A tibioperoneal trunk mycotic pseudoaneurysm successfully treated with ligation

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ABSTRACT
We present the case of a 68-year-old man with a tibioperoneal trunk mycotic pseudoaneurysm, a rarity in the modern age of antibiotics. We describe the patient’s hospitalizations and workups that ultimately led to this diagnosis and our management with open ligation without bypass. This case highlights the importance of combining a thorough history and physical examination with laboratory and imaging data while keeping in mind a broad differential diagnosis. (J Vasc Surg Cases and Innovative Techniques 2020;6:357-60.)

Keywords: Mycotic pseudoaneurysm; Tibioperoneal trunk; Aneurysm

Mycotic peripheral aneurysms secondary to bacterial endocarditis are uncommon today owing to modern antibiotics and the replacement of infected heart valves. Furthermore, the development of a mycotic pseudoaneurysm (PSA) and/or its rupture is rare. The pathophysiology includes embolization of bacteria and invasion of the intima with subsequent degradation of the intima and media, leading to aneurysm. Bacterial emboli more frequently travel to the lower extremities with the most common site being the bifurcation of the common femoral artery. Involvement of infrapopliteal vessels is even less common, with few modern case reports. Here we present the case of a patient with a history of mitral valve endocarditis and persistent left lower leg neuropathy. He was found to have a large PSA at the tibioperoneal trunk (TPT) after persistent left leg neuropathy. Written consent was obtained from the patient to include all information relative to the case as well as the images we have selected.

CASE REPORT
The patient is a 68-year-old man who initially presented with 3 days of left leg swelling, erythema and increasing pain, in the setting of 2 months of generalized fatigue. He stated that the rash started after using warm compresses (Fig 1). Laboratory results revealed a leukocytosis of 15.5 thousand/μL with 21% bands. Duplex scanning was negative for deep venous thrombosis (DVT) and computed tomography scanning showed only subcutaneous edema at the site of the rash. He was placed on antibiotics for concern of cellulitis. The infectious disease team was concerned for a vasculopathy, so a biopsy was obtained. However, final pathology only revealed perivascular lymphocytic inflammation without signs of vasculitis. Blood cultures grew out Streptococcus mutans, which was believed to be secondary to his recent dental procedures. This was followed by a transthoracic echocardiogram showing a probable mitral valve (MV) vegetation with mild to moderate regurgitation and no other structural heart disease. He was started on intravenous ceftriaxone, which was continued for 4 weeks outpatient. A 1-month follow-up transthoracic echocardiogram showed resolution of the MV vegetation, but with MV sclerosis and persistent moderate regurgitation. He continued to complain of left foot paresthesia, mostly on the bottom of foot. An electromyogram was performed and suggestive of a possible left sciatica neuropathy or lumbosacral plexopathy; however, magnetic resonance imaging of the lumbar spine was unable to explain the left foot paresthesia.

The patient then represented to the hospital 2 months later with acute onset of left leg pain and left calf swelling. He had no other symptoms, such as fever, chills, chest pain, or new changes in sensation to the extremity. Duplex imaging showed a soleal DVT. Vascular surgery was consulted at this point. A computed tomography angiography of the head and neck

Fig 1. Left leg rash seen during the initial evaluation.
was done to rule out a cerebrovascular mycotic aneurysm given the endocarditis history. Because this examination was negative, he was started on anticoagulation for the DVT.

He followed up with his primary care provider 2 months later, where the patient pointed out a palpable pulsatile popliteal mass. He continued to deny any symptoms such as fevers or chills. Laboratory results were only remarkable for a slight leukopenia of 3.7 thousand/µL and C-reactive protein of 5.8 mg/L. Repeat blood cultures were negative and a sedimentation rate was normal. Arterial duplex scanning was then done, which revealed a proximal posterior tibial artery (PTA) aneurysm measuring 5.3 × 5.0 × 6.0 cm. Given the history of bacterial endocarditis, this finding was concerning for a mycotic aneurysm. Next, computed tomography angiography was completed showing a PSA of the proximal PTA (Figs 2 and 3). An arteriogram was performed to further delineate anatomy and showed a 3-cm PSA with an origin at the distal TPT and proximal PTA (Fig 4). He was taken to the operating room, and a thigh tourniquet was placed and set to 250 mm Hg, after which the PSA was approached through a posterior popliteal incision. The popliteal artery was followed down to the TPT where the PTA and peroneal artery were identified. The PSA sac was carefully dissected off of the soleus; proximal and distal control was obtained and the patient was heparinized with activated clotting time confirmation. After obtaining control of the vessels, the tourniquet was deflated for a total tourniquet time of 63 minutes. An area at the distal TPT was in communication with the PSA sac. The sac was entered and mural thrombus was sent for culture, which was negative. The decision was made to ligate the distal TPT and origins of PTA and peroneal artery as the patient had a normal anterior tibial artery and occlusion of the TPT revealed a palpable dorsalis pedis artery and PTA from retrograde flow.

Over the next 6 months, his neuropathy progressively improved until he had complete resolution. The neuropathy was likely secondary to the aneurysm’s compression on the tibial nerve. He had a palpable dorsalis pedis and PTA. Arterial duplex examination at 6 months showed an ankle-brachial index of 1.3 with retrograde PTA flow. There was no evidence of aneurysmal degradation of the ligated vessels. He was ambulating without any claudication and had no further extremity complaints.

**DISCUSSION**

Osler is credited with introducing the term “mycotic aneurysm,” where in his 1885 Gulstonian Lecture on bacterial endocarditis he refers to “mycotic endarteritis” of aortic aneurysms. However, Osler was not the first to describe a relationship between aneurysms and endocarditis as this had been reported earlier by several authors (summarized well by Sorelius). In his lecture, Osler goes on to mention how septic emboli may cause skin changes: “in severe forms of the disease, hemorrhages are very frequent upon the skin ... they appear, in many instances, to be due to the effect of the poison, just as in other infectious disease; in others they are undoubtedly embolic.” The name mycotic would persist despite the misnomer as the infected aneurysm is rarely of fungal origin.

Mycotic aneurysms can arise through embolism, direct inoculation as in intravenous drug abuse, extension from a vegetation or foreign body, or from lymphatic spread. The bacteria invade the vessel wall causing an inflammatory response. The subsequent destruction of the intima and medial layers then leads to aneurysmal degeneration. Patient presentation can range from vague complaints such as fatigue to septic shock. If late in the course, the patient may present with rupture of the aneurysm.

Mycotic emboli causing aneurysms tend to lodge in the lower extremity and, more specifically, at the bifurcation
of the common femoral artery.\textsuperscript{4,5} Sites of embolization such as the TPT or more distal vessels are rare and found as case reports\textsuperscript{6-8}; a detailed listing of these cases in the literature was published by Leon et al.\textsuperscript{9} It should be considered that the decreased incidence of these more distal PSA may be related to decreased detection rather than truly decreased frequency. Infrapopliteal PSA are much more likely to be of a non-infectious etiology. These are most commonly secondary to trauma\textsuperscript{10,11} or an orthopedic procedure.\textsuperscript{12-16} The incidence of distal PSA is increasing with the increasing rate of transpedal and distal access for interventional and diagnostic procedures.\textsuperscript{17}

In conclusion, although these cases are rare with modern antibiotics, a vascular surgeon must keep this diagnosis on the differential. Indeed, the patient presented here would have benefited from earlier recognition of the true diagnosis and its proper treatment. Management involves antibiotics, addressing the underlying etiology of infection, and surgical repair. Surgical management of ligation with or without bypass relies on judgement, taking into account the location of the aneurysm and distal collateral flow. Obviously, prosthetic material should be avoided if possible.

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