

Adventitial cystic disease of the common femoral vein: an unusual cause of lower extremity swelling and review of the literature

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ABSTRACT

We report the case of a 61-year-old man who had presented with acute unilateral limb swelling. Computed tomography venography and duplex ultrasound demonstrated compression of the right common femoral vein by a common femoral vein adventitial cyst. Before intervention, the patient had developed an acute deep vein thrombosis of the right common femoral vein and great saphenous vein. Preoperative magnetic resonance imaging demonstrated concern for synovial connection. After 6 months of anticoagulation therapy, the patient underwent adventitial cyst excision with ligation of the hip joint articular connection. At 4 months postoperatively, the patient was symptom free without cyst recurrence. The findings from the present case support the synovial theory for adventitial cystic disease. (*J Vasc Surg Cases Innov Tech* 2021;7:610-6.)

Keywords: Adventitial cystic disease; Femoral vein; Synovial cyst; Venous thrombosis

Adventitial cystic disease (ACD) is a rare etiology of peripheral arterial occlusive disease and venous disease. ACD is characterized by the formation of mucinous cysts with the adventitia of arteries and veins, which can lead to various signs or symptoms depending on the location and degree of luminal obstruction. Most often, it affects the popliteal artery, leading to intermittent claudication.¹ Several theories exist regarding the origin of adventitial cysts; however, our center previously identified a synovial connection in 17% of the reported cases.¹⁻³ The present case is unique owing to involvement of the femoral vein and identification of a joint communication in support of the synovial origin theory. We have provided a review of the available literature on venous involvement in ACD. The patient provided written informed consent for the report of his case.

CASE REPORT

A 61-year-old man with no pertinent medical history had presented to an outside institution with a 9-day history of right lower extremity edema after prolonged air travel. Duplex

ultrasound and computed tomography venography demonstrated compression of the right common femoral vein by an adventitial cyst, without evidence of venous thrombosis.

After car travel en route to obtain a second opinion at our institution, he had noted worsening edema, now involving the thigh, and was subsequently evaluated in the emergency department. A repeat ultrasound scan confirmed the presence of the cystic mass and demonstrated an acute deep vein thrombosis of the right common femoral vein and great saphenous vein. He had no personal or familial history of thrombotic events.

On examination, the patient had palpable pedal pulses and right lower extremity edema from the ankle to the thigh, without varicosities or skin changes. No palpable lymphadenopathy or masses were present in the groin. Anticoagulation was initiated, and thigh-high graduated compression stockings rated at 30 to 40 mm Hg were placed. A repeat ultrasound scan at 6 months demonstrated recanalization of the common femoral and saphenous veins with chronic post-thrombotic changes. Magnetic resonance venography demonstrated a 3-cm cyst anteromedial to the right common femoral vein that was compressing the lumen and another 1.2-cm cyst posterior to the vein and communicating to the hip joint (Figs 1 and 2). A labral tear was identified on the magnetic resonance venogram (Fig 1, C), which possibly contributed to formation of the adventitial cyst.

A shared decision was made for operative intervention owing to the risk of recurrent thrombosis. The patient underwent open right common femoral vein cyst excision (Fig 3, A-C) and ligation of the connection to the hip joint. Intraoperative ultrasound confirmed the normal caliber of the common femoral vein without compression at completion (Fig 3, D). Histopathologic examination of the specimen demonstrated features consistent with cystic adventitial disease. The patient had an uneventful postoperative course and was discharged to home on postoperative day 1. He continued taking rivaroxaban for 1 month, followed by a further 6 months of aspirin monotherapy.

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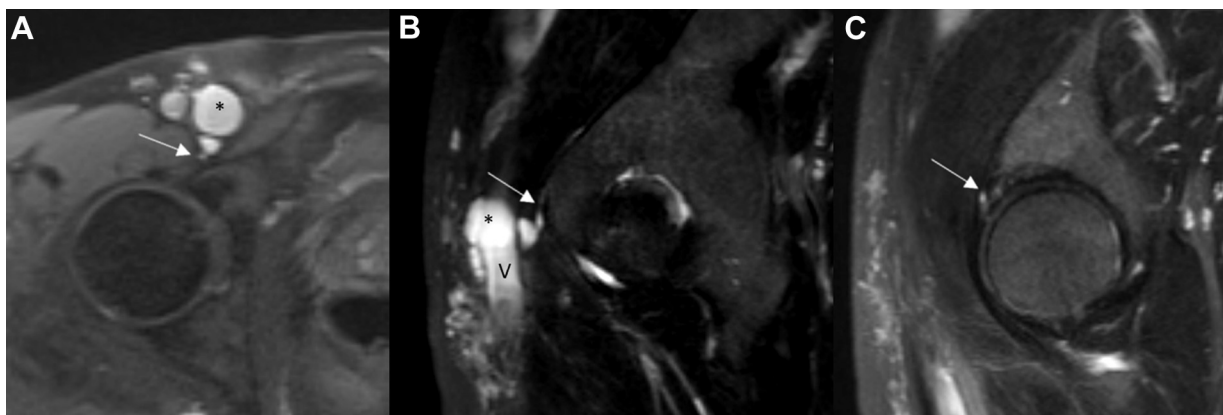


Fig 1. T2-weighted magnetic resonance venogram images. **A**, Axial cut demonstrating the joint connection (arrow) to the cyst (asterisk). **B**, Sagittal cut demonstrating the joint connection (arrow), the main cyst (asterisk), and caudal common femoral vein (V). **C**, The origin of the labral tear (arrow).

At the 4-month follow-up visit, his symptoms had completely resolved, and duplex ultrasound demonstrated no evidence of cyst recurrence.

DISCUSSION

A 61-year-old man with a venous adventitial cyst causing compression and thrombosis of the right common femoral vein was successfully treated with cyst excision and ligation of the connection with the hip joint. Venous reconstruction was not undertaken, because complete expansion of the common femoral vein had occurred after cyst removal and the joint connection was identified and ligated.

The articular origin theory of ACD has been supported by previous reports identifying a connection between a synovial joint and an adjacent adventitial cyst.^{2,4} This joint connection arises from a feeding vessel, which serves as a conduit for synovial fluid. As demonstrated in the present patient, the joint connection can be visualized with preoperative imaging in select cases.⁵ We investigated the literature to determine the incidence of joint connections in patients with ACD of the venous system.

A review of the literature identified 64 studies reporting 72 cases of ACD of the venous system (Appendix). The common femoral vein (65%) was the most commonly affected location, followed by the external iliac (18%) and popliteal (7%) veins (Table I). Additionally, ACD involving the brachiocephalic vein, basilic vein, an autogenous brachiocephalic fistula, posterior tibial vein, and small and great saphenous veins was identified.^{4,6-11} The mean age at presentation was 47.8 years (range, 5-75 years), 39 of the patients were men (54%), and the disease process was localized to the left side in 42 patients (58%). Medical comorbidities were infrequently reported, with five patients (7%) having a history of venous thromboembolism (Table I). Symptoms that indicated the need for intervention included limb swelling in most patients (85%), a palpable mass in

28%, limb pain in 15%, varices in 7%, and paresthesia in 6%. Three patients were asymptomatic, two with the finding of a painless mass and one with the cyst incidentally identified by imaging studies. ACD was initially misdiagnosed as deep vein thrombosis in 13 patients (18%) and treated with anticoagulation. To assist in the diagnosis, ultrasound (68%) and computed tomography (63%) were the most commonly used imaging modalities. A connection between the cyst and an adjacent joint space was identified in 12 patients (17%), and the connection was ligated intraoperatively in 5 patients. No recurrences were reported after ligation of the connection to the joint capsule.

The initial interventions for treatment of the adventitial cyst are detailed in Table II, with 16 patients undergoing reintervention for recurrence. Recurrence was observed in all 10 patients for whom cyst aspiration with or without sclerosant was the primary treatment, with surgical excision or resection the final successful treatment in 9 patients. In one patient treated with aspiration, recurrent aspiration and injection of sclerosant was successful through 18 months of follow-up.¹² Simple cyst drainage was the primary treatment in two patients. Both patients developed recurrence and were treated with cyst excision. Cyst excision with or without patch venoplasty was the primary treatment in 47 patients. Four patients had developed recurrence after excision and underwent repeat excision (two patients), interposition graft (one patient), or aspiration with sclerosant (one patient). Interposition grafting with a prosthetic graft or autologous vein conduit was performed in 10 patients without recurrence. A stent placed in the external iliac vein to treat one patient was complicated by in-stent thrombosis.¹³ The mean follow-up after intervention was 11 months (range, 1-48 months). Surveillance imaging was obtained for 40 patients (56%), a limitation of the reported rates of recurrence in our review. Recurrence was identified within 6 months of the primary intervention in all cases.

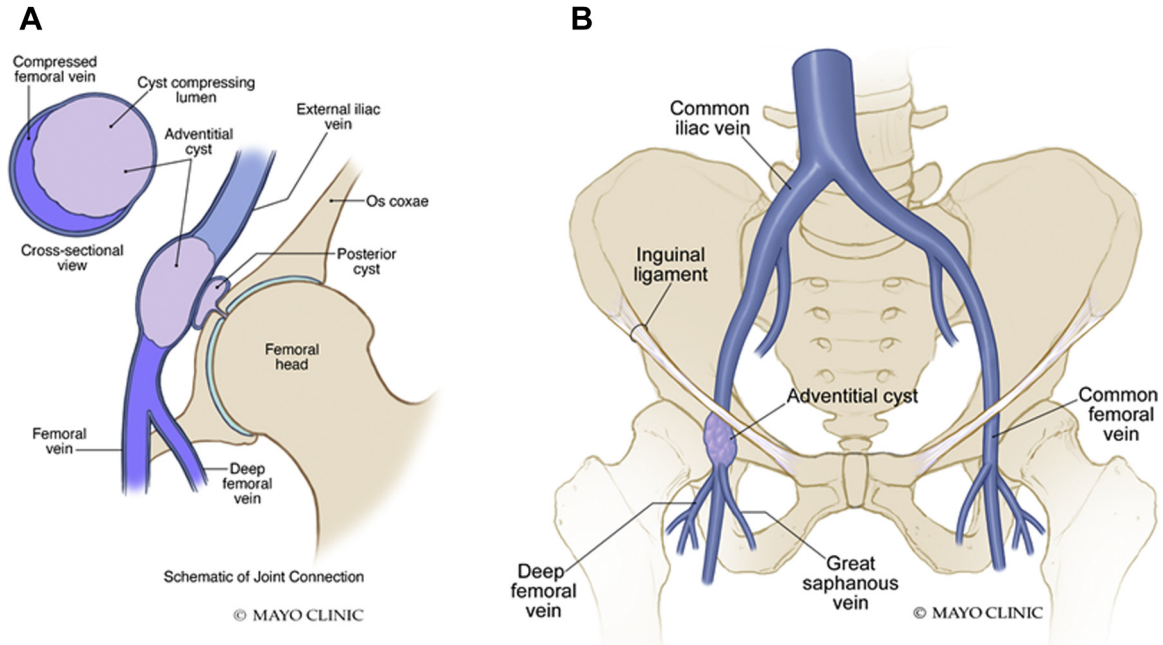


Fig 2. **A**, Illustration demonstrating compression of the vein lumen by the cyst with a connection to the hip joint capsule. **B**, Schematic diagram demonstrating the location of the adventitial cyst caudal to the inguinal ligament but cephalad to the saphenofemoral junction.

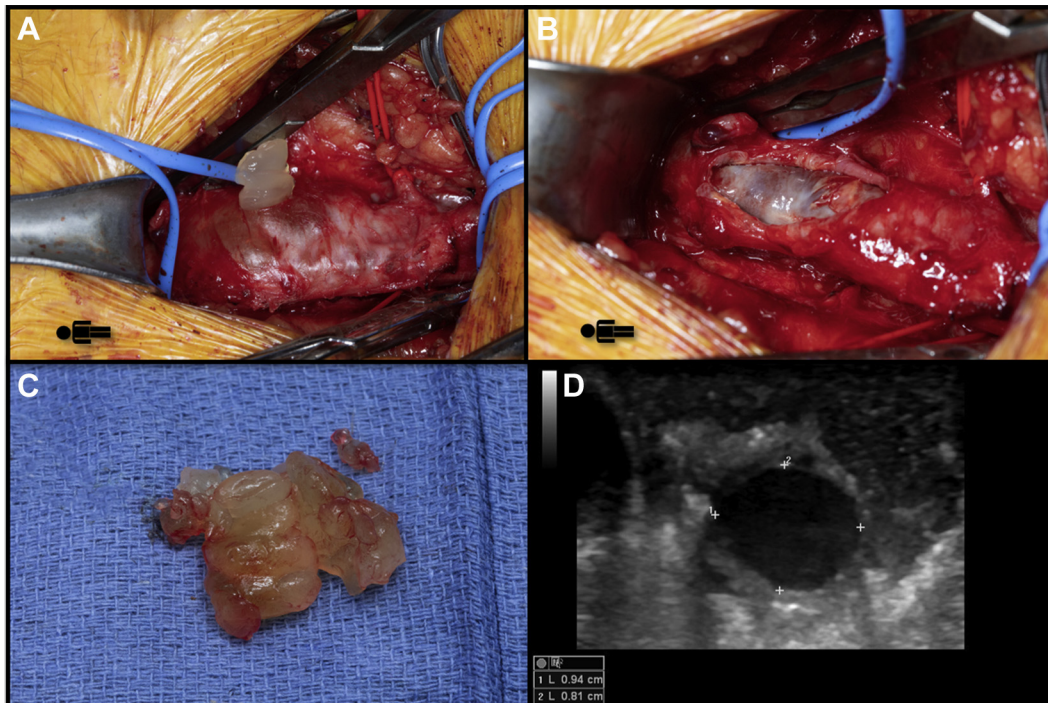


Fig 3. Intraoperative photographs of the adventitial cyst before **(A)** and after **(B)** cyst excision. **C**, Photograph of cyst contents. **D**, Select transverse view of the common femoral vein on intraoperative ultrasound demonstrating full expansion after cyst excision.

CONCLUSIONS

We have presented an unusual case of ACD of the common femoral vein with associated thrombosis and

identification of a connection with the hip joint on preoperative imaging. The patient was successfully treated with cyst excision and ligation of the joint communication.

Table I. Patient demographics and presentation

Variable	Mean ± SD (range) or No. (%)
Age, years	47 ± 14 (5-75)
Male sex	39 (54)
Left sided	42 (58)
Location	
Common femoral vein	47 (65)
External iliac vein	13 (18)
Popliteal vein	5 (7)
Short saphenous vein	2 (3)
Basilic vein	1 (1)
Brachiocephalic vein	1 (1)
Brachiocephalic AVF	1 (1)
Posterior tibial vein	1 (1)
Great saphenous vein	1 (1)
Comorbidity	
History of VTE	5 (7)
Tobacco use	3 (4)
Hypertension	5 (7)
Hyperlipidemia	1 (1)
Diabetes mellitus	1 (1)
AAA	2 (3)
Coronary artery disease	1 (1)
Presenting symptoms and signs	
Swelling	61 (85)
Pain	11 (15)
Palpable mass	20 (28)
Varices	5 (7)
Paresthesia	4 (6)
Asymptomatic	3 (4)
Claudication	2 (3)
Bruit	1 (1)
Joint stiffness	1 (1)
Weakness	1 (1)
Abdominal pain/nausea	1 (1)

AAA, Abdominal aortic aneurysm; AVF, arteriovenous fistula; VTE, venous thromboembolism.

Similar to previous reports, a connection to a joint space was identified in 17% of patients with ACD of the veins.¹⁻³ Surgical treatment with cyst excision or venous resection had a low rate of recurrence of 7%. Previous reviews of ACD have reported similar recurrence rates with surgical treatment.^{1,14-23} Simple cyst drainage or cyst aspiration resulted in unacceptably high rates of cyst recurrence when reported. Repeat imaging after intervention is warranted to monitor for cyst recurrence.

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Table II. Summary of initial treatment and associated recurrence rate with each intervention

Initial treatment	Patients, No. (%)	Recurrence, No. (%)
CE, all cases	47 (65)	4 (9)
CE without reconstruction	41 (57)	2 (5)
CE with patch venoplasty	5 (7)	2 (40)
Laparoscopic CE	1 (1)	0 (0)
CA, all cases	10 (14)	10 (100)
CA without sclerosant	9 (13)	9 (100)
CA with injection of sclerosant	1 (1)	1 (100)
VR, all cases	12 (17)	0 (0)
VR with prosthetic interposition graft	7 (10)	0 (0)
VR with vein interposition graft	3 (6)	0 (0)
VR with simple ligation	2 (3)	0 (0)
Cyst drainage	2 (3)	2 (100)
Venous stent	1 (1)	0 (0) ^a

CA, Cyst aspiration; CE, cyst excision; VR, venous resection.
^aThe adventitial cyst was never excised or aspirated and was complicated by stent thrombosis.

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APPENDIX. CASE REPORTS INCLUDED IN LITERATURE REVIEW

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