Case Reports

Journal of INTERNATIONAL MEDICAL RESEARCH

Journal of International Medical Research
50(3) 1–7

DOI: 10.1177/03000605221086149
journals.sagepub.com/home/imr
SAGE

Treatment failure after rotational atherectomy and balloon angioplasty in recurrent cystic adventitial disease of the popliteal artery: a case report

Jin Woo Yoon[1], Woosun Choi[1], Suh Min Kim[2] and Joonho Hur[1]

Abstract

Cystic adventitial disease (CAD), which usually affects the popliteal artery, is a rare vascular condition in which fluid accumulates in the sub-adventitial layer, compressing the lumen. Historically, surgical treatment is preferred over endovascular or minimally invasive techniques, due to its lower recurrence rates. Here, the case of a 67-year-old male patient, in whom rotational atherectomy was performed for recurrent CAD following surgical cyst excision and patch angioplasty is reported. The patient's symptoms recurred one day after the rotational atherectomy procedure and repeat computed tomography angiography showed recurrence of the disease. Due to gradual worsening of the condition during 8 months of follow-up, left distal femoral artery to popliteal artery (below-the-knee) bypass surgery was performed using an ipsilateral reversed great saphenous vein graft. Follow-up has continued for 2 years without complications or requirement of additional treatment. This novel case is the first report of atherectomy attempted for recurrent CAD that led to an early recurrence. Our experience emphasises that additional surgical approaches should be selected over endovascular procedures for treating recurrent CAD.

[1]Department of Radiology, Chung-Ang University Hospital, Chung-Ang University College of Medicine, Dongjak-gu, Seoul, Republic of Korea
[2]Department of Surgery, Chung-Ang University Hospital, Chung-Ang University College of Medicine, Dongjak-gu, Seoul, Republic of Korea

Corresponding author:
Woosun Choi, Department of Radiology, Chung-Ang University Hospital, Chung-Ang University College of Medicine, 102, Heukseok-ro, Dongjak-gu, Seoul 06973, Republic of Korea.
E-mail: radiochoi527@cau.ac.kr

Keywords
Peripheral arterial disease, atherectomy, angioplasty, popliteal artery, intermittent claudication, recurrence

Date received: 9 November 2021; accepted: 18 February 2022

Introduction

Cystic adventitial disease (CAD) is a rare non-atherosclerotic condition in which fluid accumulates in the sub-adventitial layer, compressing the lumen of the arteries and veins. In 80–90% of cases, CAD is located in the popliteal artery, where it may cause intermittent claudication and critical limb ischaemia.[1,2] Although several techniques, including surgical and interventional methods, have been used to treat this disease, surgical resection and graft implantation have been reported as the most reliable methods in many cases, with a recurrence rate of 1–7%.[1,3] Treatment with percutaneous transluminal angioplasty (PTA) appears unsatisfactory due to high recurrence rates,[4] however, the use of an atherectomy device has not been previously reported as a potential treatment method. Furthermore, rare cases of recurrence have been reported even after surgical treatment, but few reports have explored an appropriate treatment in the case of recurrence.[3]

Here, the case of a patient in whom rotational atherectomy was performed to treat recurrent CAD, following initial treatment with cyst excision and patch angioplasty surgery, is described. A review of the literature indicated that this is the first published case in which atherectomy was performed for the treatment of CAD in general, and particularly for recurrent CAD after surgery.

Case report

Written informed consent for publication of this case was obtained from the patient, and the patient provided written informed consent prior to the intervention described herein. As the present study is a descriptive and retrospective case report, ethics committee approval was not required. The reporting of this study conforms with CARE guidelines.[5]

A 67-year-old male patient, previously treated for CAD of the left popliteal artery, presented to the emergency department of Chung-Ang University Hospital, Dongjak-gu, Seoul, Republic of Korea, in August 2019, with acute severe claudication (Rutherford classification 3) in the left limb that had started on the previous day. He had been treated for CAD by excision of the cyst and patch angioplasty three months previously. One month after this initial surgery, occlusion of the treated artery was suspected on ultrasonography, and he underwent percutaneous thrombectomy, with successful removal of the thrombus using a manual aspiration catheter. The patient also had a medical history of hypertension, dyslipidaemia, and heavy smoking. On clinical examination in the emergency department, claudication on the left limb was induced within 3 to 5 minutes of walking. His left dorsalis pedis arterial pulse was weak and his left ankle felt cold, but his sensory and motor functions were intact without colour change. Computed tomography angiography (CTA) imaging showed that the popliteal artery was almost completely occluded (Figure 1). Other arteries, such as the external iliac, common femoral, and right popliteal arteries, were unremarkable except for atherosclerotic changes. Based on the CTA

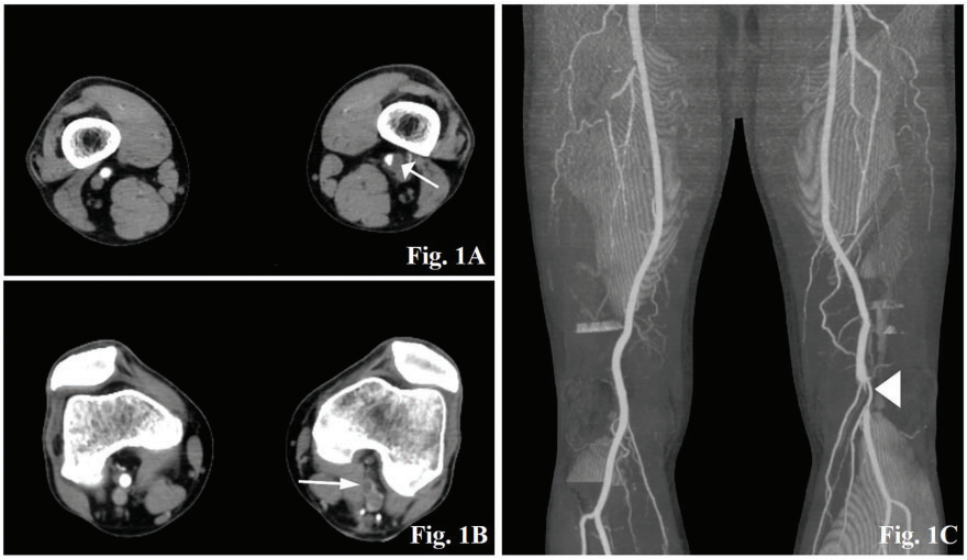

Figure 1. Computed tomography angiography (CTA) images of the lower extremity, showing: (a) preoperative initial axial image above the knee revealing a cystic mass (white arrow) compressing the left popliteal artery; (b) repeat CTA image performed 3 months after cyst excision and patch angioplasty, revealing near-complete occlusion of the popliteal artery at the level of the knee (white arrow) and normal right popliteal artery; and (c) coronal maximal intensive projection image, also performed at 3 months after cyst excision and patch angioplasty, revealing short-segment non-opacification of the left popliteal artery at the level of the knee (white arrowhead).

findings and the patient's history, the obstruction was suspected to be caused by a recurrent thrombotic occlusion. Therefore, an emergency angiography and additional interventions were performed.

Left lower-extremity arteriography was performed through an antegrade puncture of the left common femoral artery, which confirmed narrowing and thrombotic occlusion of the popliteal artery (Figure 2). In addition, because colour Doppler ultrasound, performed during angiography, revealed a cystic lesion without a Doppler signal inside the popliteal artery, the narrowing was considered likely to be accompanied by CAD recurrence. Therefore, rotational atherectomy was performed as a treatment to solve both the thrombotic occlusion and the recurrence. After placing a Spider FX embolic protection device (Covidien; Plymouth, MN, USA) in the distal popliteal artery, rotational atherectomy was performed using the Jetstream XC (Boston Scientific; Natick, MA, USA). Additionally, Passeo-18 Lux (BIOTRONIK AG; Buelach, Switzerland) drug-coated balloon angioplasty was performed. Post-procedure angiography showed that atherectomy and balloon angioplasty had resulted in a satisfactory increase in the luminal diameter (Figure 2d). However, the patient's symptoms recurred one day after the rotational atherectomy and balloon angioplasty, and repeat CTA showed recurrent occlusion of the popliteal artery with an intraluminal cystic lesion, suggesting recurrence of the disease. Despite the recurrence, the degree of claudication improved after the atherectomy procedure. Therefore, the patient was discharged with

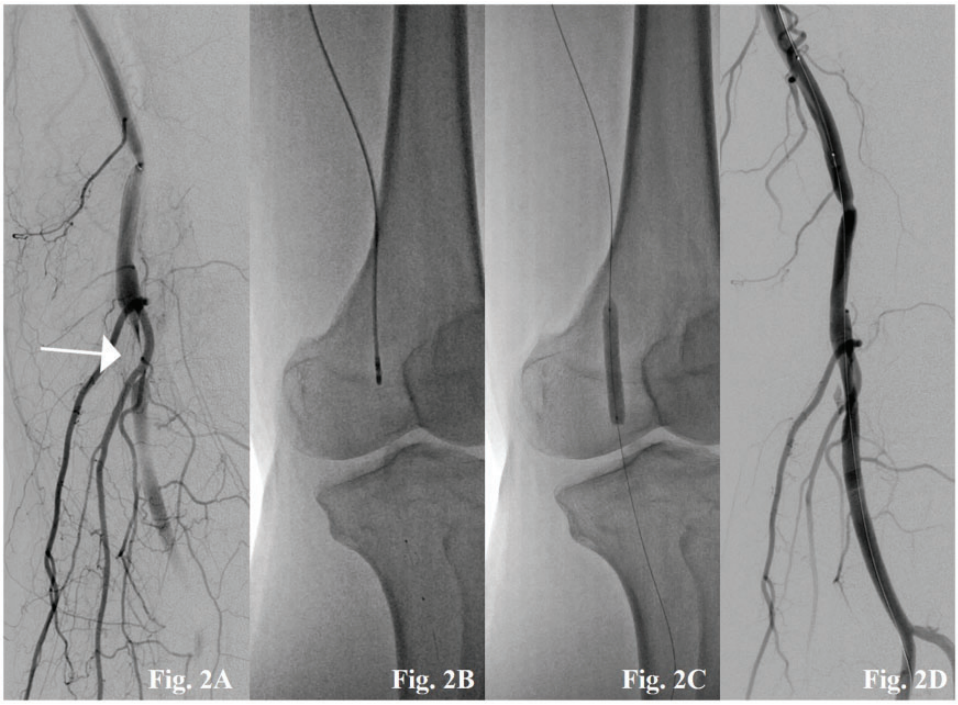

Figure 2. Representative images: (a) left lower-extremity digital subtraction arteriogram showing focal extrinsic narrowing and near-complete occlusion of the popliteal artery (arrow); spot film images at the level of the knee showing (b) rotational atherectomy of the lesion and (c) additional drug-coated balloon angioplasty; and (d) control angiogram showing the recanalized patent popliteal artery lumen after atherectomy and percutaneous transluminal angioplasty.

conservative treatment including oral antiplatelets and intensive follow-up.

During follow-up over 8 months, the degree of claudication worsened with a maximum walking distance of 200 m, and further surgical treatments were performed. Left distal femoral artery to popliteal artery (below-the-knee) bypass surgery was performed by an experienced vascular surgeon (SMK) using an ipsilateral reversed great saphenous vein graft. Improvement in claudication was observed at the 6-month postoperative follow-up (Figure 3), and the patient has continued follow-up without complications or additional treatment for 2 years since the bypass surgery.

Discussion

Cystic adventitial disease remains a rare cause of lower-limb ischaemia, with a prevalence of 0.1% among patients with intermittent claudication.[6] The exact aetiology of CAD is unknown, although four theories regarding its pathogenesis have been postulated: trauma theory, ganglion theory, systemic disorder theory, and developmental theory.[7]

Ultrasonography, computed tomography (CT), magnetic resonance imaging (MRI), and angiography are frequently used to diagnose CAD. Ultrasound imaging shows a thin echogenic line separating the lumen of the vessel and the cyst, representing the vascular intima and media.

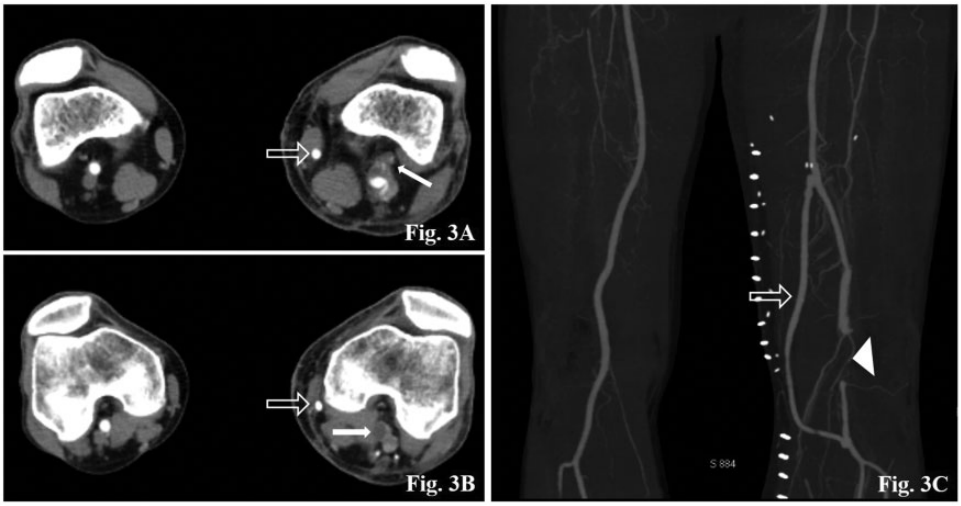

Figure 3. Computed tomography angiography (CTA) images of the lower extremity performed at 6 months after bypass surgery using an ipsilateral reversed great saphenous vein graft. Axial images immediately above the knee (a) and at the knee (b) showing the remaining cystic mass compressing the left popliteal artery (white arrows), and the patent great saphenous vein graft (open arrows). Note the cystic mass, which extends along the lateral superior geniculate artery (white arrow in A); and (c) coronal maximal intensive projection image showing the patent great saphenous vein graft (open arrow) and short-segment non-opacification of the left popliteal artery again (white arrowhead).

Ultrasonic scimitar sign due to the narrowed lumen, lack of vascular flow in the cyst, and posterior acoustic enhancement of the cyst may present on colour Doppler imaging, all of which are considered pathognomonic for CAD on ultrasound.[8] On CT imaging, a low attenuating structure can be seen intimately associated with the artery, without an intervening fat plane.[9] On MRI, the cyst will be observed adjacent to the popliteal artery lumen, with characteristic fluid signals, such as T1 hypointensity, T2 hyperintensity, and the lack of flow signal void or enhancement after the administration of contrast.[10] MRI is considered the most helpful modality for detecting the relationship between cysts and surrounding structures and excluding other pathologies, such as atherosclerotic disease or aneurysm. During angiography, variable appearances, such as hourglass or eccentric narrowing of the lumen (scimitar sign), lack of post-stenotic dilatation, or complete occlusion of the affected vessels can be observed.[8] However, some cases may display nonspecific complete luminal obstruction during angiography, which may be mistaken for an endoluminal lesion.[11]

Several techniques exist for treating CAD, including surgical intervention, percutaneous aspiration, and percutaneous endovascular intervention. Historically, surgical treatment of CAD is preferred over endovascular or minimally invasive techniques, due to considerably lower recurrence rates after surgery.[3] Previously, authors have recommended different techniques, such as complete resection with bypass, excision of the cyst, surgical aspiration of the cyst, or 'exarterectomy', a circumferential resection of the diseased adventitia.[3] Currently, the main surgical treatments are resection of the affected

popliteal arterial segment with interposition of an autologous vein graft, and cyst enucleation. Original articles and textbooks recommend popliteal artery resection with graft interposition only in the presence of arterial occlusion or involvement of the media;[2,12] however, many vascular surgeons prefer this method, even in cases in which the artery is not completely occluded.[13]

Percutaneous endovascular interventions, such as angioplasty and stenting, have been attempted with unsatisfactory results due to a high recurrence rate of 67%.[1] Cyst aspiration and endovascular interventions do not directly address CAD, which originates from the adventitial layer of the artery. As a result, the fundamental cause is not corrected and early recurrence is almost guaranteed (from as early as 8 h after treatment in some cases).[2,13] Only one case of CAD recurrence successfully treated with balloon angioplasty has been reported, in which the patient had previously undergone surgical excision.[14] Nevertheless, in the present case, the decision to perform interventional treatment was based on the patient's history of percutaneous thrombectomy to remove a thrombus from the postoperative popliteal artery 2 months previously, and on the misdiagnosis of thrombotic occlusion based on CTA findings. Angiography and Doppler ultrasound revealed an accompanying recurrence; however, it was difficult to perform surgery immediately and intravascular treatment was performed instead. It was considered that the atherectomy device would not only solve the thrombotic occlusion, but also destroy the cyst wall and consequently allow the drainage of internal fluids. However, the destruction of the cyst wall was insufficient even with atherectomy and balloon angioplasty, and CAD recurred on the day after the procedure.

Recurrence after primary surgical treatment of CAD remains exceedingly rare but may present with similar symptoms. The present patient's preoperative MRI and postoperative CTA showed that cystic masses along the lateral superior geniculate artery (a branch of the popliteal artery) remained following cyst excision with patch angioplasty (the initial surgical procedure). These cystic masses were observed even after bypass graft surgery following atherectomy (Figure 3a). Treatment failures in the published literature may be explained by the residual articular branch becoming the conduit for cyst formation and triggering propagation, thereby leading to cyst reaccumulation.[1] This hypothesis may explain and add confidence to the diagnosis of early CAD recurrence in the current patient.

In the present case, the patient's history of prior treatments, equivocal findings on CTA, and limited data regarding postoperative management of CAD, complicated the diagnosis on initial presentation. However, the patient complained of acute symptoms with evident arterial occlusion, and an urgent intervention was required. Even though the precise cause was uncertain, a decision was made to perform an immediate endovascular procedure to recanalize the popliteal artery. Despite early failure, we consider it reasonable to attempt an endovascular procedure in an emergency. In fact, we acknowledge that MRI or Doppler ultrasound prior to the treatment might have led to a more accurate diagnosis and earlier cure. Nevertheless, we believe that, once CAD is diagnosed, a surgical approach is likely to be the most effective treatment, even if it has already been treated with primary surgical resection.

Conclusion

To the best of the authors' knowledge, this is the first reported case in which atherectomy was performed for recurrent CAD. The present experience showed that endovascular treatment is not satisfactory for

CAD recurrence, even with atherectomy. Although recurrence after primary surgical treatment of CAD remains rare, it may be identified through the observation of similar symptoms following treatment. The application of atherectomy was found to be unsatisfactory in the present case, and the present report highlights the pitfalls which may be avoided by selecting surgical resection as a therapeutic approach.

Acknowledgements

The Authors thank the patient for providing this medical information.

Author contributions

WC analysed and interpreted the patient's data and reviewed the manuscript. JWY and WC were the major contributors to writing the manuscript. JH and SMK revised the manuscript. All authors read and approved the final manuscript.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Funding

The authors disclosed receipt of the following financial support for the research, authorship, and publication of this article: This research was supported by the Chung-Ang University Research Grants in 2020.

ORCID iD


Woosun Choi https://orcid.org/0000-0002-4874-3781

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