery entrapment syndrome: Anatomy, imaging techniques and findings. Abstract. Meeting presentation. 2012 European Society of Radiology. Poster No.: C-1736
- 19. Barabas AP, Macfarlane R: Popliteal artery entrapment syndrome. Br J Hosp Med, 1985; 34: 304–6
- 20. Love JW, Whelan TJ: Popliteal artery entrapment syndrome. Am J Surg, 1965; 109: 620–24
- Oliveira FM, Santos ACB, Takito AM et al: Bilateral popliteal artery entrapment syndrome: Case report. J Vasc Bras, 2008; 7(2): 159–62
- 22. Rich NM, Hughes CW: Popliteal artery and vein entrapment. Am J Surg, 1967; 113: 696–98
- Tercan F, Oğuzkurt L, Kızılkılıç O et al: Popliteal artery entrapment syndrome. Diagn Intervent Radiol, 2005; 11: 222–24
- Di Marzo L, Cavallaro A, Mingoli A et al: Popliteal artery entrapment syndrome: The role of early diagnosis and treatment. Surgery, 1997; 122: 26–31
- Baltopoulos P, Filippou DK, Sigala F. Popliteal artery entrapment syndrome: anatomic or functional syndrome? Clin J Sport Med, 2004; 14(1): 8–12
- Williams C, Kennedy D, Bastian-Jordan M et al: A new diagnostic approach to popliteal artery entrapment syndrome. J Med Radiat Sci, 2015; 62: 226–29
- Ikawa MH, Natour J, Jannini MG et al: [Popliteal artery interlocking syndrome: A differential diagnosis of limb claudication: Contribution of magnetic resonance imaging for diagnosis and evaluation]. Rev Bras Reumatol, 2007; 47(6): 442–45 [in Portuguese]
- Araújo JD, Araújo Filho JD, Ciorlin E et al: [Entrapment of popliteal vessels: Diagnosis and treatment and the concept of functional entrapment]. J Vasc Bras, 2002; 1(1): 22–31 [in Portuguese]