Vascular Specialist International
Vol. 32, No. 3, September 2016
pISSN 2288-7970 • eISSN 2288-7989

Case Report

Cystic Disease of the Groin Presenting as Compression of a Femoral Vessel

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In this study, we describe our diagnosis and treatment of two patients who presented with femoral vessel compression caused by a cystic lesion in the groin. One case was diagnosed as adventitial cystic disease (ACD) of the common femoral artery resulting in leg claudication and the other was diagnosed as a ganglion cyst (GC) causing femoral vein compression and unilateral leg swelling. The operative findings differed between these two cases with respect to the dissection of the cyst and femoral vessel, but the postoperative histological examination results were similar. The pathogenesis of ACD and GC is not fully understood, and further investigation is needed to delineate the exact pathology of these uncommon conditions.

Key Words: Adventitia, Cystic disease, Ganglion cyst, Hip, Femoral

Received August 4, 2016
Revised August 26, 2016
Accepted September 1, 2016

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Conflict of interest: None.

Vasc Spec Int 2016;32(3):124-128 • http://dx.doi.org/10.5758/vsi.2016.32.3.124

INTRODUCTION

Cystic lesions may occur around any of the joints or tendon sheaths in the body. Common locations of cystic lesions include the wrist, hand, foot, ankle, and knee, which are body surface areas where it is easy to identify cysts [1]. Cystic disease of the hip joint is a rare clinical entity and most cases have been observed incidentally without clinical symptoms [1]. However, in some cases, a cystic lesion arising from the hip joint may cause pain and symptoms related to compression of the surrounding nerves, veins, and arteries [2-4]. We describe here two patients who presented with femoral vessel compression caused by a cystic lesion of the groin. One case was diagnosed as adventitial cystic disease (ACD) of the common femoral artery causing leg claudication, and the other was diagnosed as a ganglion cyst (GC) causing femoral vein compression and unilateral leg swelling.

CASE

1) Case 1

An 18-year-old male "Wushu" martial arts athlete presented to our outpatient clinic with a palpable right inguinal mass and claudication for 6 months. His past medical history was unremarkable. On physical examination, a nontender, firm mass measuring 2 cm was palpable in the right inguinal area and the right ankle pulse was not palpable. The ankle-brachial index (ABI) was measured as 0.57 on the right side and 1.13 on the left side. Duplex ultrasonography revealed a cystic lesion that was compressing the common femoral artery (Fig. 1A). Computed tomography (CT) revealed a cystic lesion located superomedially to the right common femoral artery causing severe stenosis (Fig. 1B). Magnetic resonance imaging also showed the cystic lesion and its communication with the right hip joint (Fig. 1C).

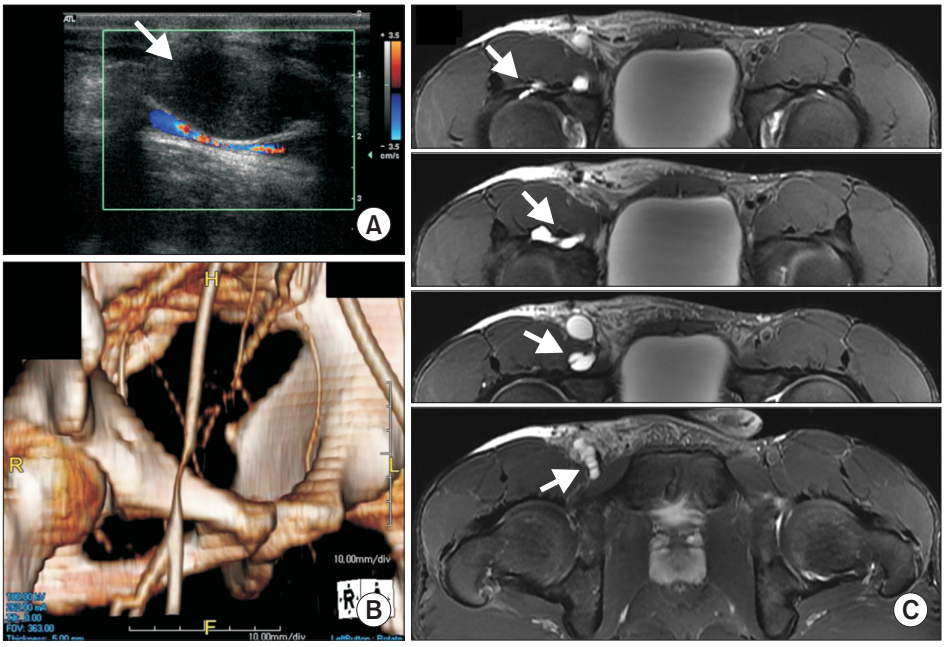

Fig. 1. Preoperative images of adventitial cystic disease in Case 1. (A) Doppler ultrasound image demonstrates severe stenosis of the common femoral artery lumen compressed by the cystic lesion (arrow). (B) Multidetector row computed tomography with 3-dimensional volume-rendering image shows a scimitar sign without features of atherosclerosis. (C) Axial T2-weighted magnetic resonance images show a cystic mass arising from the right hip joint (arrows).

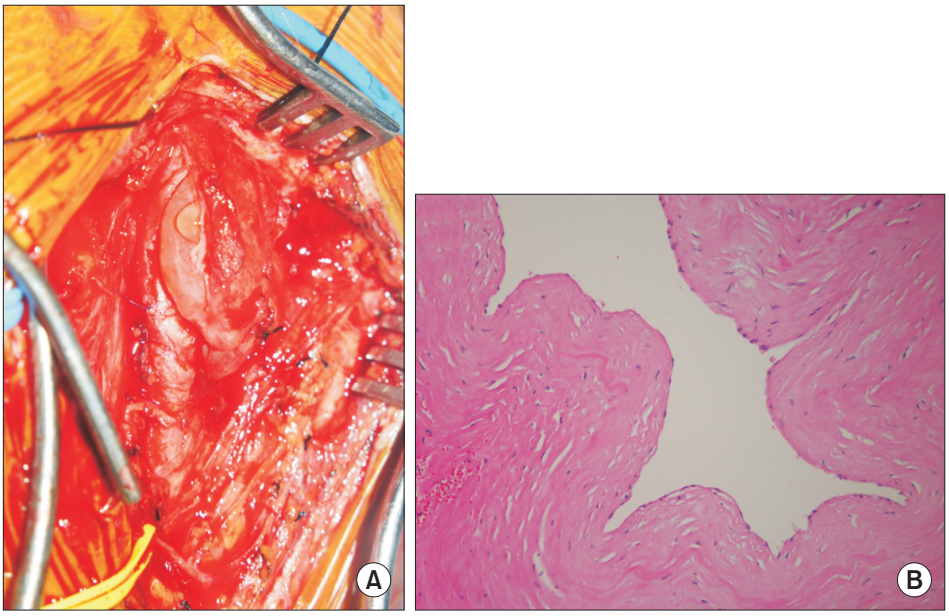

Fig. 2. Intraoperative and histological images. (A) Intraoperative image shows amber-colored gelatinous material extracted from the cyst. (B) Microscopic examination of the surgical specimen demonstrates the cystic space surrounded by dense collagenous fibrous tissue without synovial lining (H&E stain, ×200).

Preoperatively, a GC of the hip joint was suspected and an elective operation was performed. A longitudinal incision was made to the right groin, and the common femoral artery and surrounding tissue were dissected carefully. However, dissection of the arterial wall to the lesion was impossible, and we suspected ACD of the femoral artery at this point. We performed a stab incision of the cyst, and gelatinous material was extracted from the cyst (Fig. 2A). The entire length of the cyst (4 cm) was inspected through a longitudinal incision, the cyst was excised, and the connection to the hip joint was ligated. After excision of the cyst, we observed the medial layer of the femoral artery without any overlying adventitia, and we decided to insert an interposition graft with a prosthetic graft because of the friability of the remaining arterial wall and a small diameter of the saphenous vein. Histological examination of the surgical specimen demonstrated that the cystic space was surrounded by dense collagenous fibrous tissue without synovial cell lining (Fig. 2B). Postoperatively, the patient recovered without complications and was discharged 6 days after the operation. His ABI returned to normal (0.99 on the right side and 1.12 on the left side) and the claudication disappeared.

http://dx.doi.org/10.5758/vsi.2016.32.3.124

2) Case 2

A 34-year-old man presented to our hospital because of right leg swelling for 20 days. He had been hit by a blunt object 20 days before and had since developed right leg swelling. The ultrasonography performed at another local medical center demonstrated a cystic lesion in the right inguinal area, and he was referred to our hospital for further evaluation. His past medical history was insignificant. Moderate swelling of the right leg was noted on physical examination. A CT scan revealed extrinsic compression of the femoral vein caused by a cystic mass measuring 3.3×2.5 cm located posteriorly to the femoral vein (Fig. 3) and a connection of the cyst to the hip joint (Fig. 3).

A longitudinal femoral incision was made over the femoral vein and the dissection was continued to the femoral vein. During the operation, the femoral vein was freely dissected from the cyst and resection or angioplasty of the femoral vein was not necessary (Fig. 4A). After traction of the dissected femoral vein to the medial side, the cystic mass and its connection to the hip joint were resected and ligated. Microscopic examination of the surgical specimen demonstrated that the cystic space was filled with mucinous material surrounded by dense collagenous fibrous tissue without synovial cell lining, consistent with the diagnosis of a GC (Fig. 4B). Postoperatively, the patient recovered without complications and was discharged 4 days after the operation. The leg swelling disappeared during the 3-month follow-up.

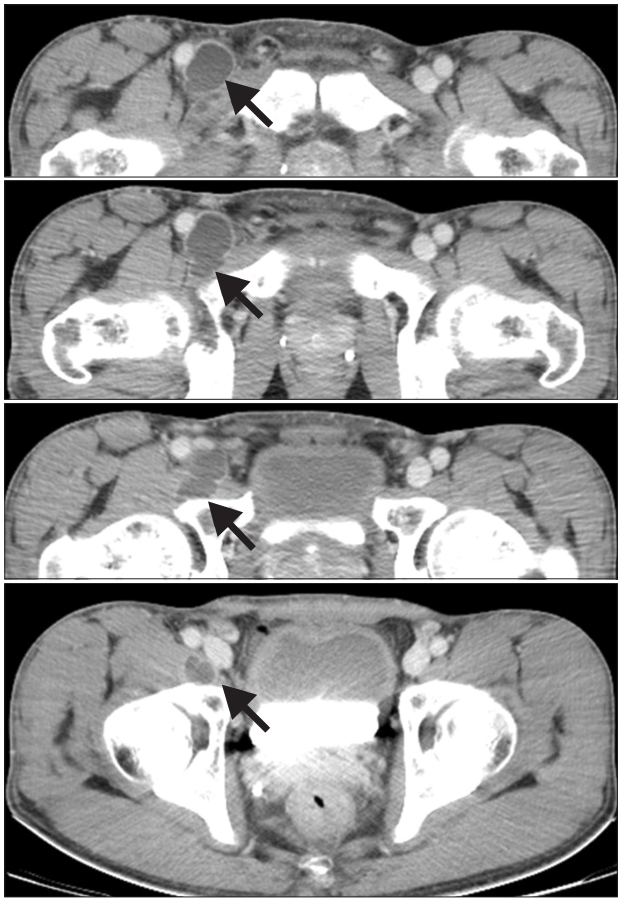

Fig. 3. Preoperative axial venous phase computed tomography image of a ganglion cyst in Case 2. The image shows the compressed right common femoral vein caused by the cystic mass and its connection with the right hip joint (arrows).

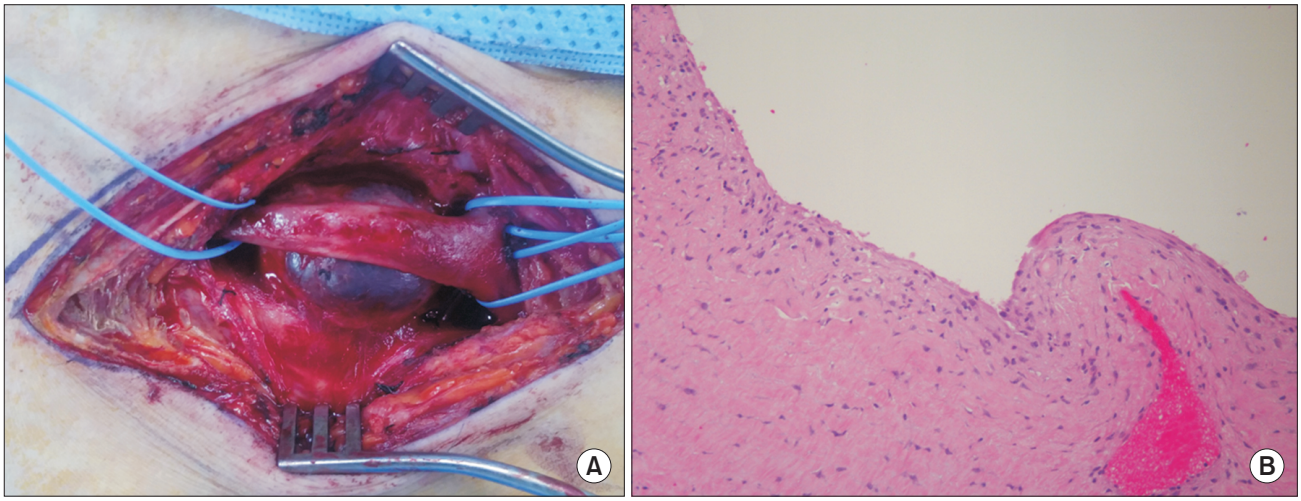

Fig. 4. Intraoperative and histological images. (A) Intraoperative image shows the cystic mass beneath the freely dissected common femoral vein. (B) Microscopic examination of the surgical specimen demonstrates the cystic space surrounded by dense collagenous fibrous tissue without synovial lining (H&E stain, ×200).

DISCUSSION

Both of our patients had a cystic lesion in the inguinal area, and imaging studies showed a connection between the cyst and the hip joint. In addition, both patients exhibited compression of a femoral vessel—to the femoral artery in one patient and to the femoral vein in the other—that caused the symptoms, which is a rare clinical presentation. However, the operative findings differed between these two cases. In the first case, the dissection of the cystic mass from the femoral artery was impossible and we could see the medial layer of the femoral artery after removal of the cystic mass, as is usually seen in ACD. In the second case, the femoral vein could be dissected freely from the attached cystic mass without injury to the femoral vein. We diagnosed the first case as an ACD and the second case as a GC based on these operative findings.

Currently, more than 700 ACDs are available in the literature with the majority of cases affecting the popliteal artery with claudication symptoms in young men without vascular risk factors [5]. Among arterial ACD, femoral arterial ACD is second largest and approximately 40 cases have been reported to date [5]. The exact etiology of ACD remains uncertain, but various hypotheses have been proposed in the literature. Levien and Benn [6] discussed four theories with respect to the etiology of ACD: (1) degenerative theory: a mucinous or myxomatous systemic degenerative condition associated with a generalized disorder; (2) repeated trauma theory: repeated trauma can cause destruction and cystic degeneration of the adventitia of the adjacent vessel; (3) ganglion theory: adventitial cysts arise as capsular structures that then enlarge and track along vascular branches to involve the adventitia of the adjacent major vessel; and (4) developmental theory: mucin-secreting cells derived from the mesenchyme of the adjacent joint are incorrectly placed in the vessel wall during development of the disease. However, the hypotheses supported by various authors are the ganglion and developmental theories [6,7]. The connection between ACD and adjacent joint as in our case has been used to support the two most popular theories concerning the pathogenesis of ACD. The proponents of the "developmental" theory consider this connection as a residual of embryogenesis. In contrast, the proponents of the "ganglion" theory see this anatomic connection as a result of herniation of synovium through a breach in an adjacent articulation [8]. In one recent literature review of 746 ACDs, all cysts were para-articular, suggestive of an association between ACD formation and its respective neighboring joint, supporting the developmental and/or ganglion theories [5]. A joint connection was identified in 122 cases (16.4%) of 746 ACDs on imaging, at surgery, or both [5]. Currently, the etiology of the disease remains controversial. However, it has been proposed recently that ACD, in which a cyst occurs within the popliteal artery near the knee, may occur by an articular mechanism and may have a conduit leading from the joint as described previously, which is similar to the development of intraneural GCs, which spread within the epineurium of tibial or peroneal nerves [7]. Therefore, the recognition of the connections and proper management of identified connections will lead to better and more durable outcomes in the treatment of these patients in the future.

Although there is confusion and no consensus on the proper terminology in the literature, two types of cysts have been described as causing cystic mass formation associated with hip joint histologically [9]. In cases involving femoral vein compression, synovial cysts are more common and present as a juxta-articular fluid-filled collection usually accompanied by a hip joint disorder such as rheumatoid arthritis, osteoarthritis, or trauma; these usually have a lining of synovial cells seen on histological analysis [10]. By contrast, GCs are extremely rare and are defined as a cystic, tumor-like lesion that is delineated by dense connective tissue and filled with gelatinous fluid rich in hyaluronic acid and other mucopolysaccharides. Most GCs display a focal cellular lining of the cavity and these cells are immunohistochemically different from synoviocytes [2,10]. A fluid-filled stalk may or may not be seen connecting the two types of cyst to the adjacent joint [9]. Our second patient, who experienced compression of the femoral vein, did not have a history of hip joint disease and was relatively young. Also, no synovial cell lining was found on postoperative histological examination. Therefore, we diagnosed this case as a GC. Previously, over 40 cases of vein compression due to a cystic lesion around the hip have been described in the literature [1]. This case serves as a reminder that when presented with unilateral leg swelling in which the common causes have been excluded, the rarer causes of limb swelling including synovial or GC should not be forgotten.

In summary, we have described two rare cases of cystic disease of the groin that presented as femoral vessel compression. The operative findings differed between these two cases in respect to the dissection of the cyst and femoral vessel, although the postoperative histological findings were similar. These cases highlight a rare clinical presentation of cystic disease and show the importance of a high index of suspicion when investigating and treating young patients with lower limb symptoms. The pathogenesis of ACD and GC is not fully understood, and further investigation is necessary to delineate the exact pathology of these uncommon conditions.

http://dx.doi.org/10.5758/vsi.2016.32.3.124

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