

Case Report

Cystic Arterial Disease Located Only in the Media of the Popliteal Artery: A Case Report

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Adventitial cystic disease of the popliteal artery is a rare non-atheromatous peripheral artery disease. In most cases, the cystic lesion is located in the adventitia of the popliteal artery. Herein, we present a rare case of cystic arterial disease in which the cyst was located only in the media of the popliteal artery. We successfully treated the cyst with resection of the affected popliteal artery and reconstruction with an autogenous vein graft.

Keywords: cystic adventitial disease, media, popliteal artery

Introduction

Adventitial cystic disease (ACD) is an uncommon non-atheromatous peripheral vessel disease, in which mucinous cysts develop in the adventitia of arteries and veins. Approximately 80% of cases of ACD involve the popliteal artery, which can cause disturbance of popliteal artery blood flow. The clinical characteristics of ACD of the pop-

liteal artery (ACDPA) include intermittent claudication in patients without atherosclerotic risk factors.¹⁾ Although the term ACD was derived from the observation that almost all mucinous cysts are located in the adventitia of the affected vessels, approximately 0.4% of cases show cysts located in the media.¹⁻³⁾ Hence, the etiology of ACDPA remains controversial. Herein, we present a case of cystic arterial disease in which the cyst was observed only in the media of the popliteal artery.

Case Report

A 65-year-old female presented with a 2 months history of progressive intermittent claudication of the right calf. She had no history of limb trauma but exhibited hypertension, hyperlipidemia, and paroxysmal atrial fibrillation that were well controlled. Her maximum walking distance was approximately 500 meters, but she did not experience rest pain at the time she presented to our hospital. On physical examination, the right posterior tibial or dorsalis pedis arteries were not palpable. The ankle-brachial index on the right leg was 0.70. Her vital signs were stable, with no significant lab-

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Received: May 7, 2019; Accepted: June 24, 2019

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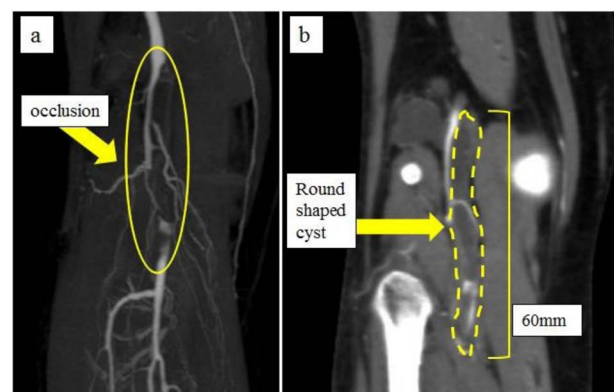


Fig. 1 Enhanced computed tomography (CT) scan showed occlusion of the right popliteal artery and remaining popliteal arterial flow below the knee (yellow circle) (a). The right popliteal artery was occluded by an approximately 60-mm-long circular cyst (yellow broken line) (b). Color figures can be viewed in the online version on J-STAGE and PMC.

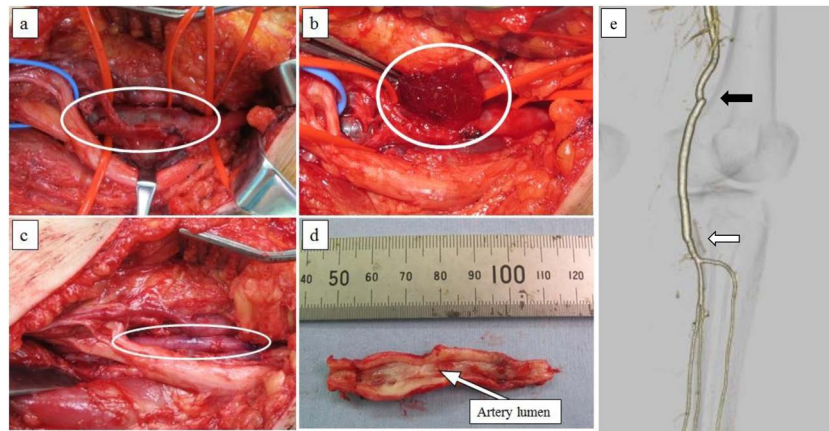


Fig. 2 Operative view showing the fusiform intramural cyst (white circle) of the right middle popliteal artery (a). The cyst was punctured, and red tenacious mucoid material (white circle) emerged (b). The resection being performed with an interposition reversed saphenous vein graft (white circle) (c). The resected lesion showed a thickened artery wall and no signs of arteriosclerosis (d). The patient's head was to the right. Postoperative CT angiography at postoperative day 7 showed sufficient bypass flow (e). Black arrow indicates the proximal anastomosis. White arrow indicates the distal anastomosis.

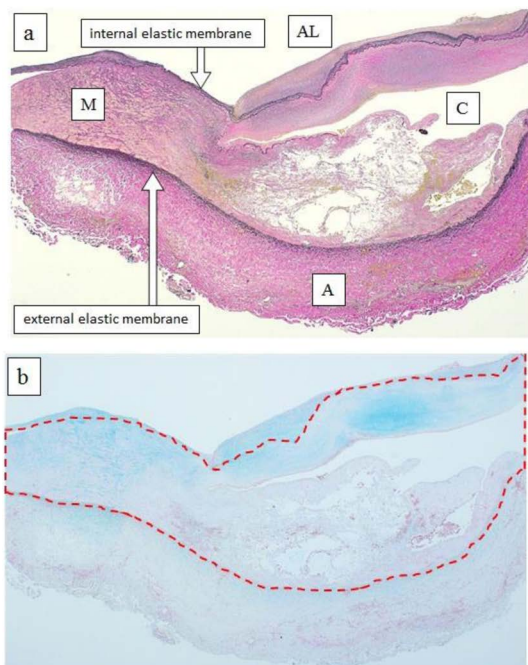


Fig. 3 Pathological analysis showed that the cyst cavity was located only within the media of the popliteal artery (Elastica Van Gieson stain, $\times 20$ magnification) (a). Almost all mucoid materials (blue stains) were recognized in the media of the popliteal artery (inside of the red broken line; Alcian blue stain, $\times 20$ magnification) (b). Color figures can be viewed in the online version on J-STAGE and PMC. AL: arterial lumen; C: cystic cavity; M: media; A: adventitia

oratory findings. Enhanced computed tomography showed an occlusion of the right popliteal artery with a 60-mm-long circular cystic arterial lesion but preservation of arterial

flow below the knee by collateral circulation (Fig. 1). The left popliteal artery and aortoiliac region showed no signs of arteriosclerosis. She was diagnosed as having ACDPA. Because she was symptomatic, with a 500-m intermittent claudication caused by the occlusive lesion, we decided upon surgical treatment. Under general anesthesia, in the prone position and through a posterior approach, a 60-mm-long intramural cyst of the right popliteal artery was exposed. No popliteal arterial pulse was noted below the knee. The cyst was punctured to reveal tenacious mucoid material, although there was no improvement in arterial flow. We then incised the cyst for drainage and decompression (Fig. 2), but the right popliteal arterial flow remained diminished on intraoperative angiography. Finally, we resected the affected popliteal artery with interposition using a right great saphenous vein graft. The operative time was 3h 56min, and blood loss was 170mL. The ankle-brachial index after revascularization improved to 1.10 in the right leg, and the right posterior tibial and dorsalis pedis pulses became palpable. On the 13th postoperative day, she was discharged from our hospital. Histopathological examination revealed that the mucoid materials were located only in the media of the popliteal artery, in contrast to our preoperative expectation. There was no evidence of communication between the cyst and the knee joint capsule (Fig. 3). The patient has had no walking difficulties for 2 years since the operation.

Discussion

ACD is a rare but an important cause of non-atherosclerotic peripheral artery disease. ACD accounts for 0.1% of intermittent claudication.¹⁾ Approximately 80% of ACD

affects the popliteal artery, with eccentric compression by the cyst disturbing arterial flow and causing intermittent claudication. ACD was first reported by Atkins and Key in 1947, with a cystic lesion in the adventitia of the left external iliac artery.⁴⁾ The first case of ACDPA was published in 1954 by Ejrup and Hiertonn.⁵⁾ A number of cases have been reported since, although the precise etiology of ACD remains unclear, and 0.4% of ACD cases show the cystic lesion located in the media of the vessels.¹⁾

Several theories have been proposed for ACD, including repetitive trauma,¹⁾ developmental,⁶⁾ articular/synovial,¹⁾ and systemic disorder origins.⁷⁾ The repetitive trauma theory suggests that repeated flexion and extension of the joint induces chronic damage of the popliteal artery, causing adventitial cystic degeneration. The developmental theory, also known as the cellular inclusion theory, suggests that mesenchymal mucin-secreting cells are derived from the adjacent knee joint to the adventitia of the vessels during development. The articular/synovial theory proposes a connection between the adventitial cyst and the adjacent knee joint capsule. In such cases, a ductus communication between the cyst and the articular capsule can be recognized in diagnostic magnetic resonance imaging and intraoperatively. Finally, the systemic disorder theory postulates that degeneration and cyst formation in the adventitia are caused by a general connective tissue disorder.

Unno et al.³⁾ reported a rare case of ACD in which the cystic lesion was located in the media; the patient often played baseball as a catcher. In that study, a communication between the medial cyst of the popliteal artery and the adjacent knee joint was found intraoperatively, while histopathological examination also identified a communication that comprised an arterial structure and the cyst in the media. The authors suggested that repetitive microtrauma caused an elongation of the cyst originating from the knee joint into the media of the popliteal artery. In the present case, the patient had no history of systemic connective tissue diseases, repetitive trauma, or even daily exercise. In addition, no connection between the medial cyst and the adjacent knee joint was found in preoperative imaging, intraoperative findings, or histopathological examination. Based on these clinical features, we support the development theory and hypothesize that mesenchymal cells may be incorporated into both the adventitia and the media.

Various treatments for ACDPA have been reported, including conservative therapy, percutaneous aspiration, surgical incision and/or excision of the cyst, resection of the affected popliteal artery, and bypass grafting.^{6,8,9)} For articular theory cases, it may be sufficient to perform ligation of the connection along with incision of the cyst and drainage. However, for occlusive cases, because of the risk of recurrence, resection of the affected artery and recon-

struction with an autologous saphenous vein graft should be performed. In the present case, we initially incised the cyst and aspirated the contents (tenacious mucoid materials). However, as blood flow did not improve, we performed resection of the affected popliteal artery and reconstruction with a right great saphenous vein graft.

Conclusion

We report a rare case of ACD with the cyst located in the media of the popliteal artery. The medial location of the cyst may have been the reason why incision of the cyst and aspiration of the content did not improve blood flow. As ACD derived from a medial cyst is rare, it is important to determine the most appropriate treatment.

Acknowledgments

We thank Andrea Baird, MD, from Edanz Group (www.edanzediting.com/ac) for editing a draft of this manuscript.

Disclosure Statement

All authors have no conflicts of interest to declare.

Additional Note

This article was written based on enough informed consent and patient's agreement.

Additional Remarks

This case report was presented at the 46th Annual Meeting of the Japanese Society for Cardiovascular Surgery, Nagoya, Japan, February 15–17, 2016.

Author Contributions

Study conception: SY, KI, KY, TM, TF

Investigation: KY

Writing: SY, KI

Critical review and revision: all authors

Final approval of the article: all authors

Accountability for all aspects of the work: all authors

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