

Bilateral external iliac artery thrombosis due to endofibrosis in a 33-year-old female triathlete

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ABSTRACT

External iliac artery endofibrosis is a rare disease predominantly affecting young, elite male athletes. This case involves a 33-year-old female triathlete who presented initially with lower extremity claudication during training. After completing a triathlon 1 year later, the patient experienced acute-onset pain in both legs. Computed tomography angiography showed abrupt occlusion of the bilateral proximal external iliac arteries. The patient underwent a right and left external iliac artery reconstruction using the ipsilateral great saphenous vein and reported significant improvement of claudication symptoms. The case highlights a rare complication of acute bilateral arterial thrombosis. (*J Vasc Surg Cases and Innovative Techniques* 2019;5:58-60.)

Keywords: Iliac artery endofibrosis; Bilateral thrombosis; Athletes

External iliac artery endofibrosis (EIAE) is a rare disease seen primarily in young, otherwise healthy endurance athletes. The disorder is characterized by fibrosis and hypertrophy of the intimal layer of the artery wall. The cause of EIAE has been postulated to be trauma from “kinking” during repetitive hip flexion, psoas muscle hypertrophy compressing the artery, excessive and tortuous vessel length, and increased cardiac output with adaptive hypertension.¹ We describe a rare case of acute bilateral external iliac thrombosis in a competitive triathlete. The patient has consented to the publication of her case details and images.

CASE REPORT

A 33-year-old female competitive triathlete was initially referred for lower extremity claudication during exercise training. She had been averaging 7070 kilometers per year of training. She described worsening bilateral hip and buttock discomfort with training at near-maximum heart rates.

Initial rest ankle-brachial indexes (ABIs) were normal (ABI right/left, 1.0/1.0); however, stress ABIs were mildly abnormal (ABI right/left, 0.8/0.8) with relatively dampened waveforms at the

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high thigh compared with the rest pulsed volume recordings and ABIs. Computed tomography angiography did not show any evidence of intimal-medial thickening, stenosis, or excessive kinking or tortuosity of the external iliac arteries.

The patient was advised to change to a less rigorous training regimen. Angiography was deferred because the patient reported significant improvement in her symptoms at 1-month follow-up.

One year later and after finishing a triathlon, the patient experienced acute onset of pain in both her legs. The triathlon was in the heat and after a plane ride to the event, which may have contributed to precipitation of the pain. She presented urgently to our office, and her resting ABIs were severely abnormal (ABI right/left, 0.35/0.41). She was admitted to the hospital, and an intravenous heparin drip was initiated. Computed tomography angiography showed abrupt occlusion of the bilateral proximal external iliac arteries with reconstitution at the distal external iliac arteries with prominent collaterals from the internal iliac arteries (Fig 1).

Given the patient's desire to continue competing in triathlons, it was decided to pursue open surgical reconstruction because it would have the best long-term outcome for patency. Other surgical techniques, such as reconstruction with prosthetic conduit and endofibrosectomy with great saphenous vein (GSV) patch angioplasty, were considered as they have been described in the literature. However, complete external iliac artery resection with autogenous GSV interposition graft was thought to be the optimal operation for this patient.

A right external iliac artery reconstruction using ipsilateral GSV interposition graft was performed without complication (Figs 2 and 3). The patient was discharged at this point and prescribed apixaban and aspirin. One month later, a left external iliac artery reconstruction using the ipsilateral GSV interposition graft was performed without complication. She was discharged on aspirin only and remains on long-term aspirin therapy for anticoagulation. Preoperative duplex ultrasound examination and intraoperative findings showed that the bilateral autologous GSVs were of appropriate caliber (approximately 6 mm) in diameter.



Fig 1. *Left,* A three-dimensional reconstruction of the computed tomography angiography image showing occlusion of the left and right proximal external iliac arteries with reconstitution at the distal external iliac arteries, with prominent collaterals from the internal iliac arteries. *Right,* A three-dimensional reconstruction after bilateral intervention.

On pathologic examination, there were notable signs of bilateral inflammation and intimal thickening from the arterial segments. At 1-month follow-up, she noted no claudication, and her postoperative resting ABIs were normal (ABI right/left. 1.0/1.0). She was not cleared for full training activities at this appointment.

DISCUSSION

Iliac artery endofibrosis is a rare nonatherosclerotic vascular disorder that was first reported in competitive cyclists in the mid-1980s.^{2,3} Published data are limited to case reports, case series, and one systematic review.

Because this condition is mostly seen in cyclists, it is thought that the extreme hyperflexion at the hip joint causes repetitive stretching, elongation, and deformation of the iliac artery. Hypertrophy of the underlying psoas muscle may further exacerbate this trauma by way of compression and displacement to the vessel. Excessive and tortuous vessel length and fixation by adjacent fascia and collateral branches make the iliac artery immobile and vulnerable to kinking and increased mechanical stress. The pathophysiologic mechanism is also thought to be related to intra-arterial factors, such as high cardiac output causing shear stress and stimulating endothelial dysfunction.³

Clinically, patients usually complain of performance deterioration during maximal efforts associated with unilateral numbness, fatigue, or cramps affecting the thigh rather than the buttock or calf. Approximately 10% to 15% of the patients will have bilateral disease on presentation. In <5% of cases, intermittent claudication reflecting subacute ischemia will be manifested because of a complication such as localized dissection or arterial thrombosis.^{4,5} This case is unique in its presentation of bilateral acute thrombosis.

Reduced exercise ABIs recorded within 5 minutes after exercise in the recumbent position can identify endofibrotic flow limitation with up to 100% sensitivity and specificity.⁴ Abnormal Doppler waveforms and peak systolic velocities can be seen after maximal exercise or with provocative maneuvers such as hip flexion.^{4,6} The role of cross-sectional imaging and invasive angiography has become limited to cases in which there is diagnostic uncertainty or to better delineate the extent and location of the disease.⁷

The initial recommendation for EIAE is to reduce the time and intensity of training. Cyclists should be instructed to raise the handlebars or to move the saddle position forward to limit hip flexion. However, it may not be realistic to expect competitive cyclists

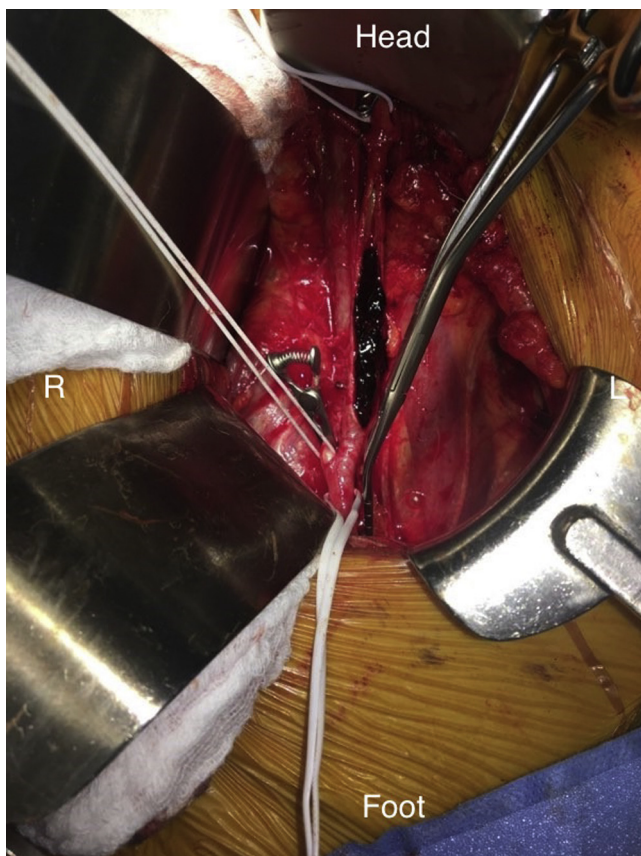


Fig 2. Intraoperative photograph demonstrating native right external iliac artery thrombosis. The procedure was an open thrombectomy performed through a longitudinal arteriotomy. Proximal control was obtained with circumferential vessel loop control of the common and internal iliac artery, and distal control was obtained with circumferential vessel loop control of the distal external iliac artery just above the inguinal ligament.



Fig 3. Complete right external iliac artery with transposition graft. The procedure and proximal and distal control are described in Fig 2. GSV, Great saphenous vein.

to embrace these alterations to their training regimen.⁴

For patients who will not adhere to a reduction in training or who develop debilitating symptoms, the best therapy is surgery. Angioplasty has not shown reliably good long-term outcomes. Endofibrotic lesions are elastic and are prone to recoiling and dissecting shortly after the procedure. Stenting is also not appropriate because the mechanical forces from cycling may cause stent fracture, migration, or plicature.⁸

Our patient had a unique presentation of bilateral acute arterial thrombosis with severely abnormal resting ABIs. To our knowledge, there has been only one other case report of bilateral acute arterial thrombosis. That patient was much older, and resting ABIs on presentation were only mildly to moderately abnormal.⁹

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