

## Cystic adventitial disease of the popliteal artery

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### ABSTRACT

Cystic adventitial disease is a rare condition that usually affects the popliteal artery, and is a rare cause of non-atherosclerotic stenosis. It is most commonly found in young or middle-aged men with intermittent claudication. Herein we present a histologically proven case of cystic adventitial disease of the popliteal artery in a 53-year-old man. We describe the imaging findings on gray-scale, Doppler and triplex ultrasonography, computed tomography with 3D reconstruction and digital subtraction angiography.

*Key words:* • popliteal artery • intermittent claudication • ultrasonography • computed tomography

Cystic adventitial disease (CAD) is a rare condition that usually affects the popliteal artery. It is most commonly found in young or middle-aged men, usually non-smokers, with intermittent claudication. The pathogenesis of the disease is unclear; however, several theories have been postulated (1, 2). Herein we present a histologically proven case of CAD of the popliteal artery in a 53-year-old man. We describe the imaging findings on gray-scale, Doppler and triplex ultrasonography (US), computed tomography (CT) with 3D reconstruction, and digital subtraction angiography (DSA).

### Case report

A 53-year-old man with a medical history remarkable only for a 30 pack/year smoking was referred to our department because of a 2-year history of intermittent claudication of the right calf that worsened during the last 4 months. Other than cigarette-smoking, the patient had no risk factors for vascular disease. Clinical examination revealed diminished popliteal and pedal pulses on the affected side. Other clinical and laboratory findings were normal, and the patient was in excellent condition.

Gray-scale and Doppler US demonstrated an ovoid cystic structure of about 20 x 15 mm, compressing the right popliteal artery and causing severe narrowing of the lumen. Spectral analysis of the popliteal artery demonstrated an abnormal arterial spectrum proximal to, within, and distal to the stenosis (Fig. 1). Subtle atheromatous changes with no significant arterial stenosis were also found in the arteries of both legs. Diagnosis of CAD was suspected, and the patient was referred for further imaging evaluation.

CT angiography of the popliteal artery confirmed the cystic nature of the lesion (23 HU) that was in contact with the popliteal artery, causing almost complete occlusion of the vessel lumen. Small arterial collaterals were also demonstrated. Further evaluation with 3D-CT reconstructions using the volume rendering technique was performed for better preoperative planning (Fig. 2).

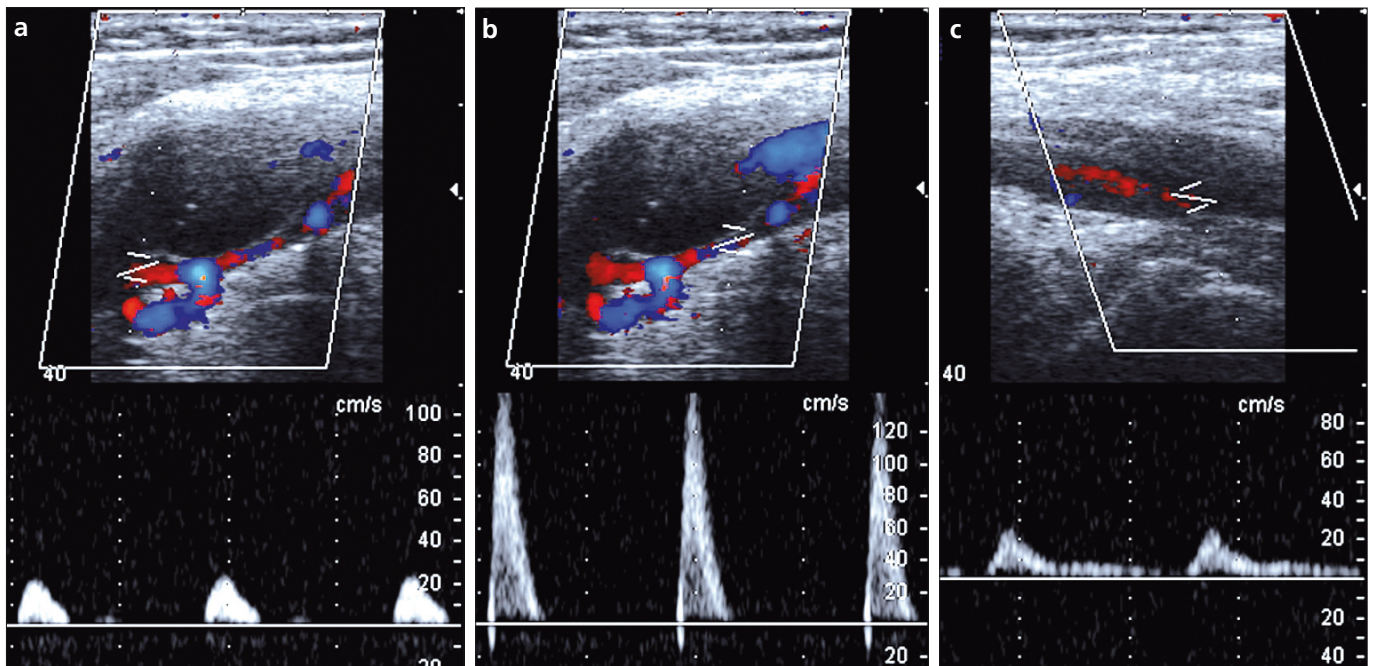
DSA was then performed in order to demonstrate more clearly the degree of the stenosis, the collaterals, and other potential stenoses from the arteries of the lower limbs not demonstrated in previous studies. DSA demonstrated almost complete occlusion of the right popliteal artery for a length of 15 mm, while there were no other stenoses in the arteries of the lower limbs (Fig. 3).

It was finally decided that surgery should be performed because of the patient's persistent symptoms. At surgery, a cystic lesion was found, filled with a viscous, mucous fluid in the wall of the popliteal artery. The wall of the cyst was carefully dissected, and the popliteal artery was decompressed (Fig. 4a). Histopathology of the surgical specimen revealed a section of the wall of a large artery, with findings consistent with CAD (Fig. 4b).

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**Figure 1.** a–c. Triplex ultrasonography of the right popliteal artery proximal to (a), within (b), and distal to (c) the stenosis, which is caused by compression of an anechoic-cystic structure. Proximal to the stenosis (a), monophasic low-velocity (23 cm/s) high-resistance spectrum is demonstrated. Within the stenosis, monophasic high-velocity (138 cm/s) high-resistance arterial waveform is indicative of high grade stenosis. Post-stenotic spectrum (c) remains monophasic low-velocity (22 cm/s) due to diminished blood inflow through the stenosis, with a diastolic component.



**Figure 2.** CT angiography with 3D reconstruction of the right popliteal artery, demonstrating almost complete occlusion of the right popliteal artery due to compression by the cyst (arrow).



**Figure 3.** DSA of the right popliteal artery in anteroposterior view. Almost complete obstruction of the right popliteal artery is observed, with minimal flow through the stenosis, creating the typical curvilinear sign due to extrinsic compression of the lumen (arrow). The distal popliteal artery and the calf vessels are intact.

There were no postoperative complications, and the patient was discharged on postoperative day 8. At 1-month and 18-month follow-up, the patient was asymptomatic with normal findings on Doppler and triplex US of the popliteal arteries.

#### Discussion

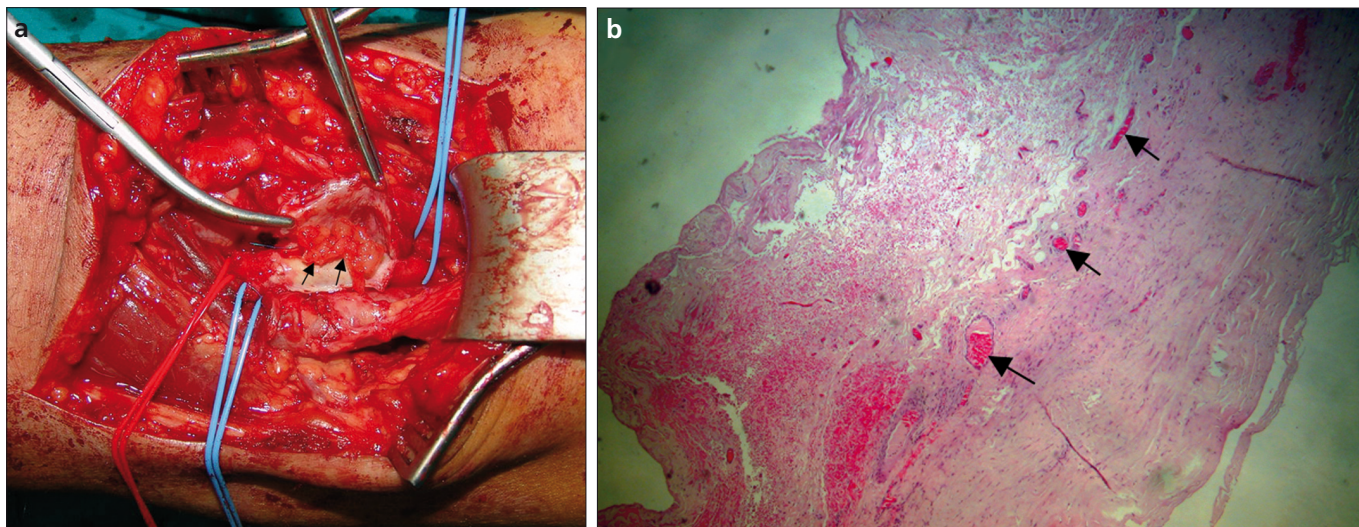
CAD is a rare, non-atherosclerotic vascular disease that was first described in

1947. Fewer than 400 cases have been reported in the literature (1, 2). The disease usually affects the popliteal arteries (about 85% of the cases) and more rarely the common femoral, external iliac, radial, and ulnar arteries (2). A case of bilateral CAD of the popliteal artery has been reported (3). Cases involving veins also have been reported (4).

CAD is characterized by a collection of a gelatinous material rich in muco-

proteins and mucopolysaccharides, clear and colorless or yellowish, located between the adventitia and the middle layer of the vessel wall, compressing the lumen of the artery (3, 5). Pathogenesis of this entity is unclear, but four theories have been proposed (3, 6). The trauma theory suggests chronic degeneration by repetitive trauma. The ganglion theory suggests that synovial cysts track along vascular branches, and finally re-





**Figure 4. a, b.** Photograph during detachment of the adventitia cyst (**a**). Viscous mucous yellowish fluid was present within the cyst (*arrows*). Histological section (**b**, HE, X40) which shows part of the wall of a large artery with hyperemic vasa vasorum (*arrows*).

side in the adventitia of major vessels. In the systemic disorder theory, these cysts are considered to be the result of a generalized connective tissue disorder. Finally, the developmental theory suggests that mucin is secreted by mucin-secreting mesenchymal cells from nearby joints that incorrectly migrated to the adventitia during embryogenesis. In some cases, connections of the cyst with the capsule of the adjacent joint were documented, supporting the latter theory (3, 7, 8).

Most affected patients are young or middle-aged men, usually with no cardiovascular risk factors. Male-to-female ratio is 15:1. CAD causes relatively sudden onset of claudication, and the symptoms may worsen with knee flexion. When there is communication of the cyst with the knee joint, exercise or repetitive trauma may cause rapid growth of the cyst, with accelerated progression of symptoms (1, 3). Spontaneous rupture of the cyst is rare, but may result in relief of symptoms (5).

Imaging evaluation should begin with Doppler US, a non-invasive and rapid method that is the most sensitive diagnostic procedure in demonstrating CAD. Doppler US alone is sufficient for diagnosis (9, 10). Supplementary cross-sectional imaging can be used when there are diagnostic difficulties, providing additional information about the morphology of the cyst and possible communication with an adjacent joint. DSA is an invasive method that may be used in selected cases (2, 5, 7, 11).

There are several treatment options for CAD of the popliteal artery. Although relatively non-invasive, aspiration of the cysts is difficult because the contents are viscous, and the cysts have a high recurrence rate (12). Endovascular treatment with angioplasty or stenting has had disappointing results, and should be avoided because these methods may injure the healthy intima of the artery (13). When the vessel is not totally occluded, as in our case, incision of the cyst with resection of the adventitia and evacuation of the contents is the method of choice, preventing recurrence. When the artery is totally occluded and thrombosed, reconstruction with an autologous vein graft is recommended (2).

The differential diagnosis should include other causes of claudication such as popliteal entrapment syndrome, peripheral arterial occlusive disease, Buerger disease, popliteal artery aneurysm, and Baker cyst (1, 2).

In conclusion, CAD of the popliteal artery, although rare, should be included in the differential diagnosis in patients with intermittent claudication.

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