Cystic Adventitial Disease Of The Common Femoral Artery: A Case Report And Literature Review
Z Gamie, A Navarro, P Curley

Abstract
Introduction: Cystic adventitial disease (CAD) most commonly affects the popliteal artery and few reports indicate that it may also be found in the external iliac and common femoral arteries. Case Presentation: We report the case of a 56-year-old female patient with a history of long distance right calf claudication found to have CAD of the right common femoral artery (CFA) and proximal profunda femoris artery. This was found by duplex sonography but not on magnetic resonance angiography. The lesion persisted after attempted percutaneous drainage and was then excised and the defect repaired with a reversed interposition vein graft. Conclusion: This case highlights the benefit of duplex ultrasound in the diagnosis of CAD, and demonstrates a previously identified shortcoming of MRA. The clinician should have a high index of suspicion for this disease in the absence of risk factors for peripheral vascular disease and should be aware that it not only affects the popliteal artery but arteries in several other locations and has the potential to reoccur.

INTRODUCTION
Cystic adventitial disease (CAD) is a rare disorder of the arterial adventitia involving part or the entire circumference of an artery.[1] CAD most commonly affects the popliteal artery[1] and few reports indicate that it may also be found in the external iliac and common femoral arteries (CFA).[2-4] It is a progressive disorder causing narrowing of the lumen and ischaemic symptoms such as claudication and in a minority can potentially present with limb-threatening ischaemia.[1] Duplex imaging can demonstrate the stenosis as an abnormality in the colour flow pattern and the cyst in the vessel wall. Magnetic resonance imaging (MRI) can demonstrate both the cyst and the extent of the arterial occlusion.[5] Diagnosis can also be made using computed tomography angiography (CTA).[1] Treatment modalities include percutaneous transluminal angioplasty (PTA), percutaneous drainage and cyst excision, with vein interposition or prosthetic grafting.[1, 5, 6] There have been reports of recurrence following PTA,[7] percutaneous drainage[8] or evacuation and cyst excision,[6] and in resection with patch grafting.[9]

We report the case of a patient with CAD of the right CFA and proximal profunda femoris artery (PFA), found by duplex sonography but not on MRA. Attempted percutaneous cyst aspiration was unsuccessful and the lesion was then excised and the defect repaired with a reversed interposition vein graft.

CASE REPORT
A 56-year-old female Caucasian patient was referred with a history of long distance right calf claudication. She complained of pain in her right calf that radiated to the thigh at a distance of 500 yards, which was worse when walking on an incline. Four months previously, she was claudicating at around 4 miles; however, there was no history of rest pains. The pains affected her lifestyle and job as a teacher. She was otherwise active and had a minimal past medical history. She was a lifelong non-smoker and was not on regular medication. On examination her BMI was 26 and she had a full complement of pulses in both legs. Her Ankle Brachial Pressure Index (ABPI) on the right was 190/150 with a biphasic tone and on the left it was 150/150 with a triphasic tone. Bloods investigations including cholesterol were unremarkable.

She underwent an exercise programme and an arterial ultrasound scan was arranged. This revealed a 2.5 x 1.5 by 1.2cm cystic structure, which appeared to arise from the right CFA, causing extrinsic compression on the underlying vessel. There was no evidence of flow within this cystic structure. There were high velocity jets seen at the CFA.
bifurcation as a consequence. Moderate velocity multiphasic flow was seen within the superficial femoral artery (SFA) (Figure 1). Appearances, though not specific, were suggestive of possible focal CAD.

**Figure 1**
Figure 1. Arterial ultrasound scan demonstrating a 2.5 x 1.5 by 1.2cm cystic structure arising from the right CFA, exerting extrinsic compression on the underlying vessel. High velocity jets can be seen at the CFA bifurcation (blue).

The cystic structure was thought to be causing compression symptoms and she underwent ultrasound-guided aspiration. However, at 2 months following the procedure she still complained of claudication symptoms. An MRA scan demonstrated no significant aorta iliac disease and there were normal appearances of both CFAs (Figure 2). There also appeared to be no significant disease affecting the PFA, SFA or the popliteal arteries. The vessels in both calves also had no evidence of stenotic vascular disease. It was arranged for her to have excision of the abnormal cystic right CFA and proximal PFA and reconstruction with a reversed upper long saphenous vein (LSV) graft (Figures 3a, 3b and 3c).

**Figure 2**
Figure 2. MRA scan demonstrating no significant aorta iliac disease and normal appearances of both CFAs.

**Figure 3**
Figure 3. (A) CAD of the CFA. Slings placed around the CFA (yellow), SFA (blue), PFA (white) and cyst (red). (B) The resected cyst containing gelatinous material. (C) A reverse saphenous interposition was placed, which resulted in normal leg perfusio

Postoperatively, her foot was well perfused and she had good popliteal and posterior tibial pulses. She was
discharged on day 1. Histology revealed a multilocular mucous filled cyst measuring approximately 15 x 8 x 7mm. Within the adventitia there were several cystic spaces with a fibrous tissue wall, devoid of an epithelial lining and containing proteinaceous debris and occasional macrophages. There were also several pools of mucin admixed with inflammatory cells and foamy macrophages. The features were compatible with cystic adventitial degeneration.

**DISCUSSION**

The male to female ratio of CAD is 5:1 and the patient is classically in the fourth to the fifth decade.[1] About 15% of CAD cases are described in extrapopliteal locations,[10] the next commonest site is the femoral artery followed by the radial and iliac arteries.[2] The first case of CAD in the CFA was described by Jaquet and Meyer-Burgdoff in 1960.[11] There have been 25 cases of CAD in the CFA reported in the literature to date, a greater proportion have been in male patients and most cases are in the 40-50 year age category (Figures 4a and 4b).

**Figure 4**

Figure 4. (A) Incidence of CAD in the CFA reported in the current literature per age category. Arrow demonstrates the category for our patient. (B) CAD in the CFA has been reported in 20 male and 5 female patients.

A number of hypotheses have been proposed for the aetiology of these lesions. The most popular theories include the synovial theory, in which the herniation of the capsule of the adjacent joint involves the adventitia of the artery,[1] and the developmental theory, in which mucin-secreting cells derived from the mesenchyme of the adjacent joint are also found in non-axial blood vessels close to the joint.[1, 12] The gold standard imaging modality is angiography but noninvasive imaging modalities such as duplex ultrasound, MRA and CTA have been suggested as new diagnostic alternatives.[13] Abnormalities present on imaging, include an indented artery (scimitar sign), an encircled artery (hourglass sign), or a completely occluded artery.[5] In our case, a noninvasive investigation, duplex ultrasound, was performed and revealed a cystic structure, which appeared to arise from the right CFA. The MRA scan was unable to demonstrate an abnormality, a limitation which has previously been reported.[9] Total resection with grafting is the recommended mode of treatment and can achieve good results.[2, 6]

For CAD in the popliteal artery, there is a reported high recurrence rate after PTA[7] and percutaneous drainage.[8] There has also been reported failure of treatment following endovascular repair.[14] There is a possibility of recurrence in an interposition vein graft used for treatment of CAD in the popliteal artery[15] and recent reports have advocated the use of a PTFE graft for its treatment.[5] However, long term outcome is unknown and there may be failure at the anastomosis due to recurrence of CAD.[5] Methods used for treatment of CAD of the CFA have included cyst aspiration or excision with or without patch repair, arterial resection, replacement with a reversed interposition vein graft, and replacement or bypass of the affected segment with synthetic graft (Figure 5).

**Figure 5**

Figure 5. Modes of treatment of CAD in the CFA currently reported in the literature.

Those treated with cyst aspiration or excision commonly recurred and some were subsequently treated with resection and replacement with a vein graft[16] or prosthetic interposition graft.[6] There has also been reported recurrence after resection of CAD and prosthetic patch repair of the CFA.[9] Recurrence in some cases may have arisen from the neighboring joint structures or the deposition of macroscopic foci during the initial resection procedure and the inherent difficulties of completely removing the synovial cells, therefore intensive follow-up is recommended.[15] In the current literature there are a few cases that have reported long term patency following cyst excision and use of a reversed interposition vein graft for CAD of the CFA.[2, 3, 17] In our case we elected for total cyst excision with the involved artery and a reversed saphenous vein interposition.
CONCLUSION

This case highlights the benefit of duplex ultrasound in the diagnosis of CAD, and demonstrates a previously identified shortcoming of MRA. The clinician should have a high index of suspicion for this disease in the absence of risk factors for peripheral vascular disease and should be aware that it not only affects the popliteal artery but arteries in several other locations and has the potential to reoccur.

ABBREVIATIONS

CAD: Cystic Adventitial Disease; SFA: Superficial Femoral Artery; CFA: Common Femoral Artery; PFA: Profunda Femoris Artery; ABPI: Ankle Brachial Pressure Index; MRA: Magnetic Resonance Angiography; CTA: Computed Tomography Angiography; LSV: Long Saphenous Vein.

References

Author Information

Zakareya Gamie, BSc, MRCS
Department of General and Vascular Surgery, Pinderfields General Hospital

Alex P. Navarro, MRCS, PhD
Department of General and Vascular Surgery, Pinderfields General Hospital

Paul J. Curley, MB BCh, FRCS
Department of General and Vascular Surgery, Pinderfields General Hospital