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☐ Case Report ☐

Adventitial Cystic Disease of the Common Femoral Artery: A Case Report and Literature Review

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Arterial adventitial cystic disease is an uncommon type of non-atherosclerotic peripheral vessel disease. Most cases of arterial adventitial cystic disease occur in the popliteal arteries; however, fewer cases have been reported in the femoral arteries. A 59-year-old male patient visited the hospital with a complaint of a swelling on the lower extremity that had begun two months earlier. Suspecting deep vein thrombosis based on a physical examination and ultrasonography from another hospital, tests were performed. Magnetic resonance imaging (MRI) was performed for exact diagnosis because venous adventitial cystic disease was suspected by computed tomography venography. The MRI indicated venous adventitial cystic disease as well. Thus, a cystic mass excision was performed. In the end, a cystic mass compressing the common femoral vein that originated from the common femoral artery was diagnosed based on the macroscopic findings. This case is reported because blood circulation in the vein was impeded due to arterial adventitial cystic disease, and the symptoms improved after the cystic mass excision and polytetrafluoroethylene roofing angioplasty.

Key words: 1. Adventitial cystic disease

2. Peripheral vascular disease

CASE REPORT

A 59-year-old man presented with a swelling of the left lower extremity that had begun two months earlier. He had diabetes mellitus and no history of trauma. No other specific sign was found on physical examination. At a local clinic, he received a diagnosis of deep vein thrombosis. For further evaluation, we performed computed tomography (CT) venography and pelvic magnetic resonance imaging (MRI). The CT venography axial images showed a 2.3 cm cystic mass compressing the left common femoral vein (Fig. 1A). The MRI

images showed a cystic mass compressing the left common femoral vein as well (Fig. 1B). We suspected adventitial cystic disease (ACD) of the common femoral vein, and the patient underwent an operation. Under general anesthesia in a supine position, the femoral artery and vein were dissected for about 5 cm with a longitudinal incision on the left inguinal area. Surgical exploration showed an approximately 2.0 cm sized cystic mass that originated from the adventitia of the common femoral artery and was compressing the common femoral vein. The cyst that had adhered to the femoral artery was removed first, and the part of the wall that was at-

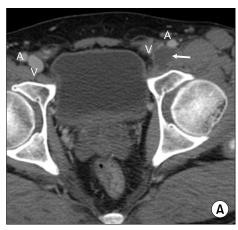
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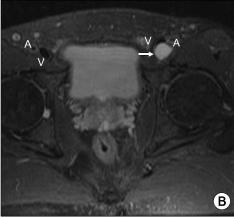
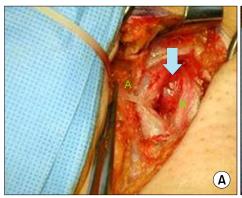


Fig. 1. (A) A mass was suspected to have originated from the femoral vein, and the computed tomography findings show that the mass obstructs the circulation of the femoral vein (arrow: mass). (B) The magnetic resonance imaging findings also show that the cystic mass in the femoral vein caused the partial obstruction of the vein (arrow: mass). A, femoral artery; V, femoral vein.



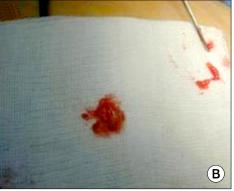


Fig. 2. (A) The mass that originated from the femoral artery (arrow) compressed the femoral vein (arrow: adventitial cyst). (B) Macroscopic findings of an adventitial cystic mass (about 2.3 cm). A, femoral artery; N, femoral nerve.

tached to the cyst in the femoral artery was partially resected. Angioplasty was performed to repair the femoral artery whose wall was partially resected. Because the patient had had a partial circulation problem in the vein before the surgery and thus venous obstruction was possible, a polytetrafluoroethylene (PTFE) patch was used for angioplasty instead of using an autologous vein graft. A cystic mass excision with PTFE roofing angioplasty was performed and a gelatinous material exuded from the mass (Fig. 2). Microscopically, the presence of a mucinous cyst and vessel wall degeneration confirmed cystic adventitial disease of the common femoral artery (Fig. 3). The final diagnosis was ACD of the common femoral artery.

DISCUSSION

ACD is an uncommon type of non-atherosclerotic peripheral vessel disease. ACD is an unusual cystic tumor of the

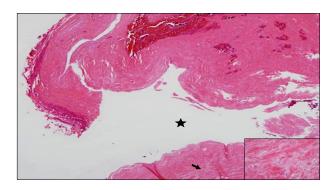


Fig. 3. The photomicrograph shows the cystic space (asterisk) in the arterial wall that was filled with gelatinous material. A portion of the arterial wall (arrow) shows dissected collagen bundles via mucoid degeneration (H&E, ×40).

blood vessels characterized by the accumulation of a mucinous substance in the adventitia [1]. ACD is an uncommon disease that was first described in 1947 by Atkins and Key [2]. ACD accounts for only 0.1% of cases of vascular disease, and among them, 85% of all cases of ACD occur in the popliteal arteries, while fewer cases appear in the femoral arteries [3]. The first case of common femoral artery ACD was reported by Jaquet and Meyer-Burgdorff [4] in 1960 [5]. It was reported that the prevalence is 5 times higher in males than in females. The age of occurrence ranges from 11 to 72 years, and the average age is 42 [1].

The pathogenesis of ACD is unknown. Four theories have been proposed about the nature of ACD: 1) the theory that it is a systemic disorder of the connective tissue; 2) the theory that it is a chronic degenerative change due to repetitive trauma; 3) the developmental theory, which maintains that a joint-related ganglion-like structure is incorporated into the vessels during embryologic development; and 4) the ganglion theory that arterial adventitial cysts originate from joint capsular synovial structures [2,3,6]. So far, obvious causes have not yet been identified. This disease is likely to have complex causes [3,6]. It should be noted that the case reported here occurred in a fisherman. Thus continuous trauma from using fishing tools in contact with the thighs was suspected to have contributed to the lesion. On the other hand, considering that the lesion occurred unilaterally and did not appear on the opposite thigh, repetitive trauma is not an obvious explanation for the lesion.

The symptoms of ACD can include unilateral claudication of the lower extremity, and in rare cases it shows ischemic neuropathy such as paresthesia, pain, and rhigosis. Other possible symptoms include arterial obstruction that results in the pulse in the femoral, the popliteal, and the dorsal pedis arteries being weak or not palpable [3,5,6]. However, the main symptom in our case was the uncommon occurrence of swelling that appeared due to the venous obstruction. In addition, the pulse of the blood vessels of the lower extremities was fully palpable.

An ACD diagnosis can be confirmed by imaging. The imaging modalities are angiography, ultrasonography, CT scan, and MRI. Recently, it has been accepted that angiography using 3-dimensional CT alone is now considered sufficient for diagnosis. CT angiography is considered to be an important test because it not only determines the site and extent of stenosis but is also useful in evaluating the entire cir-

culation system [3,5,6].

There are several methods of treatment for ACD. Aspiration of the cyst under CT or ultrasonography is a minimally invasive method, but it is difficult to perform and the cyst has a high recurrence rate [3]. Another method is surgical cyst excision without opening the artery. This method also has a higher recurrence rate than complete resection involving the vessel with artificial material interposition. Interposition provides better long-term patency than the other methods [3,6].

In this case, in the pre-surgical assessment the lesion was misdiagnosed as originating from the femoral vein instead of from the femoral artery. However, it was obvious that CT angiography was useful in the diagnostic process. In addition, recurrence will be minimized by our having performed the arterial repair and interposition as well as the cystic mass excision. Thus we report here a case of ACD, a rare disease, with unusual symptoms, that was successfully treated with surgery.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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