










Successful surgical treatment of popliteal artery cystic adventitial disease

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Abstract

We present the case report of a 43-year-old male, professional sport user, complaining of pain in his right lower extremity. We have discussed the symptoms, diagnostics with its differentiation, treatment methods and their efficacy. The diagnosis of cystic adventitial disease was made on the basis of diagnostic imaging such as Doppler ultrasound and magnetic resonance imaging. Surgical procedure is the treatment of choice, to which the patient was qualified.

Keywords: cystic adventitial disease; popliteal artery; vascular diseases

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Introduction

Cystic Adventitial Disease (CAD) is a rare vascular disease, causing blood vessel lumen obstruction, mainly of the peripheral arteries. The vessel most frequently affected by this disease is the popliteal artery. Blood flow is blocked by a gelatinous substance that expands the surface of the vessel from the outside [1]. This disease usually affects middle-aged people (40–50 years), who have no history of vascular disease. Men are five times more likely than women to develop the condition [2].

Written consent was obtained from the patient to publish the case report.

Case report

A 43-year-old man was admitted to the Department of Angiology in order to diagnose pain in the right lower

limb. About 6 months before admission to the clinic, the patient suffered a knee injury while cycling. Before the accident, the patient was physically active and played basketball in the past. After the injury, the patient began to experience periodic pain symptoms in the right calf, which subsided after about 10 minutes of rest. Subsequently, constantly occurring intermittent claudication appeared. The patient denied limb rest pain.

The patient does not have chronic diseases, and recently started smoking cigarettes (up to 5 per day). Family history unburdened. Before hospitalization, the patient was consulted by an orthopedist — a diagnosis of popliteal cyst was made and a puncture attempt was made, which was ineffective. In the physical examination at rest, pulses on the lower limbs were symmetrically present at the typical sites of the examination, and superficial sensation and tendon reflexes were preserved. There was no limitation of the range of motion in the knee and ankle joints. There were no

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skin-warming disorders of the right lower limb, and no discoloration was observed. In the measurement of the ankle joint index at rest, there was no difference between the limbs — the measured ABI on both sides was about 1.

Doppler ultrasound (USG Doppler) examination of the right popliteal fossa revealed the presence of numerous fluid spaces with diameters up to 10 mm, in the immediate vicinity of the popliteal artery. This caused pressure on the blood vessel with a narrowing of the lumen of blood flow (Fig. 1, 2). However, no increase in peak systolic velocity was observed. Suspicion of cystic degeneration of the adventitia was raised.

Magnetic resonance imaging (MRI) of the knee region was then performed, in which the popliteal artery was visualized with numerous cystic structures of various sizes visible in the proximal and distal parts (9 cm long). Cysts have a high signal in T2 and a low signal in T1, they do not contrast. The largest cyst at the height of the proximal tibia has dimensions of 1.3 cm AP x 1.4 AP x 2.7 cm CC (Fig. 3, 4). The cysts press on the lumen of the artery and cause narrowing in the distal segment (at the level of the proximal tibia) at a length of 2.0 cm to 0.2 cm in the transverse dimension (TR) and 0.7 cm in the AP dimension.

Angiosurgical treatment was recommended, to which the patient agreed. The right popliteal fossa was incised, and the popliteal artery was dissected along its entire length. At a length of about 10 cm, changes in the adventitia with large cysts in the distal part were found (Fig. 5). The adventitia from the popliteal artery was removed together with the cyst, which resulted in the normalization of the flow in the popliteal artery (Fig. 6). Pinpoint bleeding from the popliteal artery occurred, which was treated with vascular punctures 6.0. After decompression, a normal pulse was observed in the arteries below the bifurcation of the popliteal artery. Hemostasis was checked with Fibrylar, the wound was dried and sutured in layers. The wound was covered with a sterile dressing and 5000 IU UFH was administered intraoperatively.

In the postoperative period, no early complications were observed, and the patient remained in outpatient care. He returned to physical activity, and did not report intermittent claudication.

In order to assess the imaging after surgery, a follow-up MRI examination was performed after 9 months, which showed complete regression of the ganglion complex in the immediate vicinity of the popliteal artery visible in the previous examination, without stenosis of the lumen of the popliteal artery (Fig. 7). The complex of smaller ganglions is now more superficially visible in the popliteal fossa adjacent to the tibial nerve (with the largest component superficially to this nerve), about

1.5 cm in diameter, with mild nerve modeling. Several merging smaller cysts are also visible between the tibial nerve and the proximal cutaneous nerve of the medial calf. A stable image of a small several-chamber branch of the ganglion penetrating within the deep part of the popliteal fossa, laterally from the popliteal artery, under the initial segment of the soleus muscle, reaching the area of the fibula head and probably coming out of this area.

Discussion

The first report of Adventitial Cystic Disease was described in 1947 [3] in the external iliac artery. Since then, there have been several descriptions of individual cases documenting the diagnostic difficulties caused by this disease. Patients are often diagnosed by orthopedists due to the localization of complaints and the effect of mass in the popliteal fossa. It often takes a long time for patients to reach the vascular center.

The disease most often affects the popliteal artery, less often described are changes in the external iliac artery, and femoral common artery as well as in the radial, ulna, brachial and axillary arteries [4]. There are isolated reports of vein involvement [5]. CAD is most often diagnosed in young, previously non-chronically ill men with typical symptoms of intermittent claudication; hence the differential diagnosis should take into account popliteal artery entrapment syndrome. In the differentiation of these diseases, the Ishikawa symptom, which is manifested by a decrease in the pulse on the foot with passive flexion of the knee, may be helpful.

The etiology is not known, the literature cites the theory of microtrauma which causes a separation of the adventitia from the media resulting in adventitial cystic degeneration or embryological disorders with the presence of mesenchymal cells in the artery wall. Cysts may be caused by connective tissue disorders or ectopia of synovial ganglia implanting in the vessel wall (articular theory) [5].

The basis for the diagnosis is Doppler ultrasound and MRI. Doppler ultrasound is a minimally invasive, convenient diagnostic method for routine use, while MRI is more useful for assessing the morphology of the cyst and possible communication between the cyst and the adjacent joint.

Ultrasound examination is accepted as a diagnostic method of first choice, while the MR results are quite unambiguous because cysts typically appear as regions of homogeneous, low signal intensity in T1 images and multipatch high signal intensity adhering to vessels in T2 images [6].

To date, no method has been found to prevent the onset of cystic adventitial degeneration.

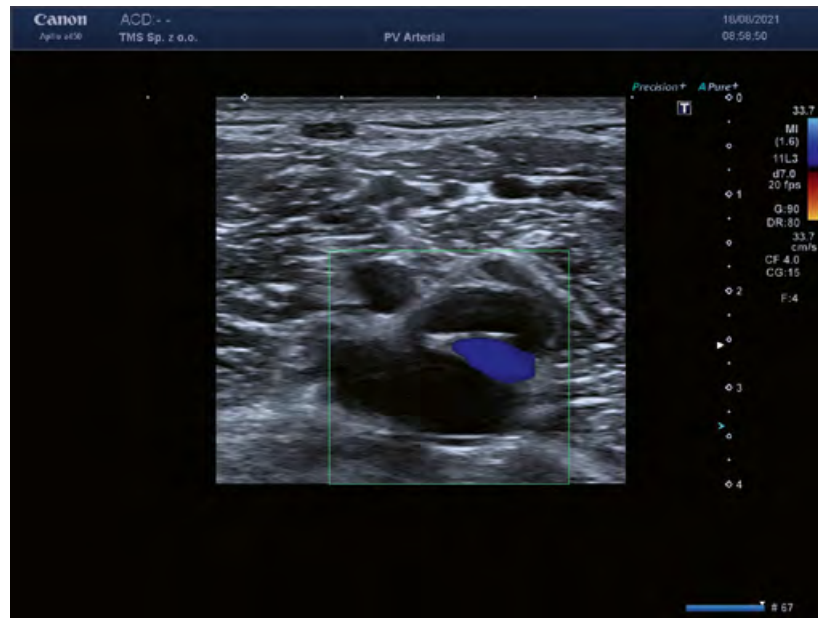


Figure 1. Transverse ultrasound through the popliteal fossa revealed a hypoechoic cyst around the popliteal artery

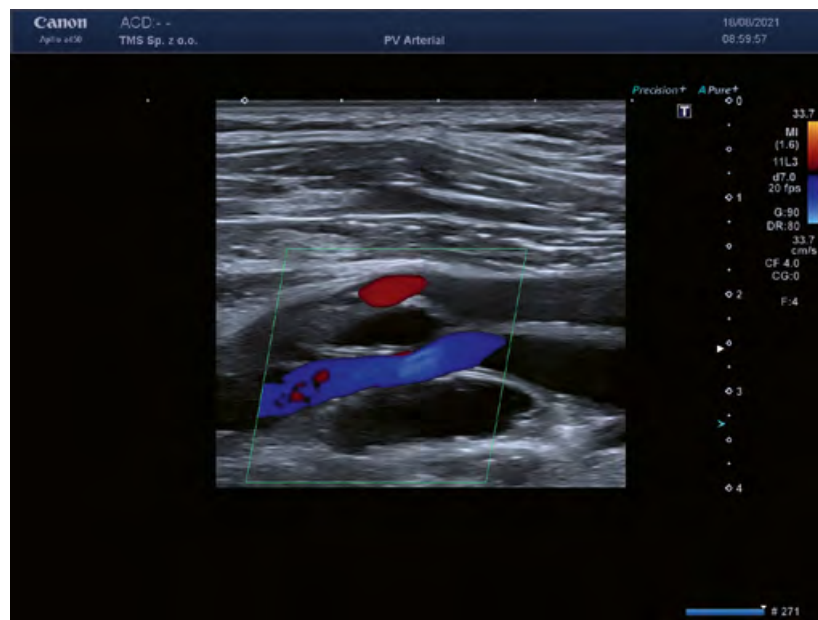


Figure 2. The longitudinal Doppler ultrasound showed compression of the popliteal artery

Small adventitial cysts causing only moderately severe symptoms may be observed as spontaneous regression of the cyst has also been reported. In case of severe symptoms or complete obstruction of the vessel, surgery is the preferred method.

The goal of treatment is complete resection or enucleation of the cyst, including the artery wall. Revascularization is usually performed with saphenous vein interposition graft. This procedure has the lowest number of relapses, but it does not guarantee that the disease will not return [7]. Alternatives to surgical

resection and revascularization include cyst aspiration. Endovascular stenting alone was not effective in a long-term follow-up, however, there have been cases of treatment of recurrence with angioplasty alone [8].

Conclusions

We described a patient with adventitial cystic disease of popliteal artery stenosis causing symptoms of lower extremity claudication. The cysts were resected by open surgery, without vein graft. The patient rema-



Figure 3. Preoperative magnetic resonance images showing cystic mass adjacent to the popliteal artery

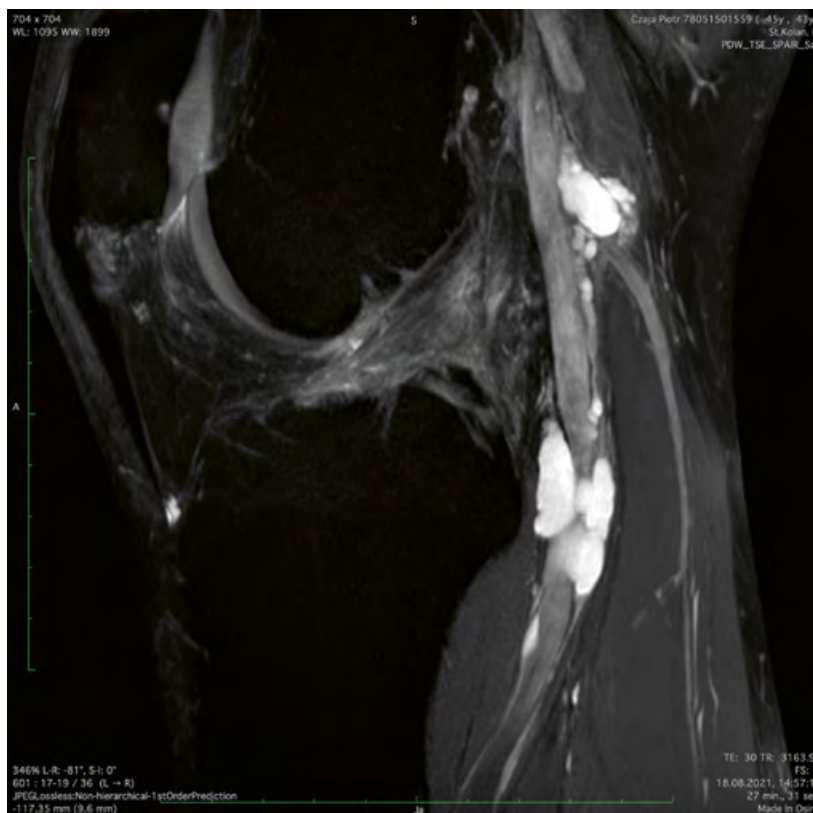


Figure 4. Sagittal view shows popliteal artery compressed by cystic mass

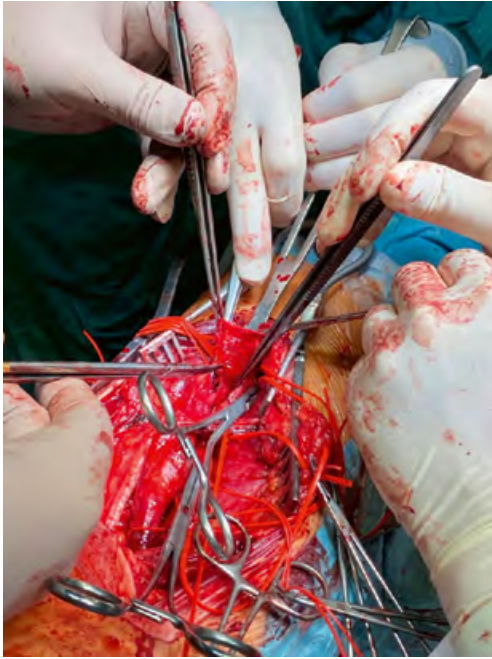


Figure 5. Popliteal artery surrounded by cysts is concentrically compressed



Figure 6. Surgical specimen comprising resected adventitial layer of artery wall with adjacent cyst



Figure 7. MRI 9 months after treatment shows normal patency of the vessel

ined free of symptoms after the procedure, and the postoperative MRI showed good patency of the Vessel.

Longer follow-up is necessary because of the risk of recurrent stenosis.

Conflict of interest

None.

References

1. Speziale F, Sirignano P, Mansour W, et al. Vascular Surgery. A Clinical Guide to Decision-making, 2022: 221–230.
2. Bartholomew AA. Cleveland Clinic medical. "Adventitial Cystic Disease: Causes, Symptoms & Treatment." Cleveland Clinic, my.clevelandclinic.org/health/diseases/16722-adventitial-cystic-disease (Accessed 20 Aug. 2023).
3. Atkins HJB, Key JA. A case of myxomatous tumour arising in the adventitia of the left external iliac artery; case report. *Br J Surg.* 1947; 34(136): 426, doi: [10.1002/bjs.18003413618](https://doi.org/10.1002/bjs.18003413618), indexed in Pubmed: [20247247](https://pubmed.ncbi.nlm.nih.gov/20247247/).
4. Smith JL, Hariri N, Oriowo B, et al. Cystic adventitial disease of the popliteal artery presenting with features of entrapment syndrome. *J Vasc Surg Cases Innov Tech.* 2020; 6(1): 75–79, doi: [10.1016/j.jvscit.2019.10.009](https://doi.org/10.1016/j.jvscit.2019.10.009), indexed in Pubmed: [32099935](https://pubmed.ncbi.nlm.nih.gov/32099935/).
5. Ching-Yee Chan M, Cornwall J, Ilonzo N, et al. Cystic adventitial disease of the popliteal vein and artery in siblings. *J Vasc Surg Cases Innov Tech.* 2021; 7(3): 545–548, doi: [10.1016/j.jvscit.2021.04.011](https://doi.org/10.1016/j.jvscit.2021.04.011), indexed in Pubmed: [34401622](https://pubmed.ncbi.nlm.nih.gov/34401622/).
6. Kawarai SI, Fukasawa M, Kawahara Yu. Adventitial cystic disease of the popliteal artery. *Ann Vasc Dis.* 2012; 5(2): 190–193, doi: [10.3400/avd.cr.11.00069](https://doi.org/10.3400/avd.cr.11.00069), indexed in Pubmed: [23555509](https://pubmed.ncbi.nlm.nih.gov/23555509/).
7. Lim JH, Chung BH, Kang JH, et al. Surgical strategy to reduce the recurrence of adventitial cystic disease after treatment. *Vasc Specialist Int.* 2019; 35(4): 217–224, doi: [10.5758/vsi.2019.35.4.217](https://doi.org/10.5758/vsi.2019.35.4.217), indexed in Pubmed: [31915666](https://pubmed.ncbi.nlm.nih.gov/31915666/).
8. Jaff MR, Bartholomew JR. Other peripheral arterial diseases. In: Goldman L, Schafer AI, eds. *Goldman-Cecil Medicine.* 26th ed. Philadelphia, PA: Elsevier; 2020: chap 72.