

True mycotic popliteal artery aneurysm in a patient with fungal endocarditis: A case report

Fungal endokarditli hastada popliteal arterin gerçek mikotik anevrizması: Olgu sunumu

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ABSTRACT

A 32-year-old male presented with pain in the left lower extremity while walking and a pulsatile mass on the backside below the left knee level. Aneurysmal dilatation was reported in the popliteal artery in the magnetic resonance angiographic examination. Following the surgical resection of the sac and revascularization, fungal fibers were detected in the histopathological examination and microbiological evaluation of the specimen. Considering the potential etiology, a detailed cardiac assessment was carried out in asymptomatic patient. On echocardiographic evaluation significant vegetation on the anterior leaflet of the mitral valve and severe mitral insufficiency were detected. With appropriate antifungal treatment and successful surgical management complete recovery was achieved.

Key words: Mycotic aneurysm, *Candida albicans*, popliteal artery, fungal endocarditis

ÖZ

Otuz iki yaşındaki erkek yürürken sol alt ekstremitte ve sol diz seviyesinin altında ters bir pulsatil kitle ağrı ile başvurdu. Manyetik rezonans anjiyografi incelemede popliteal arterin anevrizmal dilatasyonu bildirildi. Kesenin cerrahi rezeksiyonu ve revaskülarizasyonun ardından, histopatolojik inceleme ve mikrobiyolojik değerlendirmesinde mantar hifleri tespit edildi. Potansiyel etiyojisi göz önüne alındığında, asemptomatik hastada detaylı bir kalp değerlendirmesi yapıldı. Ekokardiyografik değerlendirmede ön mitral kapak üzerinde belirgin vejetasyon ve ileri mitral yetmezlik saptandı. Uygun antifungal tedavi ve başarılı cerrahi tedavi ile tam iyileşme sağlandı.

Anahtar kelimeler: Mikotik anevrizma *Candida albicans*, popliteal arter, fungal endokardit

INTRODUCTION

Arterial aneurysm may develop after a localized or systemic infection, arterial dissection or trauma. Popliteal artery is the commonest site for peripheral arterial aneurysms and the commonest etiology is atherosclerosis. On the other hand, mycotic aneurysm of the popliteal artery is extremely rarely observed and mostly associated with infected endocarditis¹. The most frequently isolated organisms are *Staphylococcus* spp. and *Salmonella* whereas fungi are very infrequent^{2,3}. The term “mycosis” refers to a fungal infection; however, mycotic aneurysm of the

vessels is a misnomer because bacterial pathogens are mostly isolated. In the literature, there is only one case reported as the aneurysm of the popliteal artery affecting both lower legs associated with *Candida* endocarditis².

Here, a “true mycotic” aneurysm that refers to an aneurysm caused by a fungus is reported with unilateral manifestation of the popliteal artery and asymptomatic *Candida* endocarditis. To us, this is the only case affecting unilateral lower extremity with asymptomatic true mycotic endocarditis.

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CASE REPORT

A 32-year-old male presented with the complaints of claudication in the left lower extremity and with a pulsatile mass on the backside below the left knee level. The patient did not have an additional disease but had intravenous drug abuse. The physical examination revealed normal heart sounds. The left popliteal artery was palpable, however pulsations of left dorsalis pedis and tibialis posterior could be palpated deeply. Magnetic resonance angiographic examination showed aneurysmal dilatation of the popliteal artery (Figure 1) below the knee level. On systematic examination any pathological finding like fever, hyper-, and hypotension, and arrhythmia were not detected. The parameters such as blood counts, CRP and sedimentation rate were entirely normal at the time of referral.

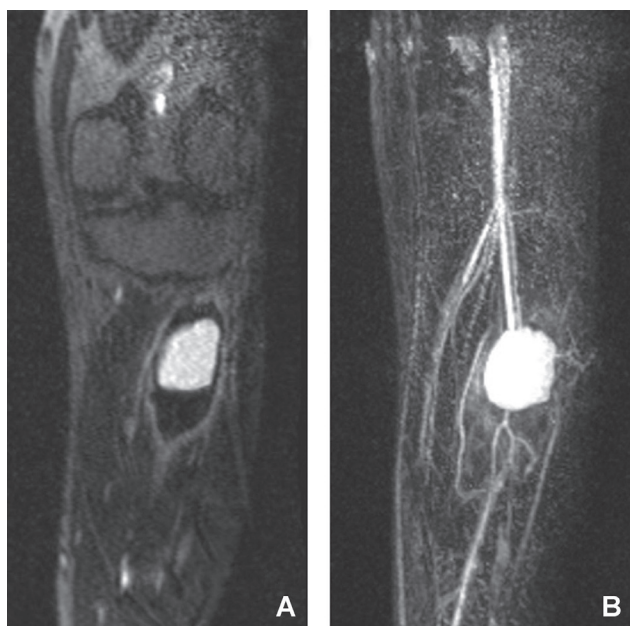


Figure 1. (A) Magnetic resonance angiography of the left leg shows the infra-popliteal localization of the aneurysm; (B) segmental obstructions of the left tibioperoneal trunk.

Under general anesthesia, the patient was prepared in the supine position. The popliteal artery exploration was performed through a below-knee medial incision. During the popliteal artery exploration, quite bulky and uliginous aneurysm sac was observed. The aneurysm sac was tightly adherent to the sur-

rounding tissues including the distal portion of the popliteal artery, tibioperoneal trunk and the popliteal vein. After systemic heparin administration, proximal and distal vascular clamps were applied. The attempt to preserve these vascular structures failed because the the presence of a gross infiltration and absence of a dissection cleavage existed between them and the aneurysm. (Figure 2A). Excision of the aneurysm with the invaded vascular structures was performed (Figure 2B). Harvesting of the left greater saphenous vein was started through the same incision which was extended proximally to the upper-knee segment. Saphenous vein interposition was performed between the popliteal artery and tibioperoneal trunk using 6/0 propylene sutures (Figure 2C). Popliteal vein was also reconstructed with saphenous vein graft interposition using 6/0 propylene sutures (Figure 2C).

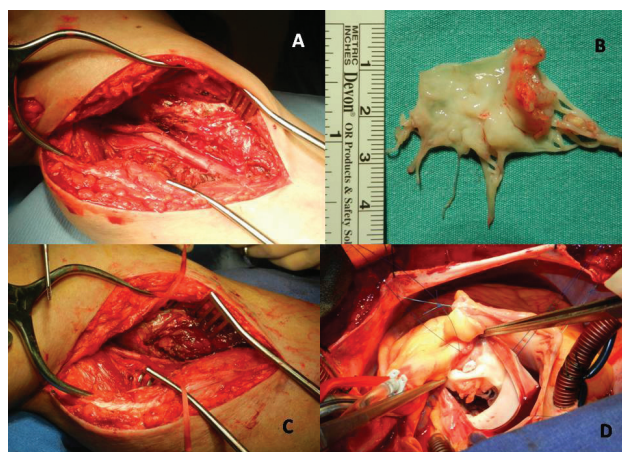


Figure 2. (A) Intraoperative picture of the aneurysm sac settled in between the popliteal artery, tibioperoneal trunk and popliteal vein; (B) Specimen of the resected aneurysmal sac; (C) Intraoperative view of the reconstructed arterial and venous system with autologous vascular grafts; (D) Intraoperative view of the mitral valve with vegetations on the leaflets.

Histopathological examination of the specimen revealed true mycotic aneurysm and microbiological evaluation was reported the agent as *Candida albicans* (Figure 3). After this result, immediate echocardiography was carried out to evaluate the source of the fungal disease based on etiology. There was a significant vegetation and severe mitral insufficiency on the anterior leaflet of the mitral valve (Figure 2D). Intravenous amphotericin B (0.6 mg/kg)

was administered daily. Prior to surgery, regular heparin (5000U, four times a day) was given to the patient for 6 days until definitive cardiac operation was performed. A mitral valve replacement was scheduled. The heart was exposed through median sternotomy. Cardiopulmonary bypass was initiated, so cross clamping was performed. Intermittent doses of antegrade tepid blood cardioplegia was administered. Superior septal incision was performed to enter the left atrium. Large vegetation on the anterior leaflet of the mitral valve was noted intraoperatively. Following excision of both leaflets of the mitral valve, a radical tissue debridement was performed. A bi-leaflet 29 no mechanical mitral heart valve (St. Jude Medical Inc., USA) was implanted using 2/0 pledgetted non-absorbable polyfilament horizontal mattress sutures. After transferring to the intensive care unit, the patient was extubated at the 8th postoperative hour. The postoperative course was uneventful.

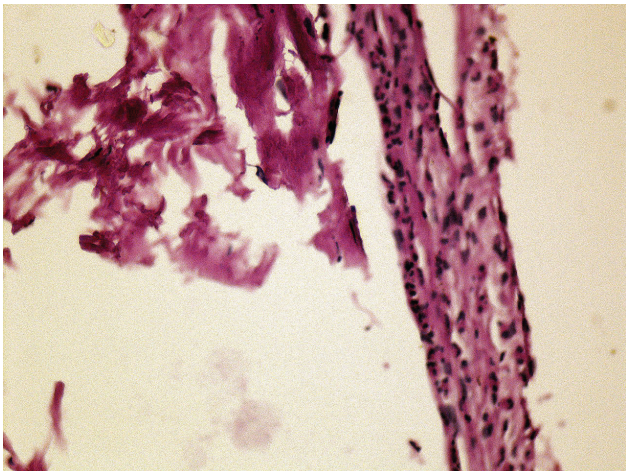


Figure 3. Histopathological evaluation of the specimen: Please note the sparse spores and hyphae (PAS stain, X200 in magnification).

During the hospitalization period antifungal treatment with amphotericin was prescribed for a total of 6 weeks. At the end of the 6th week, the patient was discharged with a complete recovery. On the 2-year follow-up, the patient was asymptomatic and doing well. Lifelong suppressive oral fluconazole therapy was recommended; however the patient refused to take oral fluconazole for such a long time.

DISCUSSION

An aneurysm is a localized permanent dilatation of an artery greater than 50% of its expected normal diameter¹. Aneurysms are classified into true and false or pseudoaneurysms. Infection, dissection or trauma may lead to an aneurysm of an artery. The term “mycosis” refers to a fungal infection in microbiology; however, mycotic aneurysm of the vessels is a misnomer because bacterial pathogens are mostly isolated rather than fungi.

An aneurysm of the popliteal artery in a patient with endocarditis was described by Tufnell in 1853⁵. Sir William Osler was the first to use the term “mycotic aneurysm” in 1885⁶. Intracranial vessels are the commonest sites for mycotic aneurysms (65%). Extracranial vessels are affected less frequently⁷. Mainly, aorta and femoral arteries are the most affected extracranial vessels^{7,8}. If located in an extra-aortic position, an infectious cause can be found in 3.6-12.3% of the cases^{8,9}. Infected aneurysms of the popliteal artery are extremely rare (1-2% of all popliteal sacculations) and infectious agents are not always detected in vessels with a clinically infected aneurysm (25%)^{2,3,10}. In our patient, *Candida albicans* was isolated both in the aneurysm sac and mitral leaflet. Moreover, histopathological evaluation also supported these findings.

Mycotic aneurysms are not frequently observed after infective endocarditis and a report described that 3-15% of the patients with infective endocarditis may complicate aneurysms¹¹. According to Wilson, mycotic aneurysms are strictly defined as infected aneurysms developing in a previously normal artery secondary to septic embolization due to bacterial endocarditis¹².

Since healthy vessel is very resistant to infection, they become infected when the patient is immunocompromised or the pathogen is extremely virulent. The common causative organisms for mycotic popliteal artery aneurysm are *Staphylococcus aureus*, *Staphylococcus epidermidis*, *Streptococcus viridans*, *Strep-*

Staphylococcus faecalis, *Streptococcus pneumoniae*, *E.coli*, *Salmonella spp.*, *Camphylobacter jejuni*, *Mycobacterium spp.* or the patients may be culture negative⁷. On the other hand, true mycotic aneurysms are associated with fungal infections caused by *Histoplasma capsulatum*, *Aspergillus fumigatus*, *Candida albicans* and *Penicillium species*¹³. Aorta is the mostly affected predilection site for the *Candida*-associated mycotic aneurysms. The most common causes of fungal endocarditis are *Candida species* followed by *Aspergillus species*¹⁴. There is only one case of *Candida* endocarditis reported in the literature whose crural arteries were affected bilaterally².

Most of the popliteal mycotic aneurysms are asymptomatic at the time of diagnosis. The most common symptoms include claudication and hypokinesia of the lower extremity, fever, fatigue, and pain. Some symptoms secondary to pressure on veins or nerves around the aneurysm may also be observed. As diagnostic tools duplex ultrasonography, computed tomography and magnetic resonance angiography can be applied.

The treatment consists of embolization or the excision of the sac with appropriate vascular reconstruction in the lower extremity². The surgical approach for infectious and non-infectious popliteal artery aneurysms is different. It is recommended to revascularize the lower extremity with an autologous graft in infectious aneurysms using polytetrafluoroethylene and polyethylene terephthalate grafts in order to prevent repeated infections¹⁵⁻¹⁷. In our patient, by taking into account of all these factors revascularization was achieved with autologous saphena magna vein graft.

Current guidelines for endocarditis suggest initial or induction therapy with amphotericin B w/o flucytosine in conjunction with the removal of vegetation, followed by chronic suppressive therapy with oral fluconazole^{18,19}. Suppressive treatment with oral fluconazole is often maintained long term, even lifetime to control the late relapse⁴. Despite recommendations for lifelong suppressive therapy, our patient

had stopped taking his oral fluconazole six months after discharge. However, on the 2-year follow-up he was asymptomatic and doing well.

The practitioner should keep in mind the possibility of true mycotic aneurysm in patients with atypically localized, de-capsulated, undistinguishable and destructive lesions. Microbiological and histopathological evaluation will accompany the diagnosis as well. Henceforth, a silent and insidiously advancing mycotic endocarditis will be distinguished and an appropriate management will be ensured just as described in our patient.

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