Case Report

Purpose: Cystic adventitial artery disease is an uncommon non-atherosclerotic peripheral vessel disease. Furthermore, cystic adventitial disease of the common femoral artery is an extremely rare entity. We report the case of a 54-year-old man complaining of intermittent claudication who was referred to our vascular service.

Methods and Results: Doppler ultrasound and multidetector-row computed tomography (CT) with 3-dimensional volume rendering revealed severe stenosis with cystic adventitial cyst in the common femoral artery. Intra-operative Doppler ultrasound showed the cyst to be multilocular type. Reversed great saphenous vein interposition was successfully placed.

Conclusion: Removal of cyst together with artery and interposition using reversed great saphenous vein is the optimal treatment procedure to prevent recurrence.

Keywords: cystic adventitial cyst disease, common femoral artery, Doppler ultrasound, bypass surgery

Introduction

Cystic adventitial artery disease (CAAD) is a rare entity and that in most patients involves popliteal artery. Desy and spinner described 587 cases of CAAD of popliteal artery in a cohort of 724 CAAD patients therefore CAAD of the common femoral artery (CFA) is an extremely rare entity.1–3 The features of this disease include intermittent claudication with middle-aged patients in the absence of atherosclerotic risk factor and finds a marked male predominance with a ratio of approximately 4:1.3 The etiology of this disease is still controversial. We reported the diagnosis and operative procedure for a case of CAAD of the CFA.

Case Report

A local physician referred a 54-year-old man to our vascular service complaining of a 3-months history of steadily progressed intermittent claudication involving the left leg after 5–10 min walking on level ground at normal speed. The patient had diabetes mellitus, history of attack of gout with hyperuricemia and no history of trauma.

On the physical examination, femoral artery and below the knee pulsations were diminished and normal femoral and below the knee pulses were palpable on the right leg. The ankle brachial index (ABI) showed 0.6 on the left side. Doppler ultrasound (DUS) revealed a severe stenosis of the eccentric circumference of CFA with a smooth arterial wall and cystic formation compressing the endolumen (Fig. 1a). Multidetector-row CT (MDCT) with 3-dimensional volume rendering in the evaluation of the lower extremity showed the local severe stenosis of the left femoral artery so called scimitar sign without further
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signs of atherosclerosis and then there were no cystic lesion of circumference of artery below common iliac artery (Fig. 1b). The lesion was diagnosed as cystic adventitia disease of femoral artery.

Under the general anesthesia a longitudinal incision was made in the left groin. The CFA was swollen, 3 cm in length and had no pulsation. Fluid collection was found through adventitia (Fig. 2a). Vascular clamps were applied to the proximal and distal common femoral artery and then incision of adventitia on the cyst was made and the mucinous specimen was then removed (Fig. 2b). After vascular clamps were removed, the pulsation of the distal common femoral artery was restored. However, intra-operative DUS detected another cyst in the posterior wall of the CFA which was diagnosed as the multilocular type. It was decided to carry out resection of the cystic lesion involving CFA. And reversed great saphenous vein graft interposition was successfully placed. Pathologic examination of the surgical specimen with stained elastica van Gieson revealed cystic space located in the adventitia (Fig. 3). The patient left hospital without sequelae. At 3 months follow up, the patient was free of symptoms and the graft remained patent without recurrence on the MDCT.

Discussion

CAAD is an uncommon non-atherosclerotic peripheral vessel disease furthermore CAAD of CFA is an extremely rare entity. CAAD was first described in 1947 by Atkins and Key. The incidence of CAAD is estimated to be 1 in 1200 cases of claudication, with cases described involving external iliac, femoral, popliteal, radial, and ulnar arteries and veins.

The literature has reported various hypotheses regarding pathogenesis of cystic adventitial disease as follows: the trauma theory; repetitive trauma, the ganglion theory; synovial cyst implant in the adventitia, the systemic disorder theory; degeneration of connective tissue, the developmental theory; mucin secreting cells placed in the adventitia from nearby joint. The patient showed no relationship between CFA and joint and there were no urate crystals in the surgical specimen, therefore obvious causes have not yet been identified.

Fig. 1 (a) Doppler ultrasound imaging severe stenosis of common femoral artery lumen compressed with cystic lesion. (b) MDCT (multidetector-row CT) with 3-dimensional volume rendering shows scimitar sign without further additional features of atherosclerosis.

Fig. 2 (a) Mucoid collection in the adventitia after incision at adventitial level of the cyst. (b) Intraoperative Doppler ultrasound revealed another cyst in the posterior of the common femoral artery.
The CAAD was diagnosed by imaging employing DUS and MDCT with 3-dimensional volume rendering or magnetic resonance angiography (MRA). Ismaeel et al. have reported that high spiral MRI with MRA is useful for identification of connections between adventitial cysts and the adjacent joint.6)

In this case after the physical examination, firstly, DUS detected severe stenosis that involved the cystic formation compressing the endolumen by cyst formation. Secondary, MDCT revealed a form of stenosis described as the scimitar sign in the left CFA without further signs of atherosclerosis. DUS provides useful information in suggesting CAAD less invasively subsequent MDCT with 3-dimensional volume rendering and MRA excludes a systemic atherosclerotic lesion.8)

Open surgery included removal of cystic lesion with CFA and subsequent interposition using great saphenous vein graft is the best therapeutic option for CAAD. Percutaneous cystic aspiration under US guide is not always possible because of the high viscosity of the content and multilocular cyst and a high recurrence rate has been described due to the mucin-secreting cells which still remain.3,9–11)

Endovascular treatment (EVT) is not only effective for CAAD but also for injured healthy arterial intima. EVT is not recommended for CAAD.

**Conclusion**

Cystic adventitial disease of CFA is a rare entity that in most patients involves the popliteal artery. As a diagnosis modality, DUS provided far more accurate physiopathological information than other imaging modalities. Intra-operative DUS is useful for the diagnosis of a multilocular cyst. Removal of cyst with artery and interposition using reversed great saphenous vein is best optimal procedure to prevent recurrence.

**Disclosure Statement**

Authors have no conflicts of interest.

**References**