A Case of Adventitial Cystic Disease of the Popliteal Artery Progressing Rapidly after Percutaneous Ultrasound-guided Aspiration

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Adventitial cystic disease is a rare non-atherosclerotic vascular disease. We report a 36-year-old man with right intermittent claudication by adventitial cystic disease. Computed tomography (CT) and magnetic resonance imaging (MRI) revealed an ovoid cystic mass compressing the right popliteal artery and causing severe stenosis of the lumen. Percutaneous aspiration was performed, which improved his symptoms. However, he complained of identical intermittent claudication two weeks later. Radiographic findings revealed that the cystic lesion had progressed rapidly. The cystic lesion was resected and the affected arterial segment was interposed. We consider that conventional surgical intervention remains the favored treatment option in the management of adventitial cystic disease.

Keywords: adventitial cystic disease; popliteal artery; percutaneous aspiration

Introduction

Adventitial cystic disease (ACD) is a rare vascular condition characterized by non-atherosclerotic vascular disease and a collection of mucinous material within the adventitia of the affected vessel. The adventitial mucoid cyst, which compresses the arterial lumen, subsequently causes intermittent claudication or limb ischemia. The symptoms usually occur suddenly as ACD often progresses rapidly. Thus, vascular surgeons should consider the diagnosis of ACD for adult male patients with recent onset of intermittent claudication in the lower extremities without evidence of atherosclerotic disease. There are various treatments for ACD, including surgical treatment, percutaneous aspiration and percutaneous transluminal angioplasty (PTA).

Herein, we describe a case of ACD progressing rapidly after percutaneous ultrasound-guided aspiration.

Case Report

A 36-year-old male presented with a 2-month history of intermittent right calf claudication when walking approximately 100 m. There was no history of trauma, manual labor, or risk factors of cardiovascular disease. On physical examination, normal arterial pulse was present bilaterally in the femoral, popliteal, dorsalis pedis, and posterior tibial arteries. However, loss of foot pulses with knee flexion was present on the affected side. His ankle-brachial systolic pressure index (ABI) was 0.89 on the right side and 1.10 on the left side. Other clinical and laboratory findings were unremarkable. Doppler ultrasound (US) examination revealed a 20-mm longitudinal hypoechoic cyst outside and behind the right popliteal artery and extrinsic compression of the popliteal artery caused by the cyst (Fig. 1A). Enhanced contrast computed tomography (CT) revealed a 21-mm severe stenosis of the right popliteal artery, compressed segmentally by a non-enhanced cystic lesion within the arterial wall (Fig. 2A). No atherosclerotic findings were confirmed.
in the artery. Magnetic resonance imaging (MRI) revealed a 20-mm multi-lobulated cystic lesion close to the popliteal artery wall showing muscle iso-intensity on T1-weighted image and high signal intensity on T2-weighted image (Fig. 2B). On the basis of the clinical and imaging findings, a diagnosis of ACD was made.

We scheduled ultrasound-guided needle aspiration of the cyst under local anesthesia. Approximately 2 mL of gelatinous material with high viscosity was aspirated (Fig. 1B). US revealed reduction of the cyst with dilatation of the lumen of the right popliteal artery immediately after the aspiration. After US-guided aspiration, the ABI improved to 1.03 on the right side, and his symptom disappeared. However, two weeks after the aspiration, he suffered from identical intermittent right calf claudication. The right side ABI worsened to 0.47. Enhanced contrast CT and MRI revealed that the cystic lesion had progressed rapidly, with a 42-mm longitudinal occlusion of the right popliteal artery (Fig. 2C, D). Thus, we performed surgical intervention.

At surgery, the patient was placed in a prone position, and the right popliteal artery was exposed through a posterior approach. A round, loculated cyst with well-defined margins was identified, and we resected the affected arterial segment and interposed an expanded polytetrafluoroethylene graft (Fig. 3A). There were no communications between the cystic lesion and the knee joint. Histological examination of the surgical specimen showed that the gelatinous material was located between the adventitia and the middle layer of the artery, and that the inner cavity of the cyst was filled with a mucinous gel-like content. These findings were consistent with ACD (Fig. 3B). His postoperative course was uneventful and the
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right calf remained asymptomatic 4 months after surgery.

Discussion

ACD is a rare vascular disorder, and was first reported in a case of a myxomatous tumor arising from the external iliac artery.\(^1\) Cysts in ACD are typically unilocular and contain mucin and hyaluronic acid. The compression of the cysts that develop within the adventitia causes stenosis or occlusion of the vessel lumen.

Clinical diagnosis of ACD is often difficult. ACD usually shows normal circulatory exploration in the lower extremities, with palpable peripheral pulses, although they might be diminished or absent depending on the grade of stenosis. Currently, conventional catheter angiography is not usually performed, and has been replaced by other angiographic images including US, CT, and MRI, which have the capacity to show both the lumen and the lesion within the vessel wall. Doppler US should be the first choice as it is easily available, rapidly performed, non-invasive and inexpensive. Moreover, it is highly sensitive in demonstrating ACD. CT and MRI are also frequently used in the diagnosis of ACD. Of these, MRI has the best its sensitivity and specificity to exclude other pathologies included in the differential diagnosis of the affected popliteal fossa. In the present case, the findings on US, CT, and MRI enabled us to make a presumptive diagnosis of ACD preoperatively, avoiding the need for conventional angiography.

Although the underlying etiology of ACD remains unknown, there are a number of proposed mechanisms including a systemic disease theory, repetitive trauma theory, synovial or ganglion theory, and embryological theory.\(^2\) Cases of ACD communication to the knee joint have been reported,\(^3,4\) indicative of a synovial or ganglion theory. However, in the present case, there was no communication between the cystic lesion and the knee joint. More recently, Levien, et al. proposed a unifying theory in support of an embryological etiology, which was based on a literature review.\(^2\) In that study, ACD was postulated to be caused by the incorporation of mesenchymal cells into the adventitia of nonaxial vessels from the adjacent joint tissue during development. This embryological theory is now the most convincing and supported theory.

Although spontaneous resolution of symptoms and lesions was previously reported,\(^5\) this is extremely rare and whether this resolution is permanent remains unclear. Currently, surgical treatment is a standard management for ACD. The main surgical treatments involve resection of the affected segment with interposition of an autologous vein graft or prosthetic graft, cyst excision, and cyst evacuation. In young patient, an autologous vein graft would have been the preferred choice. However, in the present case, the patient requested an artificial graft. Therefore, we resected and interposed the affected vessel using a prosthetic graft in accordance with the patient’s wish. In the majority of cases, surgery requires a posterior approach of the popliteal artery, as the affected

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Fig. 3 (A) Intraoperative findings showed that popliteal artery was surrounded by a loculated cyst. (B) Surgical specimen of the adventitial cyst. Note the mucoid degeneration (arrowhead) and the residual lumen of the popliteal artery.
segment is usually the middle portion of the popliteal artery. Flanigan, et al. reported that simple evacuation of the cyst is the preferred treatment unless the process has progressed to chronic occlusion and thrombosis, in which case a bypass is preferred. It was reported that the initial success rate of surgical resection was approximately 95%, and that the risk of recurrence was rare after surgery. In cases where communication with the adjacent joint is noted preoperatively, the connections should be ligated for preventing recurrence.

There are also alternative minimally invasive procedures for treatment of ACD such as percutaneous needle aspiration. Aspirating the cyst content under image guidance may be a simple alternate. There are several reports that CT- or US-guided aspiration was successful without recurrence. However, it was reported that the initial success rate of image-guided percutaneous cyst aspiration was much lower than that of surgery. The failure of image-guided aspiration may attributed to incomplete aspiration due to high viscosity of the cyst content. Further, as the cyst is not resected in image-guided aspiration, the residual cystic wall can continuously secretes mucinous fluid and result in recurrence. In the present case, the cystic lesion progressed rapidly after aspiration, which may relate to excessive mucinous fluid secretion by the residual cystic wall. It was also suggested that PTA was not an effective treatment for ACD because of the extraluminal nature of ACD. Accordingly, the incidence of recurrence after such less invasive treatments should be higher. However, recurrence can occur with all treatment approaches, including surgery, and careful follow-up is needed.

In conclusion, we consider that conventional surgical intervention, either resection with bypass or excision/evacuation of the cyst, remains the most effective treatment for the management of ACD.

## Disclosure Statement

We declare that we have no conflict of interest with respect to the publication of this paper.

## References