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CASE REPORT

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# Cystic adventitial disease of popliteal artery, the both sides of the coin: Arterial resection vs cyst excision

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**Abstract**

Adventitial cystic disease (ACD) is a rare form of non-atherosclerotic arterial stenosis. This entity accounts only for 0.1% of all vascular diseases and affects the popliteal artery unilaterally in 85% of the cases. The options for treatment ACD include excision of cysts, removal of the affected arterial segment with vein graft reconstruction or radiological aspiration. We present two cases of ACD of the popliteal artery and its subsequent management and discuss the pros and cons of the treatment's strategies.

**KEYWORDS**

cardiovascular surgical procedures, cystic adventitial disease, peripheral vascular diseases, popliteal artery

## 1 | INTRODUCTION

Adventitial cystic disease (ACD) was first described in 1947 by Atkins and Key in the right external iliac artery. This entity accounts only for 0.1% of all vascular diseases and affects the popliteal artery unilaterally in 85% of the cases.[1,2] It's a rare form of non-atherosclerotic arterial stenosis. Adventitial cystic disease of the popliteal artery usually presents as intermittent claudication. Bilateral involvement has been reported with compromise of the external iliac, common femoral, radial, and ulnar arteries. It most commonly affects men in their fourth and fifth decade of life.[1–3]

The options for treatment ACD include excision of cysts, removal of the affected arterial segment with vein graft reconstruction or radiological aspiration.[4] We present two cases of ACD of the popliteal artery and its

subsequent management and discuss the pros and cons of the treatment's strategies. Patients approved the publication of images and details related to their case.

## 2 | CASE 1

A 47-year-old man was referred to the vascular surgeon with right calf pain after walking for 10 min. The physical examination revealed palpable right popliteal mass and foot pulses at rest that persisted during knee flexion. The right and left ankle-brachial pressure indices were 1.41 and 1.3, respectively. Laboratory findings were normal, and he did not have any previous disease. Arteriography showed that the popliteal artery was compressed by a non-enhancing structure related to the arterial wall of the popliteal artery (Figure 1).

Adventitial cystic disease of the popliteal artery was diagnosed, and surgery was scheduled. An S-shaped skin incision was cut linear to the skin fold in the right popliteal fossa under general anesthesia with the patient in the prone position throughout the procedure. After dissection, a round, loculated cystic lesion encircling the popliteal artery was identified. Due to the presence of complete arterial wall compromise and arterial thrombosis, we completely resected the cyst and the affected segment of the popliteal artery and reconstructed the artery using a great saphenous vein graft. Histopathologically, the cyst had a relatively clear margin with vascular adventitia. The cyst had no arterial elements in the wall and mucinous content. The postoperative course was uneventful, and the patient was free of right calf claudication. The condition did not recur, and three-dimensional CT angiography, 6 months later, showed good patency of the vein graft (Figure 1).

## 3 | CASE 2

A 54-year-old woman was referred to the vascular surgeon in the emergency department with acute limb ischemia suspicion and right calf pain after walking for 3 min. The physical examination revealed palpable right popliteal mass and foot pulseless. Right inferior limb arterial Doppler showed triphasic flow in the foot. Laboratory findings were normal, and she only has arterial hypertension as a previous disease.

Computed tomography angiography (CTA) showed that the popliteal artery was compressed by a non-enhancing structure related to the arterial wall with the

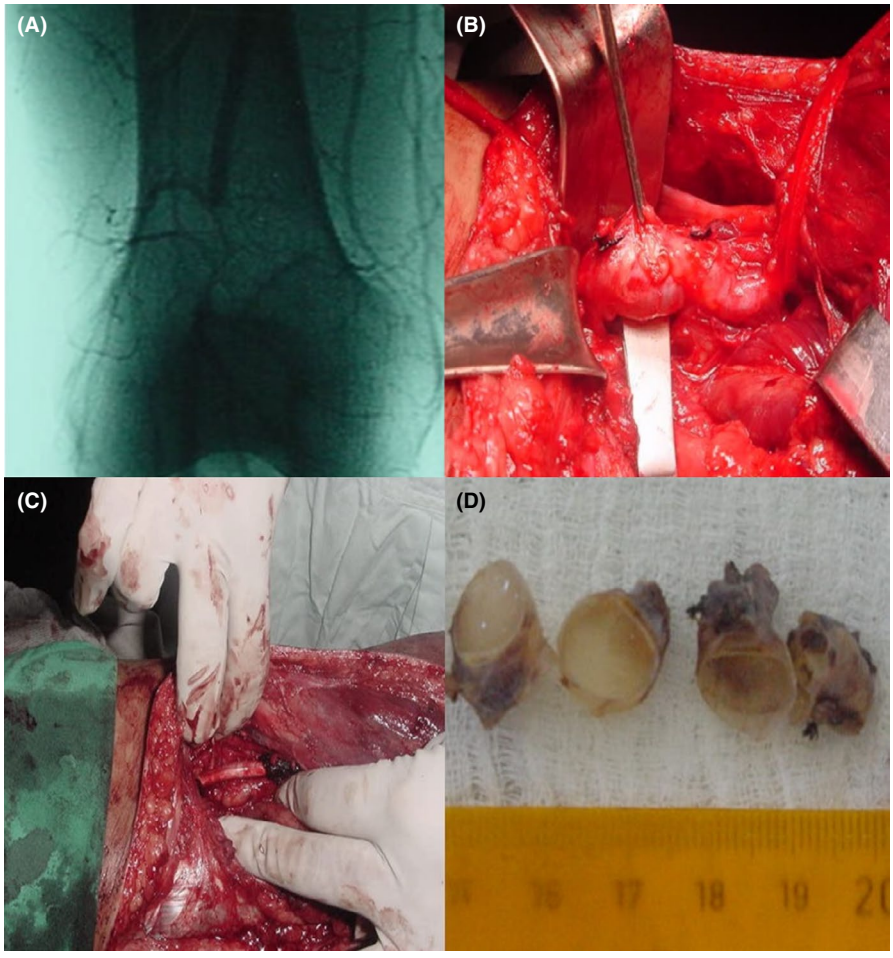


**FIGURE 1** Case 1. (A) Popliteal artery arteriography with popliteal cyst evidence. (B) Popliteal artery cyst with compromise of all the arterial wall. (C) Final result after en-bloc resection of the popliteal artery cyst with end-to-end arterial anastomosis reconstruction. (D) Macroscopic vision of the popliteal artery cyst after it's resection.

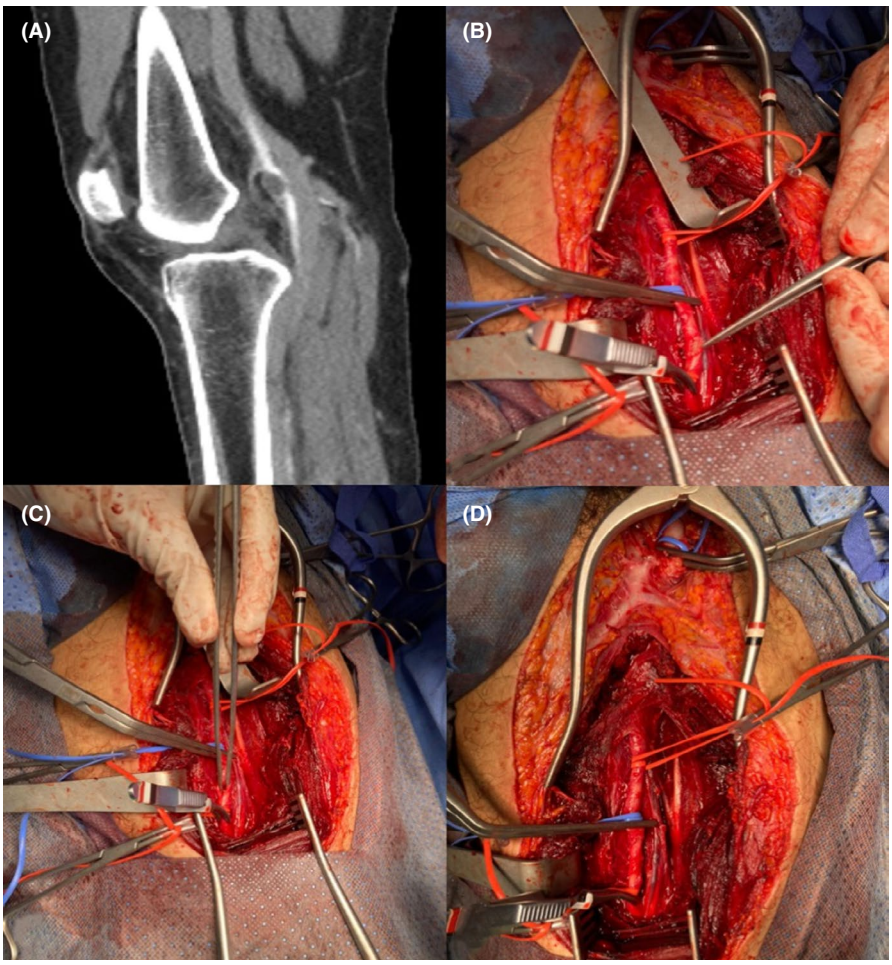


**FIGURE 2** Case 2. (A) Angiotomography that showed the popliteal artery cyst. (B) Popliteal artery with proximal and distal vascular control previous to the popliteal artery cyst excision. (C) Excision of the popliteal artery cyst. (D) Popliteal artery lumen exploration to rule out additional cyst compromise after cyst excision

typical hourglass sign (concentric, extrinsic compression) of the popliteal artery (Figure 2).

Adventitial cystic disease of the popliteal artery was diagnosed, and surgery was scheduled. An S-shaped skin incision was cut linear to the skin fold in the right popliteal fossa under general anesthesia with the patient in the prone position throughout the procedure. After dissection, a round, loculated cystic lesion encircling the popliteal artery was identified. Due to the absence of complete arterial wall compromise and no arterial thrombosis, we perform only a cyst excision. Histopathologically, the cyst had a relatively clear margin with vascular adventitia compatible with ACD. The postoperative course was uneventful, and the patient was free of right calf claudication. The condition did not recur (Figure 2).

## 4 | DISCUSSION

A single theory cannot account for the pathogenesis of all ACD patients. Levien et al. proposed the unification of the ganglion and developmental theories based on embryology of susceptible popliteal blood vessels. They refer that ganglion-like structure is incorporated into de adventitia of the vessel wall during embryological development as a synovial rest that secretes and enlarges over the years.[5–7] The findings reported by Ohta et al. support this hypothesis. They described recurrent adventitial cystic disease in an interposed vein graft. They proposed that mucin synovial cells originating from neighboring joint capsule could invade adventitia or attached to and encircled the adventitia.[5–7]

The prevalence of ACD is 1:1200 of every calf claudication case. Clinical diagnosis is often late due to affected individuals being young and having low risk for vascular disease. Scimitar or hourglass signs can be seen in the angiogram because ACD compresses the arterial lumen in an eccentric or concentric fashion. Even if the affected popliteal artery appears angiographically normal, ACD can induce symptoms during exercise. The severity of ischemic symptoms varies, and depends on the compression grade of the popliteal artery just like in our two clinical cases.[5,6] The pathological findings of ACD are intramural cysts containing mucinous material located between the media and the adventitia of the affected blood vessel as in our cases. Clinically, it should be suspected in the presence of the Ishikawa sign, which consists of the disappearance of the pedial and posterior tibial pulse when flexing the knee and reappearance of pulses when extending the knee.[2,3]

The Doppler ultrasonography is the first-line diagnostic method due to its non-invasiveness. Computed tomography (CT) and magnetic resonance imaging (MRI) allow to evaluate the cyst morphology and discard communications between the adjacent synovial articular capsules. The anatomical arterial intramural cyst extension can be established performing multi-planar sections. The MRI findings of ACD are homogenous cysts with low signal intensity on T1-weighted images and high signal intensity adjacent to vessels on T2-weighted images. Three-dimensional CT reconstructions are recommended to be performed preoperative for adequate surgical planning as in our two cases.[1,5]

Several treatments have been reported for ACD in case reports and small series. The most widely performed surgical approach is surgical cyst excision as in our second case. Tsolakis et al. showed a success rate of 94%; nevertheless, recent reports published success rates of 85% due to cyst recurrence in the long-term follow-up. Cyst excision is possible when the arterial lumen is intact and only has external compression, cyst resection resolves the compression without damaging the vascular endothelium.[6–8]

Surgical excision of the affected arterial segment with vein graft reconstruction is the second most used technique. This surgical approach presents a success rate of 93.5 to 95% as in our first case. The failure of this technique is due to neointimal hyperplasia causing stenosis and thrombotic occlusion.[3,6] Extra-anatomical bypass grafting could be done but would not resolve the local pressure effects from the continued cyst expansion.[1,5,6] Other open surgical approaches without good evidence include arteriotomy with patch angioplasty, intraoperative cyst aspiration and end-to-end native artery anastomosis.[1,2]

Open surgical approach for ACD is invasive and has potential described morbidity that includes the following: chronic pain, nerve damage, and fascial hernia. Although, the long-term surgical treatment success requires radical cyst excision and for accomplishing this an adequately popliteal fossa exploration is needed.[2,3,9]

In conclusion, ACD disease of the popliteal artery is an uncommon non-atherosclerotic cause of peripheral vascular disease in middle age subjects without specific risk factors. Surgical conservative treatment with cyst excision can be performed if no complete arterial wall alteration and no presence of arterial thrombosis.

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None.

## CONFLICT OF INTEREST

The authors have no conflicts of interest to declare.

## AUTHOR CONTRIBUTIONS

All authors contributed to the design of this manuscript. LAN and LFCV wrote the first draft. IDLM, ARNR, and DGC edited and reviewed the final manuscript. MC and MP scientifically reviewed the article.

## CONSENT

The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient.

## DATA AVAILABILITY STATEMENT

Data available on request from the authors.

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