Cystic disease of the popliteal artery: a case report

Doença cística da artéria poplítea: relato de caso

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Abstract

Although its rarity, the adventitial cystic disease of the popliteal artery (ACDPA) must be remembered in the differential diagnosis of intermittent claudication in young patient's lower limbs. Brazilian literature presents a few cases of this disease. This study is a case report of ACDPA in a male patient, with a literature review, comparing to popliteal artery entrapment syndrome.

Keywords: Intermittent claudication, popliteal artery, cysts.

Resumo

Apesar de sua raridade, a doença cística da artéria poplítea (DCAP) deve ser lembrada no diagnóstico diferencial de claudicação intermitente de membros inferiores em pacientes jovens. A literatura brasileira apresenta poucos relatos dessa doença. Este trabalho reportou o caso de um paciente masculino portador de DCAP e revisou a literatura, traçando um paralelo com a síndrome de aprisionamento da artéria poplítea.

Palavras-chave: Claudicação intermitente, artéria poplítea, cistos.

Introduction

Cystic disease of the popliteal artery (CDPA) and popliteal artery entrapment syndrome (PAES) are the leading causes of lower limb claudication in young patients. CDPA has been little studied due to its rarity. Its etiology is not clear, and its existence must always be remembered by the vascular surgeon. The Brazilian literature presents some case reports[1-5], references in meetings[6-8], and a personal communication on this theme[2].

Case description

RLS, 39-year-old Caucasian male, soccer teacher, complained of calf pain for 1 year and its worsening for 8 years, when he started presenting paleness and 100-m intermittent claudication of right lower limb, followed by right foot paresthesia. The patient's history indicated right ruptured cruciate ligament 18 years earlier, confirmed by magnetic resonance imaging, which showed anterior ruptured cruciate ligament, injured meniscus, severe arthrosis on the right knee and mild arthrosis on the left knee (Figure 1). The patient did not present risk factors for atherosclerotic disease. Physical examination revealed important muscle hypertrophy on both calves. Distal arterial pulses were present, but disappeared during passive dorsiflexion maneuvers of the feet and extension of the knees.

Doppler ultrasonography imaging suggested right popliteal artery aneurysm with disappearance of arterial flow after prolonged calf contraction (Figures 2 and 3). Arteriography of the right inferior limb showed absence of medial popliteal artery deviation (Figure 4).

Magnetic resonance imaging confirmed the aneurysmal formation in the right popliteal artery, ruling out anomalous muscle insertions (Figure 5).

Retrospectively, subtle signs of lateral entrapment of the popliteal artery in juxta-articular position were identified by angiographic images.

The treatment was open surgical intervention via posterior approach of the right popliteal fossa. A large multilocular adventitial cyst was observed in the popliteal artery,

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Figure 1 – Arthrosis and intra-articular degenerative cysts

Figure 2 – Color-assisted Doppler echography of the right lower limb revealing hypoechoic image compatible with cyst

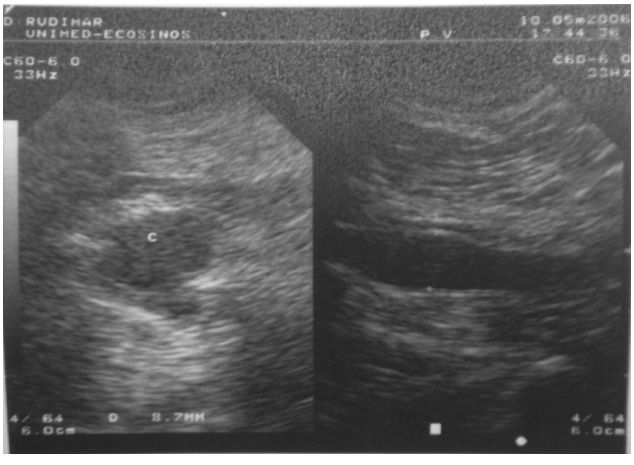

Figure 3 – Echographic evidence of arterial flow disappearance after vigorous contraction of the calves

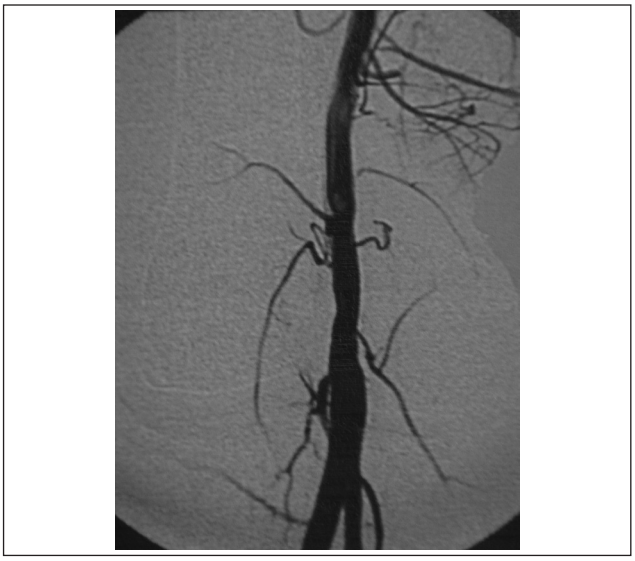

Figure 4 – Arteriography with subtle lateral fulfillment failure

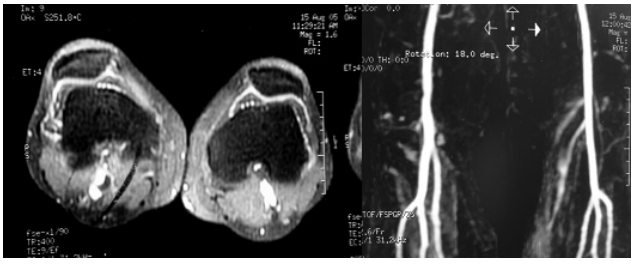

Figure 5 - Magnetic resonance imaging showing cyst and angioresonance with slight lateral fulfillment failure

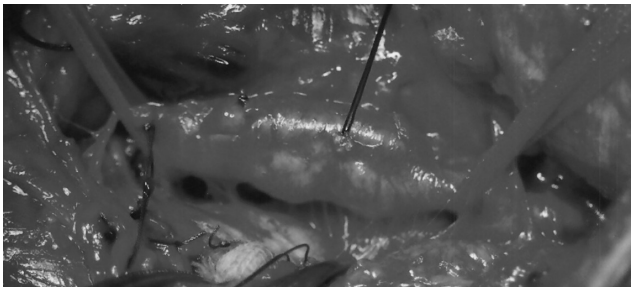

Figure 6 – Absence of communication of the cyst with adjacent structures

between its distal and medium thirds. The rest of the vessel had no anatomical alterations (Figure 6).

The exploration of the popliteal fossa did not identify Baker cyst or muscle compression.

Dissection of the popliteal artery demonstrated an important involvement of the vessel by the cystic formation, without precise limits between them (Figure 7A), thus causing lumen compression due to the firm consistency of the cysts. The slight adherence to the adjacent tissues allowed

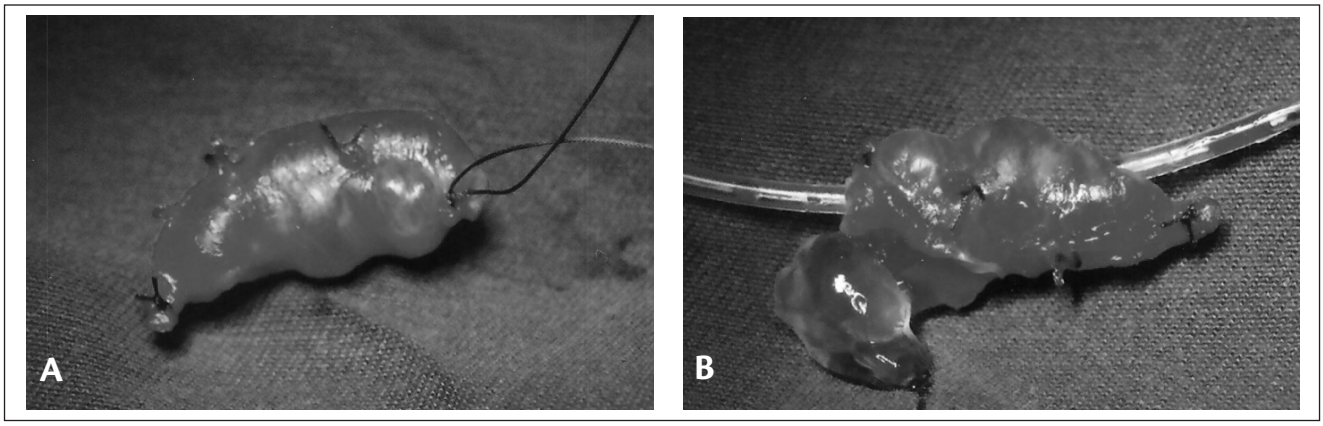

Figure 7 – Segment of the popliteal artery presenting adventitial cysts (A) and evacuated mucoid content (B)

release of the cystic mass without rupture or macroscopic extravasation of the content. Resection of the impaired arterial segment and interposition of the inverted ipsilateral internal saphenous vein segment were the chosen surgical techniques. Macroscopic analysis of the surgical specimen demonstrated a viscous substance in the cystic formation (Figure 7B). Histopathological analysis confirmed the adventitial mucinous degeneration with a material containing protein and histiocytes. The patient has had a 49-month follow-up and has reported absence of symptoms and of physical restriction.

Discussion

Lower limb intermittent claudication in young patients without evident etiological factors for arteriosclerotic disease is rare. The most common etiologies are PAES and CDPA, which is considered to be the cause of 1 in 1,200 cases of intermittent claudication[2]. Extrinsic compression and embolism of the popliteal artery, thromboangiitis obliterans and fibromuscular dysplasia[3,4] are also diagnostic possibilities.

Some particularities of the differential diagnosis between PAES and CDPA are henceforth discussed taking into account the age, sex, clinical features, alterations found in semiologic maneuvers and imaging examinations.

Age

Symptoms of PAES are common in young individuals until the third decade of life, while those of CDPA occur preferentially between the fourth and the sixth decades. It is worth emphasizing that entrapment may go unnoticed for years, hence this hypothesis should not be rejected in individuals of all age groups[1].

Genre

Both diseases are more common in males: PAES presents a frequency of 8:1 and CDPA of 4.7:1[2].

Clinical presentation

There are cases of PAES resulting only from calf muscular hypertrophy. However, the vast majority of cases are caused by congenital anomaly of the anatomical relation between the popliteal vessels and the remaining adjacent structures. In the described case, the presence of muscular hypertrophy made the diagnosis more difficult.

In both CDPA and PAES, there may be any degree of ischemic manifestations, depending on the extension of involvement and on the time of disease development. Both CDPA and PAES may also present asymptomatic periods. As a justification of this finding, some authors suggest that, in some cases, the synovial capsule and the cyst may communicate, thus modifying the intermittent pressure in its interior[9]. Spontaneous remission of the disease in a ten-year follow-up has been reported[10].

In PAES, there is bilaterality in almost 25% of the cases[11], while in CDPA there is only one case reported in literature[12].

Semiologic maneuvers

In PAES, disappearance of distal pulses with knee extension and passive dorsiflexion of the feet are frequently found, as reported by McDonald[13]. The maneuver may be performed with the aid of computed tomography scan or ultrasonography[11]. However, one should be aware of the possibility of false negative results[11] in the presence of hypertrophy of the calves, as confirmed

by echocardiography in this case. Ishikawa[14] described a maneuver of knee flexion with disappearance of the limb distal pulse related to CDPA.

Imaging techniques

Doppler ultrasonography, magnetic resonance imaging or computed tomography may be useful in the morphological identification of the artery and its relation with adjacent structures[15-17]. Due to the rarity of these findings, the image of a cyst may be mistaken for aneurysm or pseudoaneurysm, and the internal viscous substance may be mistaken for hypoechogenic mural thrombi. On the other hand, post-stenosis aneurysmal injuries may be found in 6.7% of the entrapments[18]. In CDPA, the arteriography can reveal compression of the artery affected by the cyst – the scimitar sign[15].

A detailed study can identify small alterations in images. In PAES, there may be medial deviation of the artery, presence of collateral circulation and decrease or absence of blood flow with active flexion of the feet. There are reports of entrapment without arterial deviation and with post-stenosis aneurysmal dilatation[19].

Etiopathogeny of the cystic disease of the popliteal artery

CDPA is an uncommon morphological alteration. The disease has a higher incidence in the popliteal artery. There are reports of cases with involvement of the external, femoral, radial, ulnar and brachial iliac arteries[20], and even veins[21]. Some authors report the presence of mucoid cysts originated in other sites of the vessel wall, besides the adventitia, as in the intima-media layers[20,22]. The etiology is unknown and controversial.

In 1998, South African authors presented a theory called "unifier hypothesis" considering that the cysts were traces of ganglion cells in non-axial vessel junctions[23].

Other authors indicate that repeated trauma is a possible etiologic factor[2]. The present case support this hypothesis, here represented by the degenerative alterations and ligamentous lesion of the right knee resulting from past trauma, as shown by magnetic resonance imaging.

Some authors[9,22,24] report cystic communication with the knee. This fact supports the hypothesis that adventitial cysts are actually synovial cysts that infiltrated the adventitia. Conversely, there is no explanation for the difference in the amount of hyaluronic acid found in the synovial fluid in comparison to the content of the cysts[25]. The presence of macroscopic communications of the cyst with other adjacent structures was not identified.

Saudi authors suggested, in 2007, the possibility of association of CDPA with the intimomedial mucoid degeneration. Meanwhile, other authors found it related to the etiology of aneurysmal formations[20].

Treatment

Simple aspiration of the cysts may be performed under echographic[26], tomographic[11] or transoperative[9,17] guidance. When considering the possibility of simple cyst evacuation, one must observe the structure of the reminiscent artery and pay attention to the possibility of unsuccessful intervention and recurrence, which occurs in 10% of cases in some reports[10]. Due to the viscosity of the fluid, simple aspiration may not be easy[9]. In this case, adventitia resection is an alternative[27]. Angioplasty must only be considered in exceptional cases[28,29]. When there is arterial occlusion, fibrosis or partial destruction of its wall by the cyst, excision and replacement by a venous graft are recommended, an alternative considered to be reliable and safe, with lower recurrence rate[3,4,29].

Conclusion

This paper reported the case of a patient presenting a clinical picture compatible with CDPA (which etiology remains unknown), who underwent successful conventional surgical treatment.

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Writing of the paper: DTSA and LFLA
Critical analysis: EEG
Final text approval*: DTSA, LFLA and EEG
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