A Case of Endofibrosis Presenting with Embolic Symptoms in a 43-Year-Old Cyclist

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Background: Endofibrosis is a rare clinical entity that usually manifests as claudication in cyclists and other endurance athletes. We report a case of a 43-year-old cyclist presenting with pain and cyanosis of his toes due to an embolism to his left anterior tibial artery. The source of the embolus was found to be an ulcerated, endofibrotic plaque in his left common femoral artery.

Methods: We performed an extensive literature search using the PubMed database and identified 60 results on endofibrosis. Eight articles described thrombosis relating to endofibrosis. None of the articles described an embolic phenomenon relating to endofibrosis. The following search terms were used: endofibrosis, embolic, emboli, embolism, "distal occlusion," cyanosis, thrombosis, and thrombus.

Results: The patient is a 43-year-old male cyclist who presented with pain and cyanosis of his second and third toes on his left foot for 1 week. The affected toes had a dark-purple discoloration involving the tissue overlying the distal phalanges. Computed tomography angiography showed an abrupt occlusion of the left anterior tibial artery in the mid-calf with a non-calcified plaque in the left common femoral artery. There were no other signs of arterial disease. He underwent left common femoral endofibrosectomy with patch angioplasty that revealed an ulcerated endofibrotic plaque with mural thrombus.

Conclusions: This case demonstrates an unusual presentation of a rare clinical entity. While there have been previous reports of thrombosis associated with endofibrosis, to our knowledge this is the first reported case of endofibrosis presenting with embolic symptoms.

Endofibrosis is a nonatherosclerotic arteriopathy that affects cyclists and other endurance athletes. Despite having minimal or no cardiovascular risk factors, young, highly trained athletes develop flow-limiting lesions in the external iliac, common iliac, or common femoral artery that manifest as unilateral claudication during maximal effort. These lesions can occasionally be complicated by thrombosis or dissection.

CASE REPORT

The patient is a 43-year-old man who presented with a 1-week history of pain and intermittent cyanosis of his left second and third toes. He reported having similar mild symptoms since 2007, but came for evaluation after the symptoms became severe over the past week. The pain and cyanosis were precipitated by cold weather and elevation of his foot. Nothing relieved the pain and there are no other associated symptoms; there was no inciting event that the patient could identify. He is an avid cyclist, partaking in amateur competition, cycling 4–8 hr/week since 2004. His total mileage most likely ranged from 50,000 to 100,000 total miles. He has a past medical history of hypertension and hyperlipidemia. He is a former smoker with a 16-pack-year history who quit 11 years ago.

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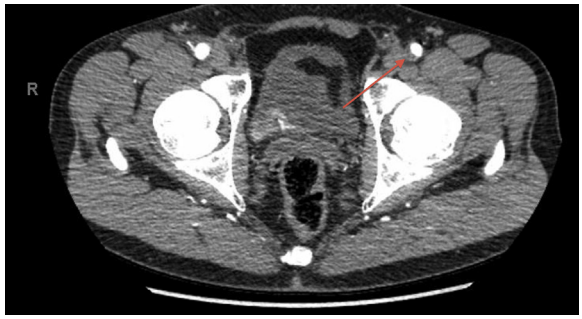

Fig. 1. CT angiogram showing heterogeneous plaque in the left common femoral artery (red *arrow*).

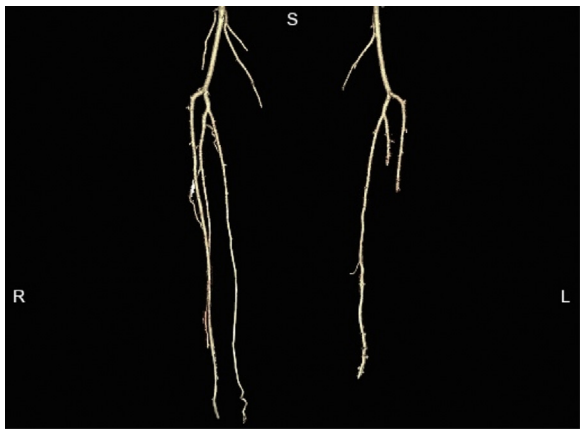

Fig. 2. Three-dimensional reconstruction showing abrupt occlusion of the left anterior tibial artery.

On physical examination, the patient is a fit, well-appearing man. His femoral, popliteal, dorsalis pedis, and posterior tibial pulses were intact, bilaterally. There was purple discoloration of the left second and third toes from the tip to distal interphalangeal joint.

Noninvasive studies revealed an ankle–brachial index of 1.22 on the right and 1.20 on the left; however, his toe–brachial index was 1.11 on the right and 0.38 on the left, suggesting a possible distal occlusion on the left.

A computed tomography (CT) angiogram was obtained which showed an occlusion of the left anterior tibial artery in the mid-calf and an atheromatous-appearing plaque with intraluminal thrombus in the left common femoral artery (Figs. 1 and 2). The anterior tibial artery appeared normal, proximal to the occlusion and there were no other signs of arterial disease. The patient was diagnosed with an embolic occlusion of his left anterior tibial artery. The source was thought to either be endofibrosis of the left common femoral artery with mural thrombus or an aneurysmal left common femoral artery with endoluminal thrombus formation. The patient opted to undergo surgical intervention to remove the source to prevent further emboli.

The left common femoral artery was dilated in the mid-portion with normal artery proximally and distally. Arteriotomy of the common femoral artery revealed a thickened, endofibrotic intima with a large ulceration of the posterior wall containing endoluminal thrombus (Fig. 3). Endofibrosectomy was performed (Fig. 4) and the artery was closed using a bovine pericardial patch.

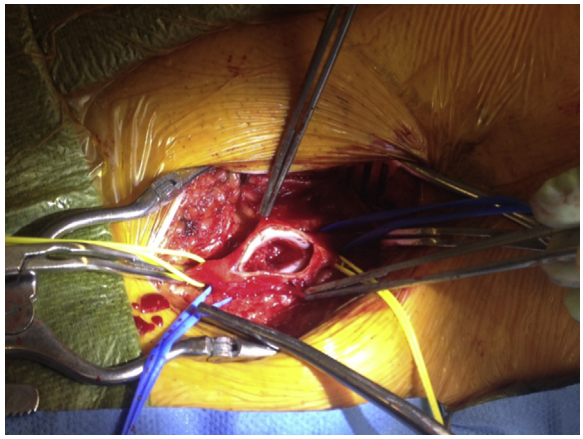

Fig. 3. Arteriotomy revealing a thickened, endofibrotic intima with a large ulceration of the posterior wall.

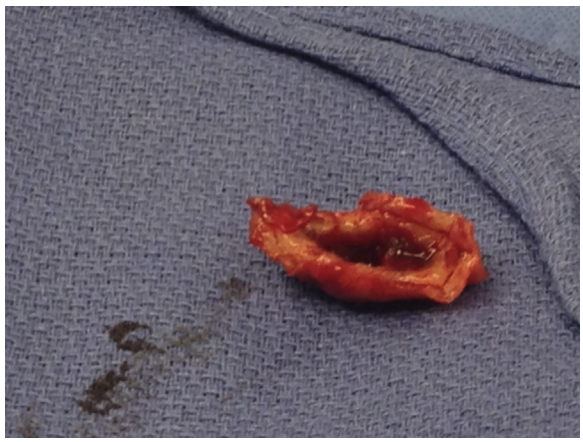

Fig. 4. Resected ulcerated endofibrotic plaque.

The plaque was sent for pathology with sections. Histology of the specimen showed ''thickening and fibroplasia of the intima. There is an overlying thrombus consisting of fibrin with scattered macrophages and lymphoplasmacytic inflammation but without typical atherosclerotic findings.'' From pathology report: ''Interpretation is that of arterial endofibrosis in the context of a 43-year-old man who is a competitive cyclist.''

The patient returned to clinic 6 months postoperatively. He had returned to his full complement of physical activity, including cycling, running, and swimming, without thigh claudication. He has some residual cramping in the anterior compartment of his left leg with strenuous exercise. He continues to take aspirin and cilostazol, which he was prescribed postoperatively. He will follow-up again at 1 year, postoperatively, and then annually with a duplex of both femoral arteries.