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Case Report

Adventitial cystic disease of the popliteal artery with intimal tear

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ABSTRACT

We report the case of a 68-year-old man with right intermittent claudication by adventitial cystic disease. We performed resection of the cyst and affected popliteal artery with interposing an autologous vein graft. Intraoperative findings revealed an intimal tear between the cyst and the compressed artery. His symptoms resolved after surgery, and the postoperative course was uneventful.
Although adventitial cystic disease with intimal tear is rare, we consider that conventional surgical intervention remains the favorable treatment option for adventitial cystic disease.
<**Learning objective:** We present a rare case of adventitial cystic disease (ACD) with intimal tear successfully treated with surgical repair. In the present case, intimal tear could not be detected preoperatively. Although this case is rare, we should pay attention to intimal tear in treating ACD. In case of ACD with intimal tear, resection of the affected artery and reconstruction with interposing graft are feasible.>

Introduction

Adventitial cystic disease (ACD) is a vascular condition that mainly affects the popliteal artery, presenting as a rare cause of non-atherosclerotic claudication. Etiologies of ACD are still controversial and some hypotheses have been reported. In general, it is said that this type of cyst grows from adventitia. We report a rare case of ACD with intimal tear in popliteal artery.

Case report

A 68-year-old man with hypertension and diabetes mellitus was referred to our hospital for evaluation of a vascular disorder. He developed intermittent claudication of the right leg, paresthesia, and limb edema during the previous 3 months. The claudication distance was about 100 m. Normal arterial pulsation was present bilaterally in the femoral, popliteal, and dorsalis pedis artery, which persisted during knee flexion. The right and left ankle-brachial pressure index were 0.64 and 1.16, respectively. Other clinical and laboratory findings were unremarkable. Doppler ultrasound revealed a 20-mm longitudinal hypoechoic cyst behind the right popliteal artery and decreased blood flow at distal lesion below the knee. Peak systolic velocity ratio of the lesion was 60 cm/s. Contrast-enhanced computed tomography (CT) showed that right popliteal artery was compressed by a non-enhancing structure related to the arterial wall. No peripheral embolism was seen. The typical hourglass sign of the popliteal artery was seen on three-dimensional CT angiograms (Fig. 1A). Magnetic resonance imaging revealed a uniloculated cystic mass measuring $23 \times 19 \times 25$ mm in the popliteal fossa of the right knee. A cystic mass was adjacent to the popliteal artery and exhibited low-signal intensity on T1-weighted images and high-signal intensity on T2-weighted images (Fig. 1B). ACD of popliteal artery was diagnosed, and surgery was scheduled.

Surgery was performed through the posterior approach using an S-shaped incision in the prone position. A round cystic lesion was identified behind the popliteal artery. Intraoperative findings showed a communication between compressed arterial lumen and cyst, containing a clear jelly-like substance through the intimal tear (Fig. 2). We resected the affected popliteal artery and interposed an autogenous great saphenous vein graft. In histopathological findings, the cyst containing mucin determined ACD (Fig. 3). The postoperative course was uneventful, and the patient was free from claudication and paresthesia. The patient was discharged on the sixth postoperative day. Postoperative CT revealed a patent graft and no residual cyst. A follow-up is underway.

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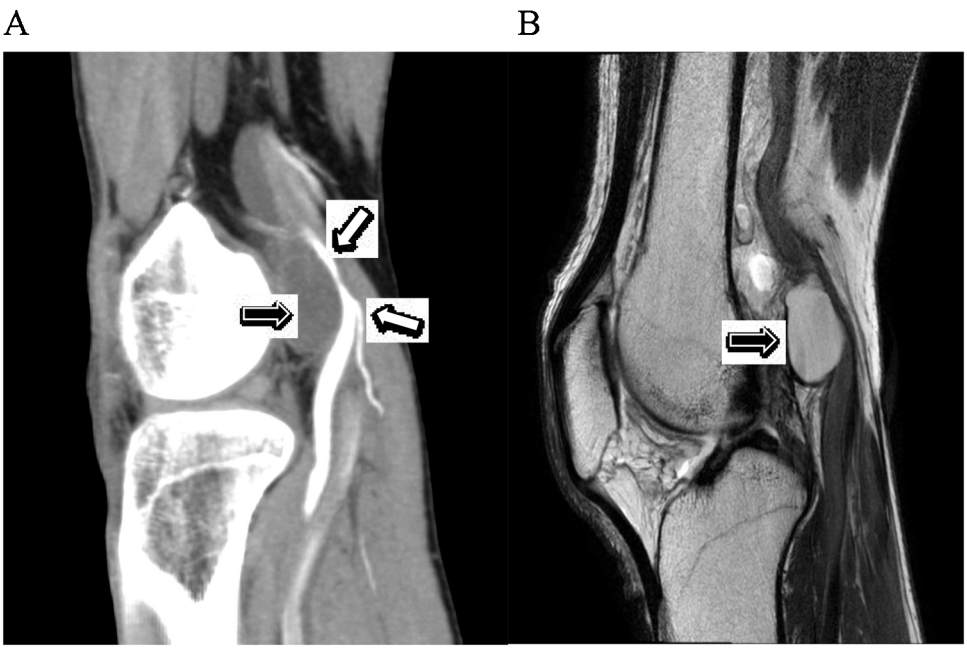

Fig. 1. Typical hourglass sign of popliteal artery (white arrow) and cystic mass (black arrow). (A) Preoperative computed tomography of right popliteal artery. (B) Preoperative magnetic resonance imaging (T2-weighted image).

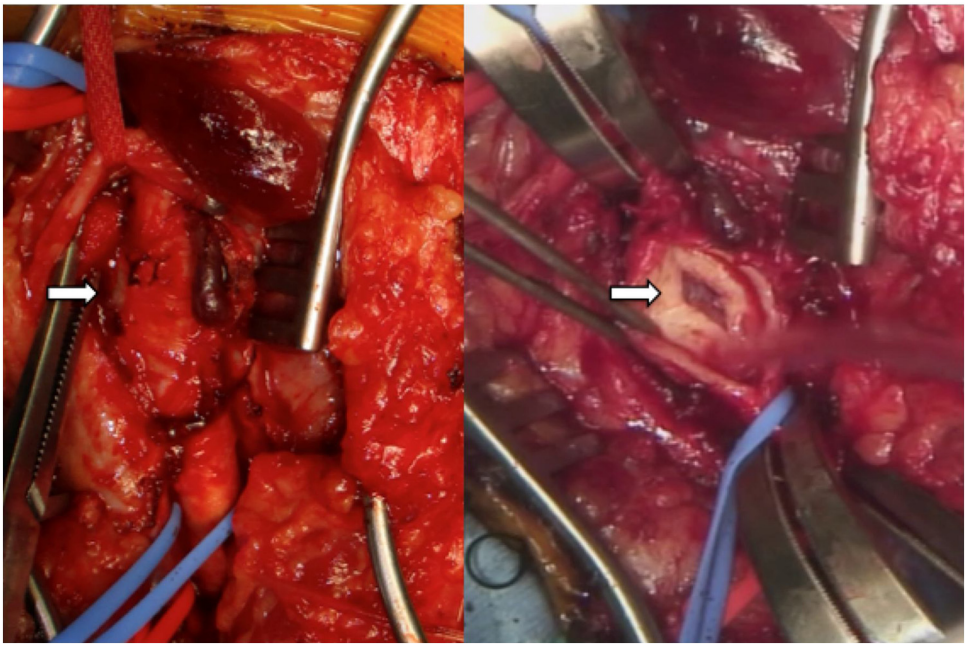

Fig. 2. (A) Jelly-like content under adventitia (white arrow). (B) Intraoperative finding revealed intimal tear (white arrow) of affected artery.

Discussion

ACD mostly affected in the popliteal artery is a rare vascular disease characterized by a mucin-containing cyst in the adventitial layer of the artery [1].

In histopathology, there are different types of cysts with viscous material consisted of proteoglycans, mucopolysaccharides, mucoproteins, and hyaluronic acid [2].

Various hypotheses regarding pathogenesis of ACD have been reported. ACD is caused by repetitive trauma, systemic disorder, migration of synovial ganglia (ganglion theory), and implantation of mesenchymal cells into the adventitia (developmental theory). Ganglion theory and developmental theory are considered as the most reasonable for explaining ACD by many reports. In the present case, there was no communication with a synovial space supporting ganglion theory, and no trauma history supporting trauma theory.

As far as searched, a report of ACD with intimal tear was not seen. We performed arteriotomy carefully not to injure posterior wall. The intimal tear is not an artificial result. We confirmed the

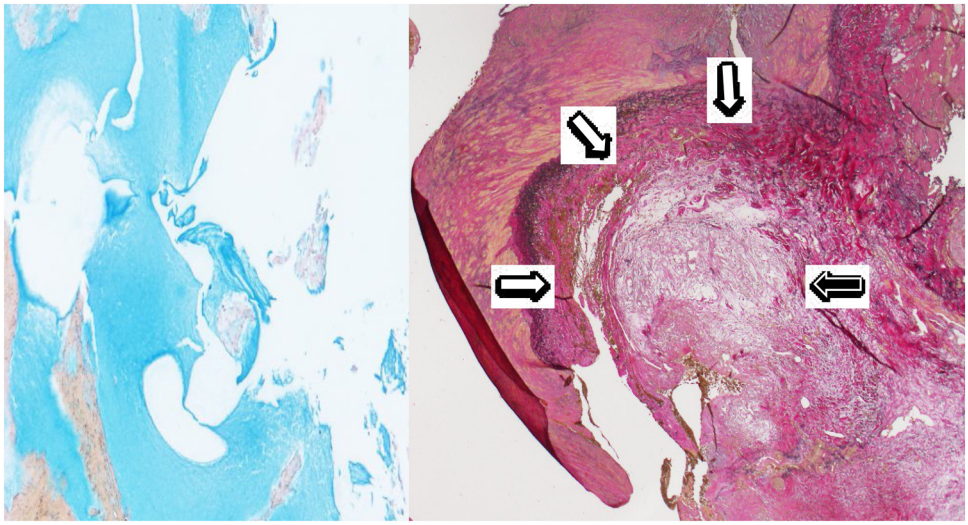

Fig. 3. (A) Cyst contains mucoid structure stained with alcian blue. (B) Photomicrograph stained with elastica van Gieson shows cystic lesion (white arrow) hyperplastic adventitia (black arrow).

intimal tear was secondary caused by cyst compressing the arterial lumen, because the content of the cyst was not hematoma but mucin pathologically. The cyst was within thickened adventitia (Fig. 3). These findings are consistent with ACD. We could not detect intimal tear retrospectively by contrast CT. This is because the cyst was full of mucin and free from hematoma. The intima that is not affected was normal.

Treatment options are excision of the cyst preserving the artery, excision of the affected artery with interposition grafting, percutaneous drainage under CT or ultrasound guide, and conservative management.

Case reports contain spontaneously resolved ACD, mainly when the arterial wall cysts have some communication with the knee joint [3], however, the natural history is typically progression toward further occlusion. This patient had symptoms caused by compression of nerve and vein such as paresthesia and limb edema, cyst removal was necessary.

Although ultrasound- or CT-guided aspiration has also been used to treat ACD, it is not always possible because of the high viscosity of the content and multilocular cyst. Although successful aspiration can improve symptoms, the recurrence after aspiration is high. Recurrence in the short term has also been reported [4].

If we had performed aspiration, this patient could have experienced complications such as bleeding or pseudo aneurysm formation because of intimal tear identified in operation.

Surgical excision of the cyst preserving the medial and intimal layers is suitable for patients without severe popliteal stenosis and minimal adherence between the cyst wall and the artery.

If we had tried to preserve the medial and intimal layers in the present case, bleeding could have occurred. We could not even detect an intimal tear by any image findings retrospectively. In cases of severe arterial stenosis or intimal tear like this case, resection of the affected artery and reconstruction with interposing graft are feasible.

It has been reported that approximately 10% of patients had recurrence of the cyst [5]. Therefore, a long-term follow up is necessary.

Conclusion

We presented a rare case of ACD with intimal tear successfully treated with surgical repair. We performed excision of the affected artery with interposition grafting. In the present case, intimal tear could not be detected preoperatively. Although this case is rare, we should pay attention to intimal tear in treating ACD.

Disclosure statement

All authors have no conflict of interest.

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