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SHORT REPORT

Case Report: Cystic Adventitial Disease of the External Iliac Artery with Imaging Features of a Complicating Proximal Dissection

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KEYWORDS
Cystic adventitial disease;
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Abstract *Introduction:* Cystic adventitial disease (CAD) is a rare cause of claudication. We report a case of CAD involving the external iliac artery, with possible cyst rupture intramurally causing significant long segment stenosis of the external iliac artery.
Case report: A 52-year-old female presented with sudden onset (over 1 day) lifestyle-limiting claudication affecting the left calf and thigh. CT angiography of the lower limbs revealed an eccentric low density wall thickening of the left external iliac artery (EIA) producing a 70% stenosis and a cystic lesion just distal to the stenosis.
Discussion: A diagnosis of cystic adventitial disease was made and the patient proceeded to iliofemoral bypass.

Introduction

Cystic adventitial disease (CAD) is a rare cause of claudication, usually of sudden onset and seen in younger patients with no risk factors for atherosclerosis.

The majority of cysts are in the lower limb, with popliteal artery predominance. Aetiology of the cysts is wide; the gelatinous material under tension may be secondary to trauma, synovial rests, or atypical mesenchymal development. CAD commonly presents with unilateral claudication secondary to cyst expansion causing arterial stenosis or, as in our case, possible intramural rupture with proximal extension causing arterial stenosis.

Case Report

A 52-year-old female presented with a 6-month history of sudden onset left-sided claudication. The patient had no risk factors for atherosclerosis. On examination no pulses were palpable in the left lower extremity while all pulses were palpable on the right.

CT angiogram of the lower limbs revealed an eccentric low density wall thickening of the left external iliac artery (EIA)

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producing a 70% stenosis and a cystic lesion just distal to the stenosis (Fig. 1(a) and (b)). Ultrasound confirmed a haemodynamically significant EIA stenosis secondary to intramural thickening and the distal EIA cystic mass (Fig. 1(c)).

A diagnosis of cystic adventitial disease with proximal intramural extension was made and the patient proceeded to iliofemoral bypass. The distal EIA showed a circumferential swelling in the wall (Fig. 1(d)) containing mucoid material.

We elected to perform an iliofemoral bypass using a PTFE conduit with distal anastomosis to the common femoral artery.

Histological examination of the excised cysts revealed a fibrous connective tissue cyst wall and cyst contents of amorphous eosinophilic material (Fig. 2(a) and (b)).

Discussion

Cystic adventitial disease (CAD) patients typically have no evidence of atherosclerosis and CAD is an important differential diagnosis for lower limb ischaemia in younger patients.

The typical distribution is popliteal, with iliofemoral involvement rare.[1] What makes this case interesting is the imaging. The imaging of the proximal and distal EIA shows two distinct appearances. The distal EIA lesion arrowed in Fig. 1(a)–(c) is an eccentric spherical cystic structure in a more typical location for CAD, whereas the more proximal EIA shows an intramural non-cystic linear lesion (arrowheads, Fig. 1(a) and (b)). This led us to conclude that the likely mechanism of the more proximal linear intramural lesion was that it represented intramural cyst rupture, causing sudden onset of symptoms.

Symptoms usually occur when these cysts compress the arterial lumen, reducing arterial flow. In our patient the cystic disease was isolated to the external iliac artery, not causing significant stenosis, but there was additional intramural extension proximal to the cyst and we postulate that it may be due to intramural cyst rupture.

Many imaging modalities may be used to diagnose CAD.[2] MRI provides useful information about cyst contents and can also assess stenosis. Ultrasound may depict a hyperechoic or hypoechoic mass in the wall of the affected vessel, or a mixture of the two, indicating cystic and non-cystic components.[3]

The most effective method of treatment is bypass and excision of the relevant vessel. Various other approaches to treatment have been described including cyst excision and cyst aspiration under ultrasound or CT guidance. Whilst these interventions carry a lower morbidity than bypass and excision of the affected segment, recurrence is more common.[4] Recurrence has also been reported following excision and bypass.[5] In our case we considered endoluminal repair of the proximal EIA, but due to the patient's age and the lack of durability of this option, and the presence of a more proximal EIA lesion this seemed inappropriate as did cyst excision alone. Bypass surgery with excision of the distal EIA cysts was performed.

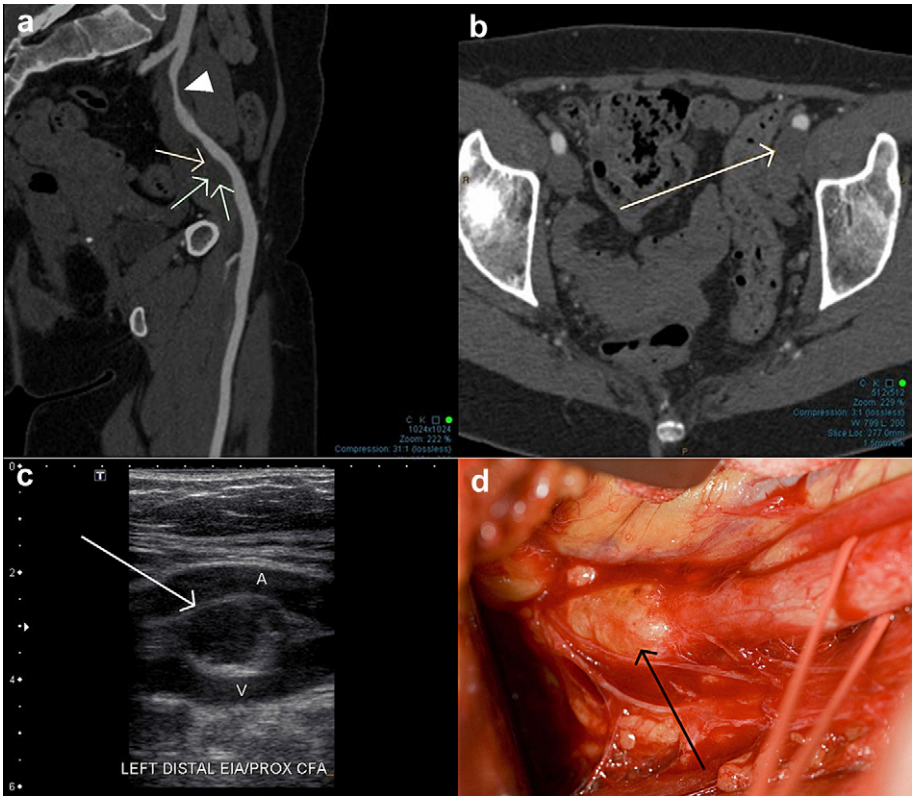

Figure 1 (a) and (b) Curvilinear and transverse CT reconstructions of the EIA. The distal EIA exophytic cyst is arrowed in both images. The haemodynamically significant more proximal EIA intramural extension can be seen in Fig. 1(a) (arrowhead). (c) Sagittal ultrasound scan showing distal EIA cyst (arrow) lying between the vein and adjacent artery. This is at the same location as arrowed in Fig. 1(a) and (b). (d) Intra-operative photo showing distal external iliac artery exophytic cystic mass (arrow).

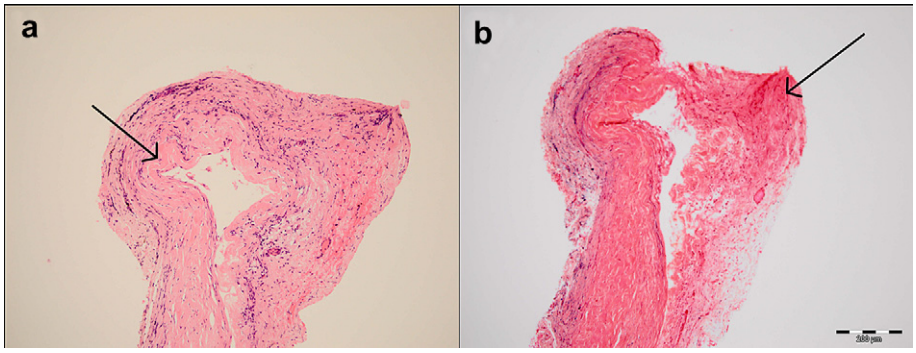

Figure 2 (a) Histology of the cyst wall showing fibrous connective tissue (arrow). (b) Histology of the cyst content containing amorphous eosinophilic material (arrow).

Conclusion

We present a case of CAD complicated by possible intramural extension causing an EIA stenosis and sudden onset of claudication. The diagnosis was confirmed by imaging and histology and treated with excision and bypass.

Conflict of Interest

None.

Funding

None.

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