Long-term outcome of partial resection in venous adventitial cystic disease

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ABSTRACT

Venous adventitial cystic disease is extremely rare. Therefore, standard treatment methods have not been clearly defined. Some reports suggest that complete cyst removal is an effective treatment. However, considering the relatively high recurrence rate, follow-up periods were short. Herein, we report the case of a 75-year-old man with venous adventitial cystic disease successfully treated with partial cyst wall excision. No recurrence was observed for 10 years postoperatively. This case suggests that complete cyst wall excision might not be necessary for the treatment of venous adventitial cystic disease. (J Vasc Surg Cases Innov Tech 2021;7:382-5.)

Keywords: Femoral vein; Venous adventitial cystic disease; Recurrence

Vascular adventitial cystic disease (ACD) is a rare cystic condition in which the vascular adventitia is filled with mucin-containing gelatinous fluid, more often in arteries.[1] Compared with arterial ACD, venous ACD is even rarer, with an incidence of only 5.3% to 9.3% of all cases of ACD.[2,3] Venous ACD occurs predominantly in the femoral (62%) and iliac veins (13%) and presents with intermittent unilateral leg swelling.[2,4,5] Several hypotheses for the etiology have been proposed but are controversial.[3,6,7] As reports are scarce, most reports suggest surgical removal as the treatment.[2,4-6]

Surgical resection is of two types, complete cyst wall removal, including the intimal wall of the vein,[8,9] and partial cyst wall removal, preserving the vein intima.[5,10] For both techniques, few reports have long-term follow-up,[10] and some reported postoperative recurrences,[9,11] so the optimal method has not been determined.[2,4]

Herein, we report a case of venous ACD of the common femoral vein (CFV) in which the cyst contents were surgically removed and the cyst wall was partially resected. The patient has survived without recurrence for 10 years. Patient consent was obtained for publication of the case details and images.

Although similar techniques have been reported, this is the first report of a case with long-term postoperative follow-up.

Our case suggests that it may not be particularly necessary to perform complete resection for the treatment of venous ACD.

CASE REPORT

The patient was a 75-year-old man, weighing 48.7 kg and 147 cm tall, with histories of diabetes mellitus and hypertension but no history of trauma. He visited our hospital because of intermittent swelling of the lower extremity for 3 months with a nonpulsatile mass in the right inguinal region. Blood tests revealed a D-dimer level of 0.7 ng/mL and HbA1c level of 6.4%. Blood counts and other biochemical test results were normal.

Preoperative imaging is shown in Fig 1. Duplex ultrasound (DUS) imaging revealed a cystic mass on the posterior wall of the right CFV with a smooth surface and a homogeneous internal hypoechoic appearance with acoustic shadowing. The cystic mass compressed the femoral vein and severely narrowed the lumen of the vein. Deep vein thrombosis and abnormalities were not observed in the femoral and popliteal veins; furthermore, there were no significant abnormalities associated with the femoral and popliteal arteries. Computed tomography revealed a multilocular low-density mass behind the right CFV. The cystic mass was 4 cm in diameter and had no contrast-enhancing effect. The mass crushed the vein and caused the vein lumen to collapse markedly. Magnetic resonance imaging revealed a cystic mass with low and high signals on the T1-and T2-weighted images, respectively. On the computed tomography and magnetic resonance imaging scans, the cyst had no connections with surrounding structures such as the femoral artery, nerve, or hip joint. On the basis of these findings, we made a diagnosis of venous ACD of the femoral vein and performed surgical treatment.

At surgery, we made an incision in the inguinal region, controlled the CFV, and then dissected the cyst. No heparin

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Author conflict of interest: none.

For this study, informed consent from the patient and approval of the research ethics committee of Iwate Prefectural Isawa Hospital (approval No. 2020-17) were obtained.

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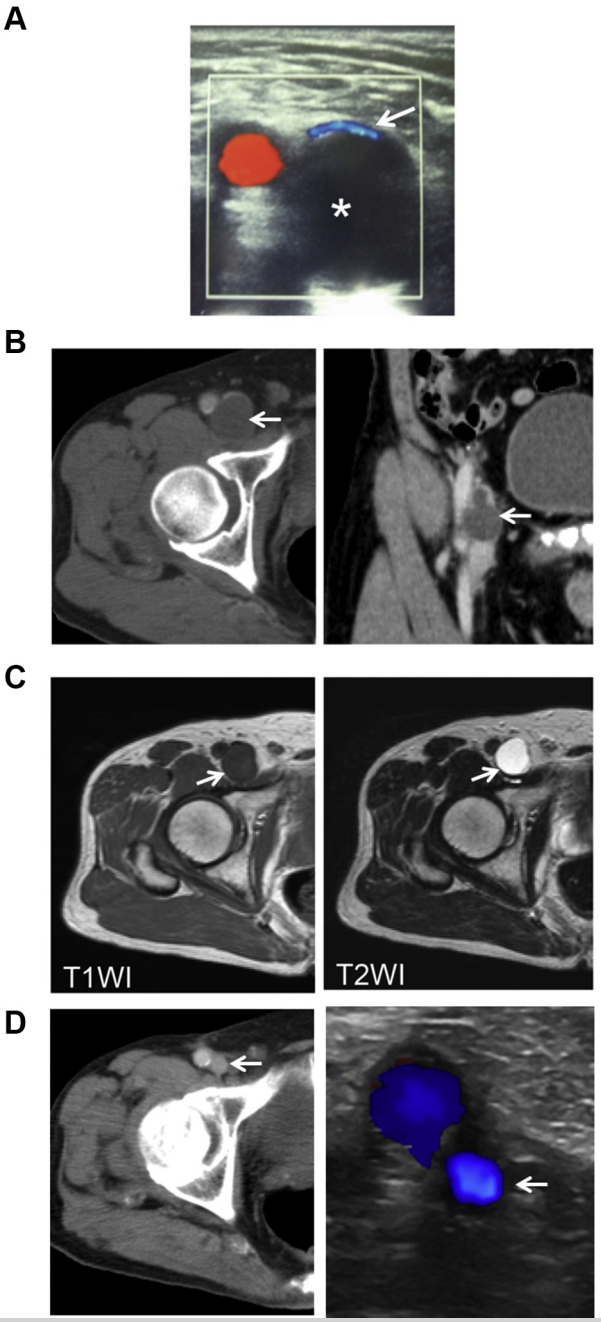

Fig 1. Preoperative imaging. **A,** duplex ultrasound (DUS) images of the right thigh show a hypoechoic mass (*asterisk*) with an acoustic shadow behind the femoral vein. The femoral vein (*arrow*) is seen to be compressed by the cystic mass. **B,** Contrast-enhanced computed tomography (CT) images show a multilocular mass with low density (*arrow*). No contrast enhancement is observed inside the mass. **C,** Magnetic resonance imaging images reveal a cystic mass with low intensity (T1WI) and high intensity (T2WI) (*arrow*). **D,** CT images (*left*) at 8-year follow-up and DUS image (*right*) at 10-year follow-up show no recurrent cyst, and the femoral vein is patent (*arrow*).

was given, and the vein was not clamped during the cyst excision. The cyst had no adhesion to the femoral artery or other surrounding tissues, except the femoral vein. No structure was

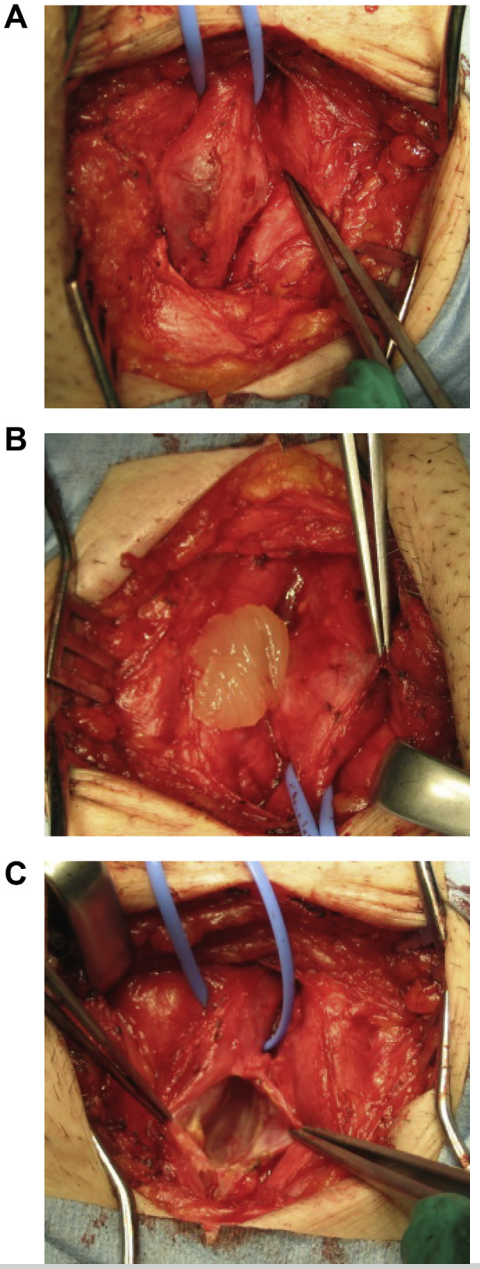

Fig 2. Surgical findings. **A,** The cyst is adherent to the femoral vein. **B,** Incision of the cyst revealed a clear, *yellow*, jelly-like mucoid content. **C,** The inner surface of the cyst.

found connecting to the joints. We could separate the cyst from the femoral artery easily, but the cyst was tightly adherent to the femoral vein, so we could not detach it from the vein. We made an incision in the cyst and removed all the lucid, yellow, jelly-like mucoid contents. Then, we resected the cyst wall as much as possible without injuring the vein lumen (Fig 2). We confirmed that the venous blood flow was preserved on intraoperative DUS, so we left a portion of the cyst wall, without resecting the vein circumferentially. Histologically, the cyst wall was composed of collagen tissue and many histiocytes but no lining cells such as epithelial or synovial cells (Fig 3).

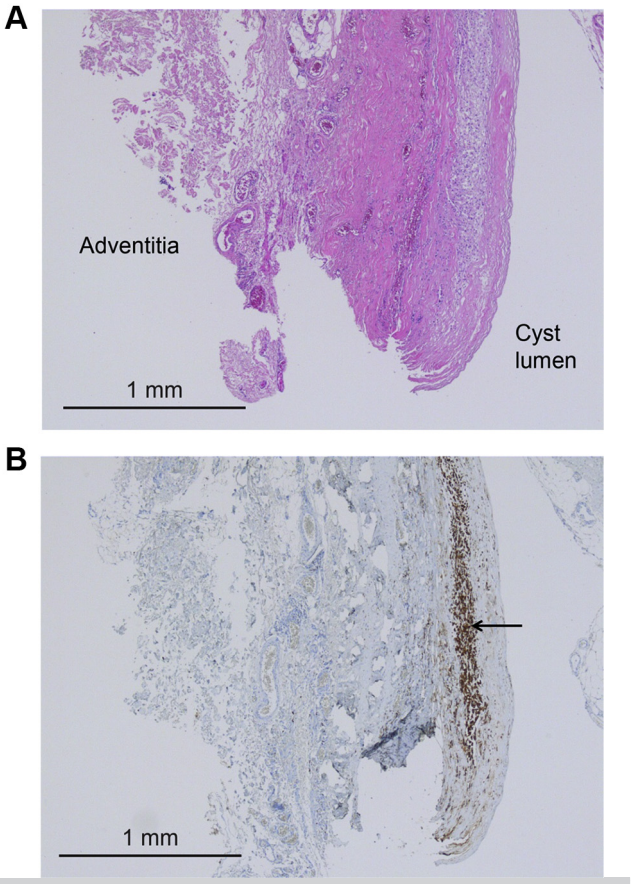

Fig 3. Histological findings. **A,** Hematoxylin-eosin staining revealed that the cyst wall was composed of collagen tissue. **B,** CD68 immunostaining showed the presence of many histiocytes in the cyst wall (*arrow*) and no lining cells such as epithelial cells and synovial cells.

The patient left the hospital, recovering from the thigh swelling. We did not use any anticoagulants postoperatively. The patient was followed up with DUS at 3 and 6 months, and every year for 10 years. We found no recurrence of the thigh swelling or cystic or aneurysmal changes in the vein (Fig 1, *D*).

DISCUSSION

The diagnosis of this case is appropriate because the symptoms, imaging, and pathological findings are consistent with previous reports.[2,4,5,12] The etiology of this case is unclear, as the patient had no history of trauma or systemic disease and no synovial tract was identified.[3,6,7]

The optimal treatment of venous ACD remains controversial because of its rarity. Minimally invasive treatments such as needle aspiration, ethanol injection, and endovascular therapy have been reported in a few cases, but with frequent recurrences.[13,14]

Surgical resection is the treatment of choice in most cases,[2,4-6] and the methods can be divided into complete and partial resections of the cyst wall.

Some reports suggest complete cysts resection to prevent recurrence,[11] whereas several cases of recurrence have been reported even in cases with complete resection.[8,9] Therefore, whether complete resection is effective in preventing recurrence is unclear. Moreover, complete cyst wall resection may have disadvantages such as the risk of thrombus at the venous repair site and problems with long-term patency.

In this study, we performed a partial cyst wall resection, leaving the cyst wall on the venous side, and the clinical symptoms were relieved. Furthermore, no recurrence was observed in the long term. Histologically, the inner wall of the ACD is covered with fibrous connective tissue and is not lined by a cell layer of epithelial or synovial cells.[12] Also there are no mucus-producing cells on the inner surface of the cyst. This suggests that leaving the cyst wall may not affect recurrence.

In our case, the partial cyst wall resection preserved the intact venous intima, eliminating the need for venous repair and the risk of thrombosis. Furthermore, the preserved cyst wall was sturdy and did not result in a venous aneurysm in the long term. However, if the preserved vein wall was fragile or if there was a residual venous stenosis, complete removal of the cyst and venous reconstruction may be necessary. If the integrity of the remaining vein wall is maintained, it can be reconstructed by patching; if most of the vein wall is missing, graft interposition may be necessary.

Notably, recent reviews support the joint (synovial) etiology theory.[2,5,15] In our case, a joint connection could not be identified; however, we may have unintentionally ligated the joint connection. If a synovial tract is identified, it should be ligated to prevent recurrence.[2,6,16]

Finally, few reported cases have long-term follow-up. Postoperative follow-up was either not performed or was limited to a short period.[8-11]

We are the first to report a case of venous ACD with no recurrence for 10 years after partial cyst wall resection. However, this is only one case report, and we believe that more cases must be reviewed in the future to develop optimal treatment strategies to prevent recurrence.

CONCLUSIONS

We used a surgical approach for partial cyst wall resection for ACD of the femoral vein. The patient has survived for 10 years without recurrence. Hence, our report suggests that complete cyst wall resection with venous reconstruction may not be necessary for venous ACD.

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