



Inadvertent Stenting and Percutaneous Aspiration for Treatment of Adventitial Cystic Disease in the Popliteal Artery: A Case Report

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Adventitial cystic disease (ACD) is a rare, non-atherosclerotic disease that mainly affects the popliteal artery. Treatment is primarily surgical as endovascular approaches are affected by high recurrence rates. However, some studies have reported successful endovascular treatments of popliteal ACD cases. A 55-year-old female presented with right calf claudication. Computed tomography angiography revealed segmental occlusion of the right distal superficial femoral artery. Subsequently, a drug-eluting stent was successfully deployed. However, an unusual adventitial cystic lesion occluding the lumen that was characteristic of ACD was detected during a postoperative imaging review. It was aspirated using an ultrasound-guided percutaneous needle and drained using a pigtail catheter for 24 hours. Follow-up images after 39 months showed a patent artery with no recurrence of any cystic lesions, highlighting successful ACD treatment via stenting, ultrasound-guided aspiration, and cyst drainage. Stenting and cyst aspiration can be an alternative option for selected patients with ACD.

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INTRODUCTION

Adventitial cystic disease (ACD) is a rare non-atherosclerotic disease that affects various arteries and veins, particularly the popliteal artery (PA). Patients are typically young and middle-aged and often have no risk factors for atherosclerosis [1,2]. ACD causes accumulation of mucus material under the adventitia of blood vessels causing narrowing of those vessels and claudication [3]. For young patients with intermittent claudication and no risk factors for peripheral vascular disease, it is important to differentiate between ACD, PA entrapment syndrome (PAES), PA aneurysm, and fibromuscular dysplasia. Imaging studies, including ultrasonography, computed tomography angiography (CTA), and magnetic resonance imaging (MRI) are essential diagnostic

tools for ACD. Treatment is primarily surgical, although endovascular interventions have been reported. Endovascular treatments are not recommended due to poor outcomes because of high recurrence rates [4]. However, several studies have reported successful treatment using stenting in some ACD cases [5,6]. Herein, we report a case of popliteal ACD treated with stenting followed by ultrasound-guided percutaneous needle aspiration and cyst drainage. This case report was approved by the Institutional Review Board of the Seoul National University Hospital (IRB no. 2102-107-1197).

CASE

A 55-year-old female presented to the outpatient clinic

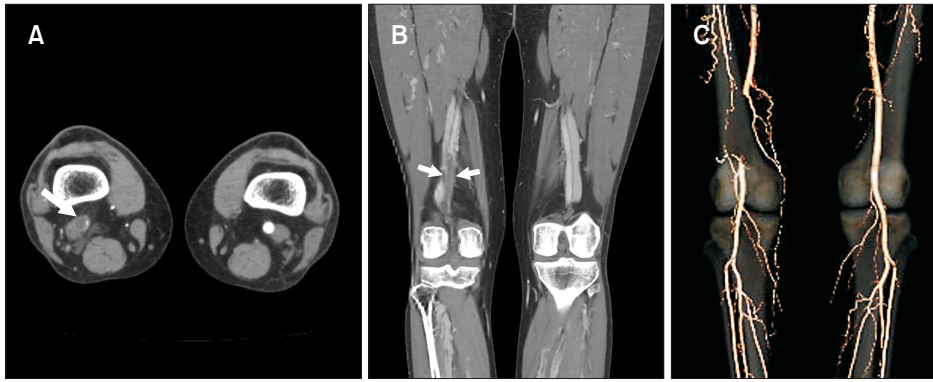


Fig. 1. Pre-interventional computed tomography scan showed occlusion of the distal superficial femoral artery to the proximal popliteal artery occlusion (arrows). (A) Axial image. (B) Coronal image. (C) Three-dimensional volume rendering anteroposterior view.

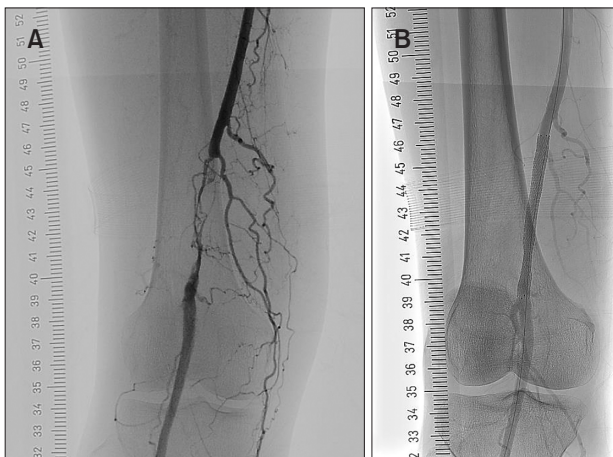


Fig. 2. Treatment of adventitial cystic disease using stent placement. (A) Angiogram confirmed a segmental right distal superficial femoral artery to proximal popliteal artery occlusion. (B) After stenting, the occluded segment was well recanalized.

with severe right calf claudication after walking 100 m which had started 7 months earlier (Rutherford class 3). The patient had no comorbidities except for hypertension. The patient's ankle-brachial index (ABI) was measured as 0.81/1.21 (right/left). CTA revealed a segmental right distal superficial femoral artery to proximal PA occlusion (Fig. 1). Digital subtraction angiography confirmed the CTA findings (Fig. 2A). Balloon angioplasty was performed several times with a 5 mm×8 cm Mustang (Boston Scientific Co., Natick, MA, USA) in the narrowed segment; however, significant residual stenosis of >50% was consistently observed. A 6 mm×8 cm drug-eluting stent (Eluvia, Boston Scientific Co.) was deployed resulting in good recanalization (Fig. 2B). When crossing the occluded lesion, a 0.014" Command ES guidewire (Abbott Vascular, Santa Clara, CA, USA) and 0.018" Rubicon™ 18-Support Catheter (Boston Scientific, Marlborough, MA, USA) were used with no difficulty in intraluminal passage.

However, a postoperative image review during a mul-

tidisciplinary conference revealed a cystic lesion with diameter of 9.3 mm compressing on the right PA which was compatible with ACD. Therefore, to reduce external compression and recurrence the cyst was drained using ultrasound-guided percutaneous needle aspiration with a 22-G spinal needle and 6-Fr pigtail catheter. At the proximal PA, a well-defined, hypoechoic, unilocular, non-vascular mass surrounding the vessel was identified. Aspiration was performed with a syringe containing contrast medium. A small amount of gelatinous substance containing debris lighter than the contrast medium was aspirated (Fig. 3). After drainage for 24 hours using a pigtail catheter, follow-up ultrasound revealed no remaining cysts and the catheter was removed immediately. The patient's ABI improved to 1.10/1.18. Recovery was uneventful, and the patient was discharged with aspirin and cilostazol. Follow-up duplex ultrasonography at 39 months showed a patent artery and no signs of cyst recurrence (Fig. 4).

DISCUSSION

ACD is a rare condition characterized by mucus-containing cysts that develop in the adventitial layers of the blood vessels. When a cyst compresses an artery it causes narrowing or occlusion which fosters claudication symptoms similar to other arteriosclerotic diseases. ACD incidence is estimated to be 1 in 1,200 cases of claudication. ACD develops near joint spaces and can occur in the external iliac, femoral, popliteal, radial, and ulnar arteries or iliac and femoral veins [7,8]. If a cyst is compressing a vein, swelling may also occur. However, owing to the rarity of ACD, there are no definitive treatment guidelines meaning many treatment options have not yet been validated.

There are several different treatment options for ACD, including percutaneous cyst aspiration with or without sclerotherapy, cyst excision, and vascular resection with an interposition graft or bypass. Although it is necessary to understand etiology to explain the differences in vari-

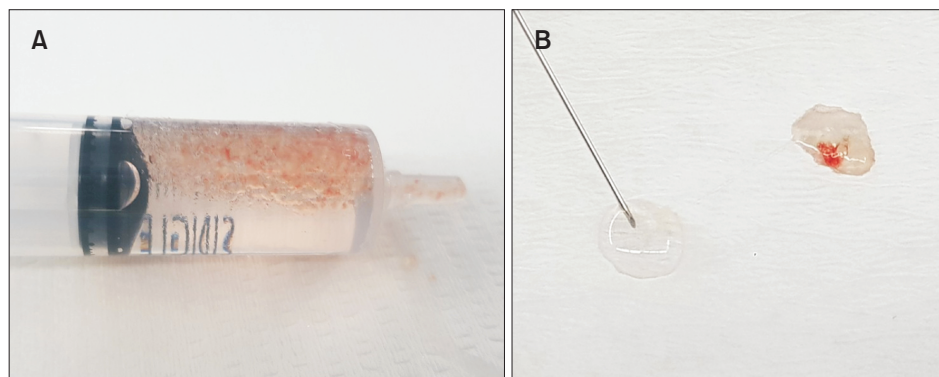


Fig. 3. Cyst aspiration with a syringe containing contrast medium. (A) A small amount of gelatinous substance containing debris lighter than the contrast medium was aspirated. (B) Gelatinous fluid on the right is easily distinguishable from the contrast medium on the left.

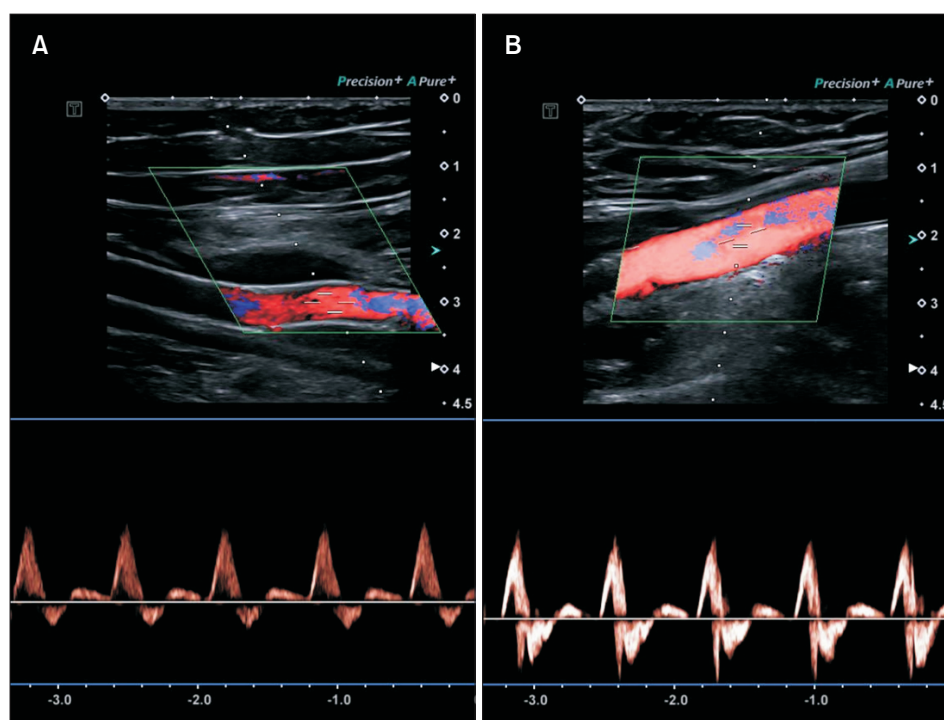


Fig. 4. Follow-up duplex ultrasonography 39 months postoperatively revealed normal flow within the right distal superficial femoral artery (A), and popliteal artery (B).

ous treatment methods, it is not precisely known how and why these cysts form. Various theories on the etiology and pathogenesis of ACD have been proposed, such as the repetitive trauma theory, systemic disorder theory, developmental theory, and articular theory [9,10]. However, considering ACD pathogenesis, the articular theory seems most appropriate. Based on this theory, ligation of the connection between the joint capsule and adventitial cyst is essential to prevent recurrence [11]. Cysts commonly had communication channel with the adjacent joint along with the capsular branch of the affected artery, most commonly the middle genicular artery [12,13].

Lim et al. [14] reported that vessel excision and interposition grafting are better at preventing ACD recurrence than simple cyst excisions. Therefore, treatments described in previous reviews have mostly been surgical with cyst

drainage for expansion of the arterial lumen or arterial reconstruction using bypass [15,16]. Surgical managements are thought to provide the good long-term patency and outcome with low recurrence rate of ACD after revascularization surgery (0%-10%) [2,15,17]. Whereas endovascular techniques, such as balloon angioplasty, are affected by high failure rates and are seldom used [18,19]. Khoury [18] reported stenosis of a PA near the knee joint diagnosed as ACD. It was treated using balloon angioplasty only in which restenosis was identified within 24 hours. In another case, the lesion was located above the knee and stenting was performed without cyst aspiration. Consequently, an interposition graft was required one week later because the stent became occluded [19].

Nonetheless, endovascular treatment achieved no recurrence for >3 years in this case. After cyst aspiration,

good decompression was attained. It is thought that no recurrence occurred because the joint connection was compressed while expanding the stent. Needle aspiration and puncture is a less invasive approach than surgery and can decrease the size and symptoms of cysts. When the involved vessel is not occluded, cyst aspiration can be initially performed without leaving a scar. In addition, if the affected vessel does not cross a joint, stenting can also be considered as a preliminary stage of surgery with caution. If follow-up imaging studies reveal that the patient's symptoms have reoccurred after employing these approaches, re-aspiration or radical surgery should be performed after sufficient discussion with the patient [9].

We explained to the patient that surgery could be performed if the cyst reoccurred after preferentially trying an easier method first. Fortunately, it did not recur so both the patient and doctors were satisfied. Since surgical operation was not performed, joint connection could not be identified; therefore, ACD could not be definitively confirmed. However, considering the imaging findings and contents of the cyst, an ACD diagnosis was regarded accurate.

Making a general recommendation for this rare disease based only on this patient is difficult. However, primary cyst aspiration including stenting can be a reasonable option for selected patients. In the past, there have been many opinions stating that simple aspiration is not helpful [20]. Nevertheless, low risk cases using aspiration with good outcomes are gradually being reported meaning aspiration can be considered as an initial treatment option.

Notably, our diagnostic process was unsatisfactory. In a relatively young patient with vascular occlusion without atherosclerotic findings, diagnoses of non-atherosclerotic PAD, including ACD or PAES, should be ruled out. In this case, additional preoperative ultrasound or MRI examinations would have helped for differential diagnosis. Fortunately, after the procedure, the possibility of ACD was confirmed through multidisciplinary team meetings. Multidisciplinary team approaches are mandatory for modern

vascular surgery.

In conclusion, although endovascular techniques such as stenting are generally considered inferior to surgery in the treatment of ACD, stenting and cyst aspiration can be an alternative and viable option for selected patients, whose lesion is unilocular cyst, facilitating decompression or is placed above the knee and not across the joint making it suitable for stenting, and no definite joint connection. However, serial follow-up imaging is mandatory to detect possible recurrence.

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CONFLICTS OF INTEREST

The authors have nothing to disclose.

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Concept and design: SKM. Analysis and interpretation: HYW. Data collection: HYW, SH, HJJ. Writing the article: HYW. Critical revision of the article: SKM. Final approval of the article: all authors. Statistical analysis: none. Obtained funding: none. Overall responsibility: SKM.

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