External iliac artery dissection secondary to endofibrosis in a cyclist

Thomas D. Willson, MD, Elizabeth Revesz, MD, Francis J. Podbielski, MD, and Matthew J. Blecha, MD, Chicago, Ill

Endofibrosis of the external iliac artery is an uncommon disease affecting primarily young, otherwise healthy, endurance athletes. Thigh pain during maximal exercise with quick resolution postexercise is characteristic of the so-called cyclist's iliac syndrome. We report an unusual case in which the typical endofibrotic plaque was accompanied by dissection of the external iliac artery. The patient was treated surgically with excision of the affected artery segment and placement of an interposition graft. This case highlights an unusual finding in association with external iliac artery endofibrosis and provides an opportunity to briefly review the literature on the subject. (J Vasc Surg 2010;52:219-21.)

Arterial endofibrosis is an uncommon arterio-occlusive condition that affects high-performance endurance athletes, cyclists in particular.¹⁻¹⁰ Patients typically present with rapid fatigue and thigh claudication, which resolves quickly postexercise. Most patients are found to have a simple focal stenosis secondary to fibrotic plaques within the external iliac artery. We are familiar with only two previous reports of arterial dissection in this syndrome. We describe the presentation, treatment, and pathologic findings of a 47-year-old man who presented with a symptomatic right external iliac artery dissection.

CASE REPORT

A 47-year-old man was referred, relating a several-week history of significant cramping and pain in his right thigh and right calf during exercise. He had no significant past medical history, was taking no medications, and related no history of trauma to his right leg. He was a lifelong non-smoker. He had no family or personal history of collagen vascular disease.

The patient, who is an avid cyclist, had initially noticed rapid muscle fatigue in his right leg when riding his bicycle. He also experienced more severe symptoms when attempting to run. He typically cycles more than 200 miles each week without difficulty, a routine he began around age 25.

On physical examination, common femoral pulses were palpable and strong bilaterally. There was no sign of ulceration or embolization in either lower extremity.

A resting Doppler examination revealed an ankle-brachial index (ABI) of .9 on the right and 1.05 on the left. Subsequent postexercise Doppler study revealed a severe drop in the right-side ABI from 1.17 to 0.34 at 1 minute, consistent with an

From the Saint Joseph Hospital/Resurrection Healthcare Department of Surgery.

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Reprint requests: Dr. Matthew J. Blecha, Resurrection Health Care - St. Joseph Hospital, Department of Surgery, 2900 N. Lake Shore Dr., Chicago, IL 60657 (e-mail: matthew.blecha@yahoo.com).

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occult occlusive arterial lesion. The ABI returned to normal at approximately 30 minutes postexercise. Computed tomography (CT) angiography (Fig 1) identified a dissection of the proximal to midportion of the right external iliac artery with arterial wall thickening and mild aneurysmal dilatation. No stenoses, aneurysms, or other dissections of the arterial system in either leg were found.

Based on the symptoms and the results of our workup, the patient was prepared for open repair of the dissection with an interposition graft. Patch angioplasty was not considered to be an option, given the presence of a frank dissection. Endoluminal stenting was not performed, given a high potential for restenosis or stent kinking in this anatomical location and given the patient's desire to return to cycling.

The right external iliac artery was exposed via an oblique abdominal flank incision. The arterial system was dissected out from 3 cm above the right iliac bifurcation down to the level of the inguinal ligament (Fig 2). A rind of firm, fibrous tissue was found encircling the external iliac artery. This segment had a slight aneurysmal dilatation and a dark discoloration. Interposition graft placement was performed using a 5.5-cm-length of ringed 8-mm PTFE. Prosthetic conduit was selected because the patient's saphenous veins were only 3 mm in diameter creating a significant size mismatch.

Pathologic examination revealed a 4.5-cm-long segment of artery with chronic inflammation, hematoma, and dissection. A dissection flap was readily visible within the specimen (Fig 3). The true lumen was noted to be smooth and yellow-tan with some surface irregularities. Histologic findings revealed a subendothelial plaque with abundant myofibroblasts and a disrupted internal elastic membrane. These findings correlate with previous reports of endofibrosis.⁴

The patient was discharged home on the third postoperative day. He was cleared to reinitiate cycling 16 weeks postoperatively. The patient is 9 months postsurgery, and he has been cycling for 6 months without a recurrence of symptoms or drop in postoperative ABI. He will be followed long term with serial history and ABI.

DISCUSSION

This case report details an external iliac dissection in a healthy 47-year-old cyclist. Dissection is a rare presentation

Fig 1. Computed tomography angiogram revealed a dissection of the proximal to mid-portion of the right external iliac artery with minimal aneurysmal dilatation.

of arterial endofibrosis, occurring in only 1% of patients in one series.¹ Arterial endofibrosis generally, while uncommon, is an important clinical entity affecting otherwise healthy cyclists and other endurance athletes.¹⁻¹⁰ Prompt recognition can lead to appropriate management and permit affected patients a quick return to their previous lifestyle.

The epidemiology of external iliac artery endofibrosis is not well described. It has been reported that 20% of top-level cyclists will develop sports-related flow limitations of the iliac arteries.³ Endofibrosis is most often described in cyclists, but cases have been reported in other groups of endurance athletes including triathletes, runners, cross-country skiers, rowers, and rugby players.¹⁻¹⁰ Patients do not necessarily need to be professionals. Chevalier has published two series in which amateur and recreational cyclists make up the vast majority of cases: 76% to 85% of affected cyclists.^{1,9}

Lesions are bilateral in 15% of patients at initial presentation.⁶ This patient had no evidence of left-sided disease by history, CT findings, or ABI. In previous case series,

patients who have had definitive (surgical) treatment have returned to their previous level of activity without recurrence of symptoms. There is a single case report of dissection occurring on the opposite side after a return to highlevel cycling after contralateral dissection in the absence of endofibrosis.¹¹

Endofibrosis development has been linked to both anatomic and mechanical factors. Studies of affected athletes have noted tethering of the external iliac artery (EIA) to the psoas muscle in 50% to 64% of cases. 1,3,4 Further, the middle portion of the external iliac artery is subject to both high shear forces and high blood flows during maximal exercise. A variety of etiologic factors have been suggested, including external compression due to psoas muscle hypertrophy, high-flow conditions due to increased cardiac output and adaptive systolic hypertension, and kinking/tortuosity. Lengthening of the EIA is noted in endofibrosis patients as the arteries to the psoas muscle in the central portion of the vessel create an immobile segment "working against" the relatively mobile adjacent segments of EIA. The subsequent excess length predisposes the artery to repetitive kinking during the pedaling process. 4 This results in repeated trauma to the arterial wall, which can cause an inflammatory reaction.²

Many diagnostic tests have been proposed, but proper validation has been difficult. Careful history with focus on the location and duration of claudication has been shown to be highly sensitive. An ABI of <0.54 has a specificity of 100% but was only 43% sensitive. In patients with unilateral complaints, an ankle pressure difference of 23 mm Hg was 73% sensitive and 95% specific.⁵ Duplex examination detects both stenosis and elongation of iliac arteries reliably but is user-dependent. ^{2,7,8} CT angiography is highly sensitive in detecting stenosis, dissection, or arterial wall thickening.

Surgery is the primary treatment in individuals who want to continue their sporting lifestyle.⁶ The most common method of repair is endofibrosectomy with patch angioplasty. In more complex cases, such as this one with dissection, reconstruction with either autogenous or prosthetic interposition graft has been documented to have excellent results, with 90% primary patency and 99% return to sport, including return to high-level competition.^{1,9} Transluminal angioplasty and stenting techniques have both been proposed but are not generally considered appropriate for treating endofibrotic lesions of this sort. Unlike atherosclerotic lesions, these lesions are elastic and tend to re-expand or dissect within days of angioplasty. 2,4,7,8,10 A stent placed in the external iliac artery would be subject to the same forces as the artery itself if the patient returned to his previous level of activity. Potential fracture, plicature, or migration of the stent were all concerns. Long-term stent patency in an artery still tethered to the psoas muscle in a patient planning to return to cycling would be tenuous.

External iliac artery endofibrosis is an uncommon diagnosis but one that must be considered in young, healthy



Fig 2. A rind of firm, fibrous tissue was found encircling the external iliac artery. This segment had a slight aneurysmal dilatation and a dark discoloration.

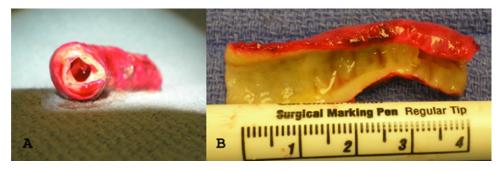


Fig 3. Dissection flap is readily visible within the excised segment of diseased external iliac artery.

endurance athletes with complaints of claudication. History and physical examination in conjunction with pre- and postexercise ABI and CT imaging can help establish the diagnosis. Once diagnosed, surgical reconstruction is the preferred method of treatment.

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