

CASE REPORT

Cystic adventitial disease of the popliteal artery with unusual spontaneous regression: A case report with literature review

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Abstract

This report highlights the case of cystic adventitial disease of the left popliteal artery in a 45-year-old male patient. Imaging modalities confirmed the diagnosis and high resolution MRI found a cystic connection to the adjacent knee joint. The evolution was unusual with spontaneous regression of the symptoms.

KEYWORDS

adventitia, cysts, magnetic resonance imaging, popliteal artery, spontaneous remission

1 | INTRODUCTION

Cystic adventitial disease (CAD) is an uncommon non-atherosclerotic peripheral vessel condition characterized by the collection of mucinous material in the adventitia.¹ Its predilection site is the popliteal artery but can affect other peripheral vessels.¹ It is most commonly described in male patients in their fourth or fifth decade.¹ Here, we report a case of CAD of the left popliteal artery in a 45-year-old man complaining of intermittent claudication in the lower left limb with an unusual spontaneous regression of symptoms within three months of follow-up.

2 | CASE HISTORY

A 45-year-old man has presented, for one year with stage 2 intermittent claudication according to the Rutherford classification with recent worsening. He had no medical history and specifically no atherosclerosis risk factors apart from smoking. He recalled no previous trauma or injury to the lower limbs. Computed tomography (CT) angiography revealed a pre-occlusive luminal stenosis with artery enlargement (Figure 1). A focal thrombosis was first suggested, but the absence of atherosclerotic lesions prompted us to look for local conditions favoring this thrombosis such as trapped popliteal artery syndrome.

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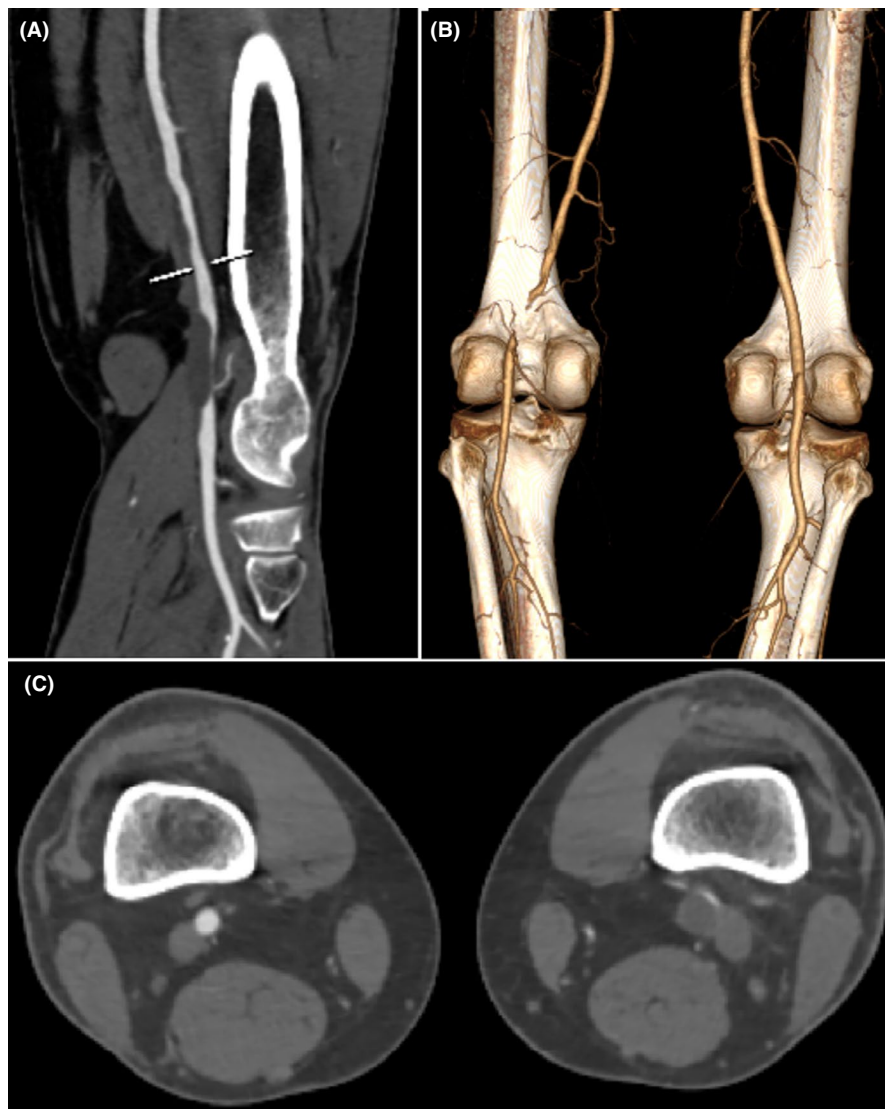


FIGURE 1 CT angiography images: (A) curved image of the left popliteal artery: A cystic lesion in the arterial wall with pre-occlusive luminal stenosis. (B) Posterior volume rendering image: Stenosis of the left popliteal artery. (C) Axial image of the right and left popliteal arteries: Cystic lesion of the left arterial wall (white arrow). Note the pre-occlusive stenosis of the arterial enhanced lumen compared to the contralateral side

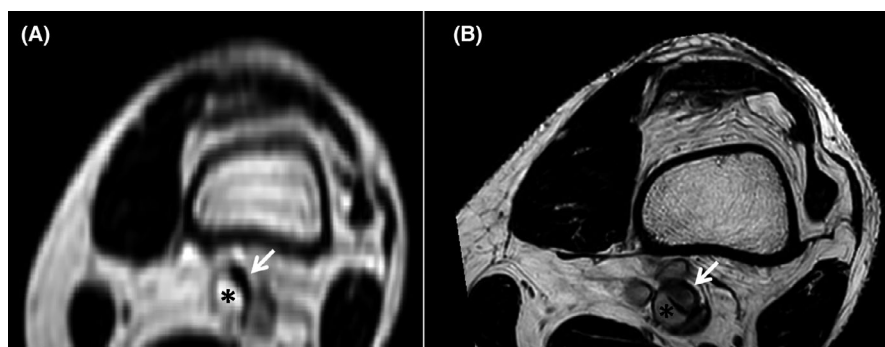


FIGURE 2 Reformatted axial T2 WI from the MRI angiography performed on presentation (A) and a native axial T2 WI on the same level three months later (B): A marked re-expansion of the popliteal artery lumen (white arrows) with persistence of the artery wall cysts (black asterisks)

Thus, a magnetic resonance imaging (MRI) angiography was done.

The lesion had a low signal on T1-weighted images (WI) and a high signal on T2 WI without enhancement after administering a contrast agent, confirming its cystic nature. Arterial stenosis was also noted. Multilocular cysts connecting the artery wall lesion to the knee joint cavity next to the lateral femoral condyle were observed.

The patient was diagnosed with CAD with tight stenosis of the left popliteal artery, and surgical resection with vascular reconstruction was scheduled. Meanwhile, a platelet anti-aggregating agent and vasodilator treatment with functional rehabilitation were both prescribed while awaiting the surgical procedure. The patient made progress, and the walking perimeter improved under conservative treatment. Thus, a new exploration was done.

A second MRI examination (with higher spatial resolution sequences) was performed and revealed a marked re-expansion of the arterial lumen (Figure 2). Clearer images of the cystic connection with the knee joint were also obtained (Figure 3).

A duplex ultrasound with dynamic maneuvers showed no hemodynamically significant stenosis in the left popliteal artery with normal flow velocity waveforms in the downstream leg arteries (Figure 4).

The operation was canceled, and the patient underwent a conservative follow-up to check for symptom recurrence or total regression of the adventitial cysts. As of August 2021, after two years of follow-up, the patient has remained asymptomatic with no changes in either the duplex ultrasound or the MRI angiography examination.

3 | DISCUSSION

Atkins and Key reported the first case of CAD in 1947.² Since then, more than 500 case reports and case series have been published.

The popliteal artery is by far the most affected site with 80% of cases.¹

The patients are predominantly male with a sex ratio of 4/1, and the mean age of diagnosis is 46 years old.¹

They present with intermittent vascular claudication in 81% of the cases, with a history of trauma reported in only 4% of those.¹

Some complications reported included intimal tear³ and arterial thrombosis.⁴

Imaging modalities, mainly MRI and CT angiography, confirm the diagnosis and eliminate differential diagnoses including popliteal artery entrapment syndrome, atherosclerotic disease, dissection, and vascular tumor.

A high spatial resolution MRI is also an important component of preoperative evaluation and follow-up.⁵ It shows the cystic lesions in the vessel adventitia and the

percentage of lumen stenosis as well as demonstrates connections between the cysts and the adjacent joint.

The etiology of CAD remains a highly debated topic. Four major theories have been put forth: repeated trauma, systemic disease, developmental process,⁶ and articular (synovial) connection.⁷

The trauma theory has been challenged because of the rarity of cases—only 4%—caused by a history of trauma. Furthermore, CAD, which is rare in itself, should be more common if caused by a traumatic event or repeated micro-traumatic episodes.

The lack of association between a systemic disorder and CAD renders the systemic disease theory unlikely. In fact, only five cases of bilateral CAD have been reported and no patients had multiple adventitial cysts at different anatomic sites simultaneously.

The developmental theory states that mesenchymal mucin-secreting cell rests become incorporated within the adventitia of vessels during development.⁶

The articular theory seems the most plausible. The fact that all cysts in the reported cases were para-articular strongly suggests an association between CAD formation and its respective neighboring joint. As shown in our observation and in previous recent reports, high spatial resolution MRI sequences reveal cystic connections more frequently.

Spinner et al. put forth evidence to support a joint connection in all cases of CAD.⁷ According to their hypothesis, an articular vascular branch would be the conduit for cyst propagation from the joint to the parent vessel through adventitial dissection. In the example of CAD of the popliteal artery, the joint connection would be the middle genicular artery.⁷

Several treatment methods have been described for CAD. Ultrasound-guided percutaneous cyst aspiration and endoluminal angioplasty are less invasive alternatives, but are associated with a higher incidence of recurrence.¹ This seems consistent with the fact that the articular connection would not be resected by these techniques.

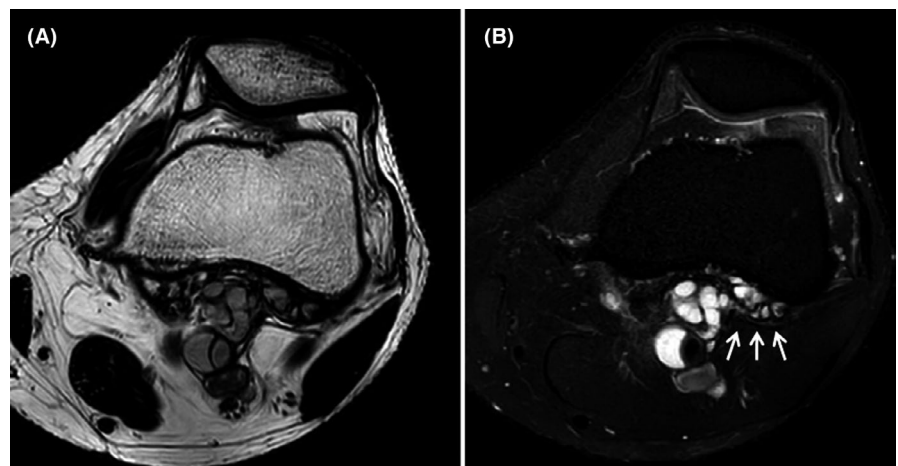


FIGURE 3 (A) Axial T2 WI and (B) axial T2 SPAIR WI: Wall cyst of the popliteal artery with cystic multilocular connection to the intra-articular space, under the joint capsule (white arrows)

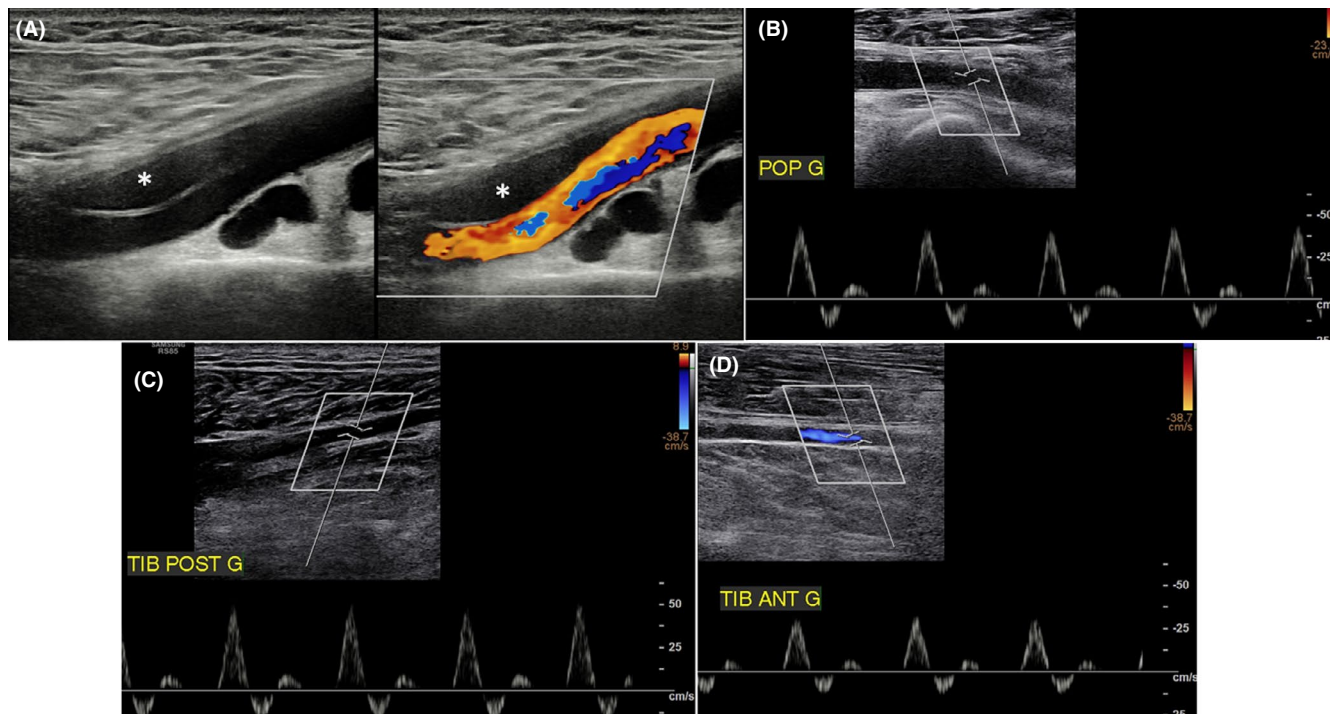


FIGURE 4 Duplex ultrasound images: (A) Anechoic lesion in the popliteal artery wall without color Doppler signal within (white asterisk). (B,C,D) No hemodynamically significant stenosis on the popliteal artery and the downstream leg arteries

The standard treatment remains cyst resection and vessel reconstruction with vein autograft.⁸ Open cyst evacuation with removal of the cyst wall is another surgical option.

Based on a systematic review,¹ it is believed that the key to a definitive procedure would be ligation of the articular connection.

Spontaneous symptoms or cyst regression are rare, with only 9 previous reported cases in the literature.⁹

This unusual outcome suggests that conservative medical treatment and walking rehabilitation should be done as a first recourse, especially in closely monitored cases with few or no significant symptoms. Such a conservative method may require long-term follow-up because recurrence of symptoms and/or adventitial cyst reaccumulation can occur as long as joint connections persist.¹⁰ This follow-up is based on clinical symptoms, physical examination, and primarily Doppler ultrasound. For the first year, close follow-up is recommended every three months, bi-annual visits the second year, and annually afterward. MR angiography may be prescribed once a year for three to five years. In case of frequent and close recurrences, or acute limb ischemia, surgical treatment should be considered.

4 | CONCLUSION

CAD is a rare condition affecting the popliteal artery in most cases. A high spatial resolution MRI is a key diagnostic

tool and helps detect joint connections. Adopting the joint synovial connection theory will make it possible to better understand the pathogenesis of CAD and come to a consensus on a treatment plan, namely to include systematic connection ligation, thereby improving patient outcomes by reducing recurrence rates. Spontaneous regression of cysts is rare and would justify conservative treatment in paucisymptomatic and non-limb-threatening cases with medical supervision and close ultrasound monitoring before considering surgery.

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CONFLICT OF INTEREST

None.

AUTHOR CONTRIBUTIONS

Mohamed Chaabouni involved in conception and design of study, literature search, and drafting of article. Ines Baccouche, Monia Attia, and Chaker Jaber involved in design of manuscript, literature search, and drafting of article. Meriem Affes, Saoussen Hantous, and Salma Kchaou involved in radiographic image acquisition and interpretation. Henda Nèji involved in design of manuscript, drafting of article, and final approval. All authors read and approved the final version of the manuscript.

ETHICAL APPROVAL

Written informed consent has been obtained from the patient.

CONSENT

The authors have confirmed during submission that patient consent has been signed and collected in accordance with the journal's patient consent policy.

DATA AVAILABILITY STATEMENT

The data of this case are available from the corresponding author upon reasonable request.

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