Adventitial Cystic Disease of the Popliteal Artery

Shun-ichi Kawarai, MD,1 Manabu Fukasawa, MD,2 and Yu Kawahara, MD2

We describe a patient with adventitial cystic disease of the popliteal artery with intermittent claudication involving the right calf during exercise. Magnetic resonance imaging (MRI) and computed tomography (CT) revealed a cystic lesion that encircled and compressed the popliteal artery. Resection of the cyst involving a segment of the affected popliteal artery and interposing an autologous vein graft resolved the symptoms, and the postoperative course was uneventful. The cyst was histologically similar to a ganglion.

Keywords: intermittent claudication, autologous vein graft, ganglion

INTRODUCTION

Adventitial cystic disease is a rare vascular condition that mainly affects the popliteal artery and usually presents as intermittent claudication during exercise. We describe the recent management of an otherwise healthy adult man who developed intermittent claudication of the calf while walking.

CASE REPORT

A 45 year-old man was referred to an orthopedic clinic with right calf pain after walking for 10 minutes. Three months of drug therapy did not relieve this symptom, and he was referred to our clinic for evaluation of a vascular disorder. A physical examination revealed palpable right popliteal and foot pulses at rest that persisted during knee flexion. The right and left ankle-brachial pressure indices were 1.32 and 1.22, respectively. Other clinical and laboratory findings were normal, and he was in good health except for mild hypertension and mild hyperlipidemia.

Magnetic resonance imaging (MRI) proceeded in the axial and sagittal planes. T2-weighted imaging revealed a multi-loculated cystic mass with high signal intensity measuring 25 × 30 × 45 mm in the popliteal fossa of the right knee. A cystic mass was adjacent to and encircled the popliteal artery (Fig. 1A). The cyst had a thin wall and no solid contents (Fig. 1B). The signal characteristics of the mass were comparable with those of ganglion on T2-weighted images, and the intensity was homogeneously low on T1-weighted images.2,3)

Contrast-enhanced computed tomography (CT) showed that the popliteal artery was compressed by a non-enhancing structure related to the arterial wall. The typical hourglass sign (concentric, extrinsic compression) of the popliteal artery seen on three-dimensional CT angiograms is shown in Fig. 2.3)

Adventitial cystic disease of popliteal artery was diagnosed and surgery was scheduled. An S-shaped skin incision was cut linear to the skin fold in the right popliteal fossa under general anesthesia with the patient in the prone position throughout the procedure. After dissection, a round, loculated cystic lesion encircling the popliteal artery was identified (Fig. 3). The cyst was soft, elastic and contained a clear jelly-like substance. The dense adhesion ruled out cyst evacuation to preserve the popliteal artery and incomplete resection might have led to recurrence. We completely resected the cyst and the affected segment of the popliteal artery and reconstructed the artery using a great saphenous vein graft.1,4)

Histopathologically, the cyst had a relatively clear margin with vascular adventitia. The cyst had no arterial elements

1Department of Cardiovascular Surgery, Hachinohe City Hospital, Hachinohe, Aomori, Japan
2Department of Cardiovascular Surgery, Yamagata Prefectural Central Hospital, Yamagata, Yamagata, Japan

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Corresponding author: Shun-ichi Kawarai, MD. Department of Cardiovascular Surgery, Hachinohe City Hospital, 1 Bishamontai, Tamukai, Hachinohe, Aomori 031-8555, Japan
Tel: +81-178-72-5111, Fax: +81-178-72-5115
E-mail: shun_z_kawai@hospital.hachinohe.aomori.jp
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in the wall and mucinous content, which were consistent with a ganglion. The postoperative course was uneventful, and the patient was free of right calf claudication. The condition did not recur, and three-dimensional CT angiography, six months later, showed good patency of the vein graft.

DISCUSSION

Adventitial cystic disease is rare, accounting only for 0.1% of all vascular diseases.1) Atkins and Key first described adventitial cystic disease in 1947 that affected the right external iliac artery.5) Over 300 cases have been reported since then. Adventitial cystic disease affects the popliteal artery unilaterally (85%) most often and usually presents as intermittent claudication, although bilateral involvement6) has also been reported and the external iliac, common femoral, radial and ulnar arteries have been affected.4,6)

Over 80% of individuals who develop adventitial cystic disease are men aged between 40 and 50 (range of 11–70) years, and the prevalence of this disease is 1:1200 cases of calf claudication.3) Because the affected population is usually young and at low risk for vascular disease, a clinical diagnosis is often difficult. Adventitial cysts compress the arterial lumen in an eccentric or concentric fashion and appear as scimitar or hourglass signs, respectively, on angiograms.2,3) The severity of ischemic symptoms varies, and mainly depend on the condition of the affected popliteal artery. However, even if the affected popliteal artery appears angiographically normal, adventitial cystic
disease can induce symptoms during exercise. Adventitial cystic disease is considered to cause functional flow obstruction during dynamic exercise. Because the pulse in the right foot of our patient was normal at rest but diminished after exercise when symptoms developed, surgery was required to relieve the symptoms.

The etiology of adventitial cystic disease is still under debate, and trauma, ganglion, systemic disorder, and embryonic development have been proposed. The trauma theory suggests that repetitive trauma causing degeneration of the arterial adventitia ultimately leads to cystic change. The ganglion theory suggests that synovial cysts form nearby joints track along vascular branches and finally reside in the adventitia of major vessels. The systemic disorder theory perceives adventitial cystic disease as a part of generalized connective tissue disorder. The developmental theory suggests that mesenchymal cells rich in mucin from nearby joints incorrectly migrate to the adventitia during embryogenesis. However, no single theory can account for the pathogenesis of all clinical cases. Levien et al. proposed a unifying hypothesis on the basis of embryology of susceptible blood vessels and the affected site. They claim that a joint-related ganglion-like structure is incorporated into a target vessel during embryological development and that this synovial rest or ganglion secretes and enlarges over the years to develop within the adventitial wall later in life, implying that both the ganglion and developmental theories are applicable.

The typical, pathological findings of adventitial cystic disease comprise intramural cysts containing gelatinous material located between the media and the adventitia. The cyst was in contact with the popliteal artery but...
located outside the adventitia in our patient. The cyst had mucinous content but no vascular elements in the wall. These findings were similar to those on ganglion and are inconsistent with most reported cases. However, Ohta et al. described recurrent adventitial cystic disease in an interposed vein graft and found a compressed lumen with intimal hyperplasia and no evidence of intra-adventitial invasion of the vein graft. They postulated that mucin-secreting synovial cells originating from neighboring joint capsule had directly invaded damaged adventitia or had simply attached to and encircled the adventitia.  

The pathological findings of our surgical specimen support this hypothesis.

Gray scale Doppler ultrasonography (U.S.) is a less invasive, convenient diagnostic method for routine clinic application, whereas CT and MRI are more useful for evaluating cyst morphology and possible communication between a cyst and an adjacent joint. The MRI findings of adventitial cystic disease are quite clear as cysts typically appear as regions of homogeneous, low signal intensity on T1-weighted images, and of multi-loculated high signal intensity adjacent to vessels on T2-weighted images. Three-dimensional CT reconstructions using volume rendering or MRA should be performed for optimal preoperative planning. Conventional digital subtraction angiography (DSA) is rarely applied because this modality is invasive.  

The differential diagnosis includes popliteal artery entrapment syndrome (PAES), and multi-planar data acquisition can determine the anatomical extent of arterial intramural cystic lesions, thus helping to discriminate this condition from adventitial cystic disease.  

The management of adventitial cystic disease varies depending on the condition of the affected vessels. Cyst aspiration is less invasive, but it is associated with a high rate of recurrence. Removal of the cyst content (evacuation) while preserving the involved artery can be effective and resection of the affected segment of the vessel and interposing a vein graft or prosthesis is promising. Long-term follow-up is mandatory because adventitial cystic disease can recur or grafts can become occluded. Endovascular interventional therapy has yielded unsatisfactory outcomes.  

Adventitial cystic disease of the popliteal artery is an uncommon but important cause of peripheral vascular insufficiency in younger individuals without specific atherosclerotic risk factors. Thus, adventitial cystic disease should be considered when relatively young individuals present with intermittent claudication.  

**CONCLUSIONS**

We described a patient with adventitial cystic disease of the popliteal artery with intermittent claudication. The cyst involving the affected popliteal artery was resected, and an autologous vein graft was interposed. The patient remained free of symptoms after the procedure, and postoperative three-dimensional CT images showed good patency of the vein graft. Long-term follow-up is mandatory because of the potential for recurrence or graft occlusion.

**REFERENCES**