Popliteal Adventitial Cystic Disease. Case Report

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Cystická degenerace adventicie
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Klaudikace
Otevřené chirurgické výkony

SOUHRN

Cíl: Popisujeme případ pacienta, u něhož byla stanovena diagnóza cystické degenerace adventicie popliteální tepny jako vzácného onemocnění; zároveň představujeme diagnostický algoritmus a proces rozhodování.
Metody: Popisujeme případ 51leté zdravotní sestry, která si stěžovala na rychle progredující bolest v pravém lýtku během cvičení. Nejdříve jí byla stanovena chybná diagnóza a byl u ní indikován endovaskulární výkon. Až po vyšetření metodami CT a MR byla stanovena správná diagnóza a byl proveden otevřený chirurgický výkon. Po odstranění cysty z adventicie zůstala adventicie otevřená. Díky zdravé a nejspíše pevné stěně tepny nebyla nutná žádná další rekonstrukce.
Výsledky: Pacientka byla propuštěna dva dny po výkonu s hmatným distálním pulsem a beze známek ischemie pravé dolní končetiny. Pooperační průběh byl nekomplikovaný. Histologicky se cysta podobala gangliu.
Závěr: Domníváme se, že diagnosticky náročné případy, jako byl tento, vyžadují multidisciplinární přístup, aby bylo možno správně určit zdroj ischemie u poměrně zdravého pacienta bez známek aterosklerózy postihující několik úrovní organismu. Jako léčbu první linie je nutno vždycky zvážit, zda se lze vyhnout rekonstrukci tepen, zvláště u pacienta bez vhodné žilní spojky.

Keywords:
Claudication
Open surgery
Popliteal adventitial cystic disease

ABSTRACT

Aim: We want to present a case of a patient who was diagnosed with the rare condition of popliteal adventitial cystic disease and to describe the diagnostic algorithm and the decision making process.
Methods: We report a case of a 51-year-old female nurse complaining of rapidly progressing claudication involving the right calf during exercise. She was misdiagnosed at the beginning and endovascular procedure was scheduled. Only by performing CT-scan and MRI the right diagnosis was made and open surgery was performed. The adventitial cyst was excised and the adventitia was left open. No further reconstruction was needed due to healthy and presumably strong arterial wall.
Results: The patient was discharged 2 days after the intervention with palpable distal pulse and no signs of right lower limb ischemia. Postoperative course was uneventful. The cyst was histologically similar to a ganglion.
Conclusion: We believe that in challenging diagnostic cases like this multidisciplinary approach is important to correctly identify the source of ischemia in a relatively healthy patient with no signs of multilevel atherosclerosis. Avoiding arterial reconstructions especially in a patient without appropriate venous conduit must be always considered a first line treatment.

Introduction

Cystic adventitial disease (CAD) is a rare but well documented form of nonatherosclerotic arterial stenosis; it affects the popliteal artery most often but can affect any artery or vein.[1] A total of 85% of cases is found usually in the popliteal region. The patients are generally young and otherwise healthy with no signs of multilevel atherosclerosis. That is the reason why durable and long-lasting treatment strategies should be always considered.

The incidence of CAD is 1 in 1 200 cases of claudication and 1 in 1 000 femoral angiograms with a male predominance of 15 : 1.[2]

Case report

A 51-year-old female referred to our hospital complaining of claudication in the right lower extremity which was getting worse in the last couple of months. According to

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the patient the pain started a year ago when she visited a vascular specialist and conservative therapy was prescribed together with appropriate exercise regime. The conservative therapy gave an unsatisfactory result and the patient has been experiencing severe pain walking very short distances for one month. The patient has tried several pain medications with little or no effect.

Our patient had no previous medical history of hypertension or coronary heart disease. Furthermore we couldn't confirm atrial fibrillation or any previous embolic incidents.

Upon examination the patient had palpable femoral pulse on the affected extremity, barely palpable popliteal and absent distal pulse. Resting ankle brachial index (ABI) was 0.35 on the right and 1.0 on the left side.

The patient was scheduled for angiography due to the lack of iliac artery disease or abdominal aortic aneurism confirmed by thorough ultrasound examination. The angiography showed severe stenosis of the popliteal artery in P1 segment around 4cm in length. The rest of the vessels had no signs of atherosclerotic disease and the patient had 3 vessels run off below the knee (Fig. 1).

Nevertheless the angiography picture rose the suspicion of external compression which led to change of strategy and CT-angiography was performed a couple of hours later (Fig. 2). The CT-angiography showed dilated popliteal artery around 10–11 mm with thrombosis

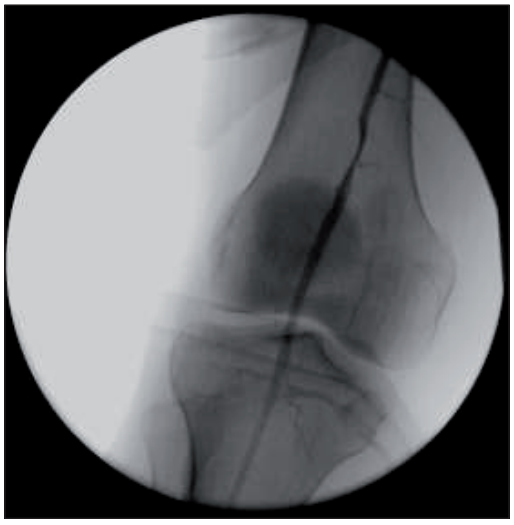

Fig. 1 – Angiography showing stenosis of the popliteal artery.

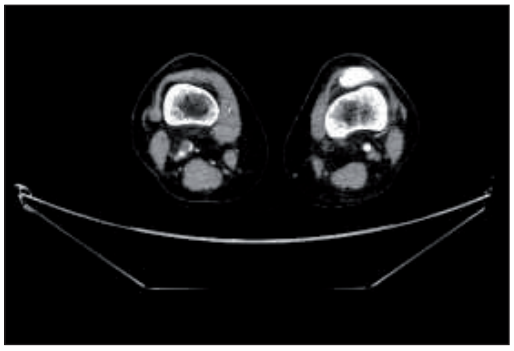

Fig. 2 – CT-angiography showing dilatation of the popliteal artery.

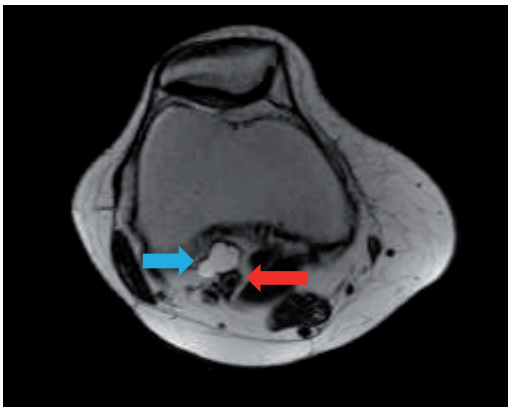

Fig. 3 – (A) MRI showing cystic formation (blue arrow) in close contact with the popliteal artery (red arrow).

Fig. 3 – (B) MRI showing genicular branch with cystic degeneration.

narrowing the arterial lumen about 90%. At this point we consider the diagnosis of popliteal artery aneurysm (PAA). The patient was reevaluated and our surgical team had a discussion with the head of the radiology department.

Upon deeper examination small cystic-like formation was found around the popliteal artery and the differential diagnosis was broadened. Three hypotheses were proposed – popliteal adventitial cystic disease, ganglion or popliteal entrapment syndrome. In order to establish the most likely diagnosis MRI was performed which led us to the conclusion that CAD is the most likely cause of the popliteal artery stenosis (Figs 3A, 3B).

Surgical technique

Although the popliteal artery could have been reached with medial incision above knee we decided to perform a posterior approach which would give us access to the posterior wall of the artery where we suspected the cyst would be (Fig. 3A). This decision was made based on thorough ultrasound examination and carefully studied 3D reconstructions prior to surgery. An S-shaped skin incision was cut linear to the skin fold in the right popliteal fossa under loco-regional anesthesia with the patient in the

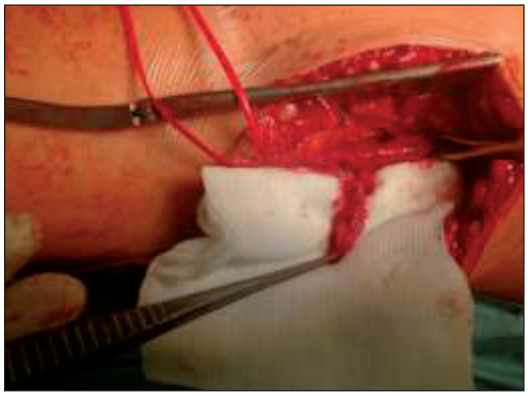

Fig. 4 – Genicular branch with cystic degeneration.

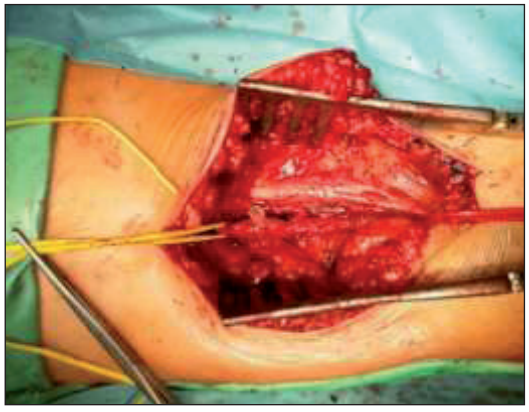

Fig. 5 – Evacuation of gel-like substance beneath the adventitial layer.

prone position throughout the procedure. Once we had control of the popliteal artery we noticed cystic dilatation of the arterial wall around 4–5 cm in length. Furthermore we saw most likely a genicular branch arising from the popliteal artery which showed signs of cystic dilatation and we followed the branch to the knee capsule and ligated it as far as possible because it was considered a potential source of disease recurrence (Figs 3B, 4). First we resected this branch and sent it for histology exam. Then we performed a light incision alongside the affected part of the popliteal artery, opening only the adventitial layer and we evacuated sufficient amount of gel-like substance which we sent again to histology (Fig. 5).

Once we had all the pathologic structured resected a discussion was carried out whether to close the adventitial layer with a direct suture, place a patch or leave it wide open. Prior to operation we checked the arm veins, GSV and SSV and they were all considered inappropriate for bypass grafting so interposition was not considered. Another option would be using a synthetic graft but due to inferior patency this option was abandoned. Due to the otherwise healthy artery, the immediate pulse restoration and the unknown etiology of the disease which could lead to another subadventitial cystic formation in this region we decided to leave the artery with no further reconstruction (Fig. 6).

The patient was discharged two days after the operation with palpable distal pulse and no signs of ischemia.

One month after the initial operation the patient came back and another CT-angiography was performed.

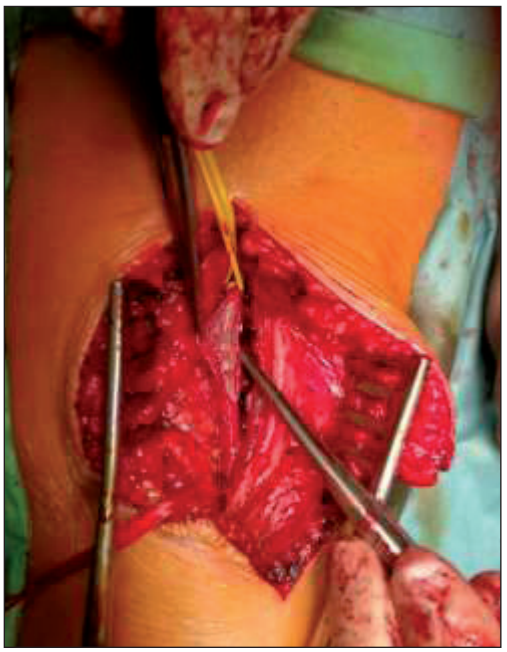

Fig. 6 – Popliteal artery after resection of the adventitial cyst.

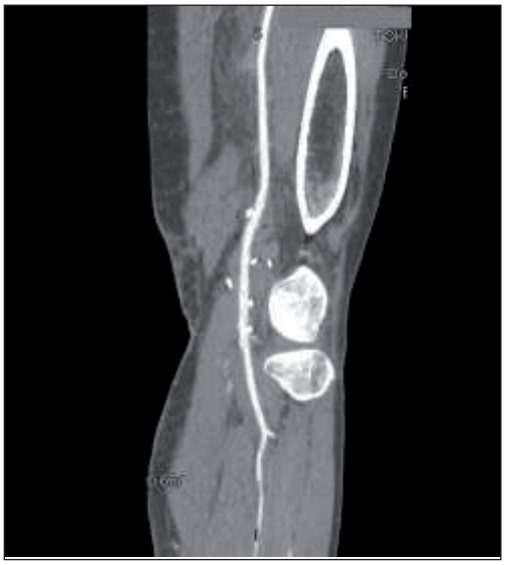

Fig. 7 – Patent popliteal artery.

It demonstrated patent popliteal artery with no signs of disease (Fig. 7).

Discussion

The etiology of CAD remains controversial and four potential theories have been proposed, which include the following: 1) repetitive local trauma, 2) systemic disease, 3) synovial/ganglion, and 4) embryological.[3]

The repetitive local trauma theory postulates that due to the location of affected vessels near joints, they are subject to repetitive microtrauma that can result in disruption of the adventitia from the media, resulting in intramural bleeding and development of cysts due to en-

zymatic activity within the vessel wall.[4] There have been reports of trauma induced cysts; however, this theory is controversial as CAD can occur in children and is rare in athletes.[5,6] Linquette et al. proposed the systemic disease theory which suggests that CAD is related to a generalized disorder of myxomatous degeneration. This theory has failed to gain adequate support as patients did not show systemic lesions on follow-up.[7,8] The synovial/ganglion theory and the embryological theory have recently gained more favor in the etiology of CAD. The synovial/ganglion theory proposes that adventitial cysts develop due to migration of synovial ganglions from the adjacent joint capsule or tendons, along vascular branches to the adventitia.[9,10] In our case this seems to be the most logical explanation to this condition because we were able to identify the genicular branch which we followed to the capsule. The embryological theory hypothesizes that during development, mesenchymal cells are incorporated into the adventitia from adjacent joint tissue. These cells then secrete mucin over a number of years giving rise to adventitial cysts which have the potential to encroach on the arterial lumen.[5,8] Because there is no single unifying theory able to account for the pathogenesis of all clinical cases, CAD may be better explained by a combination of multiple theories.

Like all vascular conditions CAD can be treated endovascularly or with open surgery. In the literature there's a lot of negative experience regarding mini invasive treatment with PTA and/or stent placement including early restenosis or instent thrombosis. That is the reason why endovascular approach has limited place in the treatment of this non-atherosclerotic condition.

The most cited treatment modality includes cyst incision which leads to decompression of the artery and return to normal lumen diameter. The success rate is up to 94% but failures are not uncommon due to cyst recurrence.

The next approach would be venous interposition with resection of the diseased segment of the artery. In our case we excluded this option due to the lack of appropriate autologous vein material. The success rate cited in the literature is between 93.5–95%.[11] Bypass failure is like with every other surgical bypass and is due to neointimal hyperplasia with subsequent stenosis and thrombosis eventually.

Interposition using prosthetic graft is another viable option but has inferior patency rate compared to venous interposition and in our opinion should be avoided in young patients with long-life expectancy

Radiologically guided aspiration of cysts has been reported with mixed results. Some cases have required multiple aspirations before satisfactory resolution, some attempts have failed due to thickness of the fluid, and some have been successful. Although cyst preserves autologous endothelium, which is highly desirable, long-term CAD does recur after aspiration rendering this treatment strategy as ineffective.[12]

No matter which treatment modality is chosen careful follow-up is mandatory. Our protocol includes follow-up visits 2 weeks after the intervention, at the end of months 3, 6, 12, and then once a year. At the end of the sixth month the patient had no signs of ischemia and ultrasound exam showed no abnormalities. We scheduled a MRI test at the end of the first year.

Conclusion

The case we presented emphasizes the importance of multidisciplinary approach because the patients with CAD are often unsuccessfully treated endovascularly before they get the right diagnosis. Clinical history, physical examination, and CT or MR angiography are crucial in making a definitive diagnosis. We believe that in relatively young patients with no signs of atherosclerosis with suspected popliteal artery pathology MRI is the best diagnostic modality to distinguish between CAD, ganglion or popliteal entrapment syndrome.

Open surgery remains the treatment of choice for these kinds of lesions but more data is required. Due to the lack of venous conduit we decided to cut the adventitia open and evacuate the cyst leaving it without any arterial reconstruction. The early results are very promising but longer follow-up is needed.

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