[Primary Care]

An Unusual Case of Leg Pain in a Competitive Cyclist: A Case Report and Review of the Literature

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Cycling has become a popular recreational and competitive sport. The number of people participating in the sport is gradually increasing. Despite being a noncontact, low-impact sport, as many as 85% of athletes engaged in the sport will suffer from an overuse injury, with the lower limbs comprising the majority of these injuries. Up to 20% of all lower extremity overuse injuries in competitive cyclists are of a vascular source. A 39-year-old competitive cyclist had a 5-year history of thigh pain during cycling, preventing him from competing. The patient was eventually diagnosed with external iliac artery endofibrosis. After conservative treatment failed, the patient underwent corrective vascular surgery with complete resolution of his symptoms and return to competitive cycling by 1 year. Since its first description in 1985, there have been more than 60 articles addressing external iliac artery endofibrosis pathology.

Keywords: cycling; endofibrosis; pain; leg

A 39-year-old male competitive cyclist presented with a 5-year history of left thigh pain during maximal and submaximal insertions. The patient has been training for 15 years, averaging 800 hours of training per year, and was the previous national elite road champion and national marathon mountain bike champion.

The patient reported a burning sensation in the left thigh muscles starting at the vastus medialis and spreading to all the quadriceps muscles and the gastrocnemius muscle. When the pain occurs, he is forced to stop pedaling for a few moments, after which he can resume riding at a slower pace.

He had no medical history of smoking, diabetes, high blood pressure, hypercholesterolemia, or coronary heart disease, and he was not taking any medications.

Physical examination revealed normal blood pressure, and pulses were palpated at all spots, with equal and good sensation in both legs. Hip and knee range of motion were normal, and provocative tests were negative for impingement, meniscal injury, or instability. Circumference of the left thigh had been reduced by 2 cm as compared with the right (Figure 1).

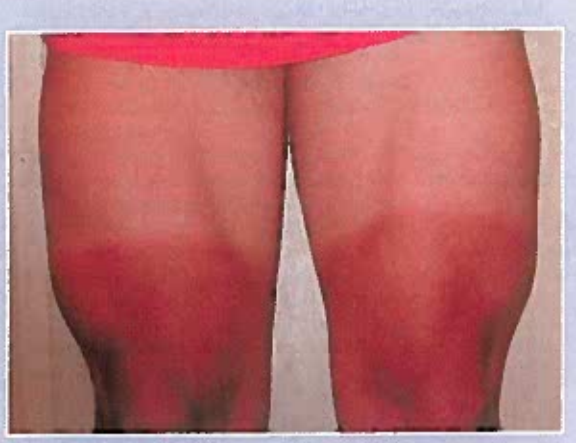

Figure 1. At physical examination, circumference of the left thigh had been reduced by 2 cm as compared with the right.

Blood examinations, including a complete blood count, chemistry panel, prothrombin time, partial thromboplastin time, and international normalized ratio, were normal.

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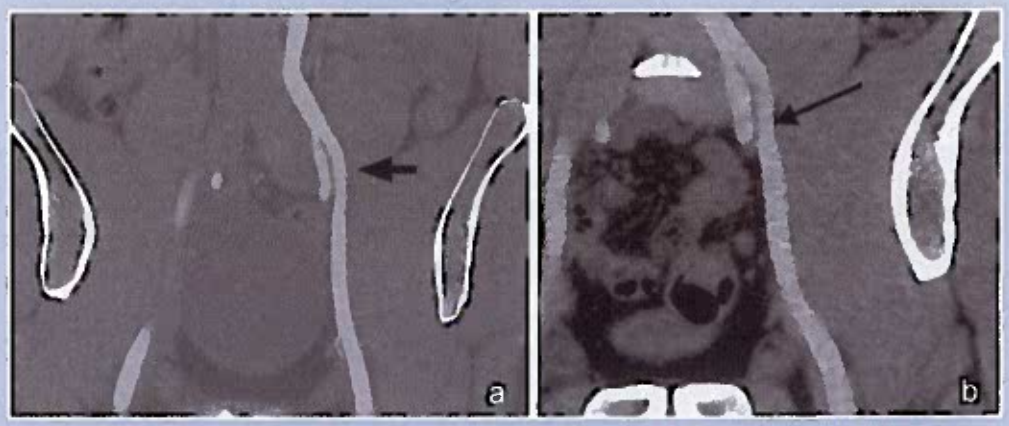

Figure 2. Computed tomography angiography. (a) No exertion: 28% stenosis in the left external iliac artery. (b) After exercise: 45% stenosis in the left external iliac artery.

Pelvic and lower extremities showed no abnormalities, including groin ultrasound and magnetic resonance imaging of the lumbar spine, pelvis, and hips. Computed tomography (CT) angiography demonstrated 28% stenosis in the left external iliac artery, and after exercise, demonstrated 45% stenosis at the same place (Figures 2a and 2b). A diagnosis of external iliac artery endofibrosis (EIAE) was established.

The patient underwent extensive conservative treatment, which included physical therapy, changing of cycling position, training, and antiatherosclerosis medication. However, he was not able to compete at the highest level. The patient no longer wished to continue conservative treatment; therefore, he was referred to a vascular surgeon specializing in this pathology.

Under general anesthesia, the diseased artery was approached, the endofibrotic lesion was removed, and the artery was shortened by 1.5 cm (Figures 3a-3c). The artery was closed using an autologous vein patch (Figure 3d). Following surgery, the patient was instructed to refrain from physical exertion for 6 weeks, after which he started gradual return to sport over an additional 6 weeks. By 3 months postoperatively, he was training on the bike, and by 6 months, he regained his presurgery condition and was pain free during maximal exertions. Eighteen months following surgery, he remains pain free.

DISCUSSION

In 1985, the first description of EIAE was presented with regard to 2 competitive cyclists.[34] Indeed, it is most prevalent in cyclists, professional and recreational; however, it has been described in other sports such as running, speed skating, triathlon, tennis, soccer, and more.[6,8,15,17,18,20] In cycling, there is a positive correlation between both the distance and intensity of cycling and the development of EIAE.[4,12] The actual prevalence of this condition is hard to evaluate, but in professional cyclists, vascular injuries have been estimated to account for as much as 20% of all overuse lower extremity injuries.[29] More than 85% of the patients are men[27]; however, research might be biased because of the high prevalence of men participating in endurance sports. The condition is bilateral in 15% of the cases,[13] and there is a left iliac artery predominance.[1,2]

Endofibrosis is a pathologic process in which progressive stenosis of the artery lumen is caused through thickening of the arteries intima. Histologic endofibrosis is a different entity compared with atherosclerosis and fibromuscular dysplasia.[14,34,35] Endofibrosis lesions have an accumulation of subendothelial loose connective tissue containing collagen, elastin, and smooth muscle, whereas in arthrosclerosis, there are densely packed collagen fibers and widespread calcifications. The pathologic process usually does not involve the arterial endothelium. The lesion may be complicated by overlying thrombus, dissection, or atherosclerosis.[11,19,22,36] The most common sight of pathology is the external iliac artery, which is affected in more than 90% of cases.[9,22,23] However, there have been reports of involvement of the common iliac, common femoral, and profunda femoris arteries as well.[32]

There are several local and systemic factors that contribute to the development of EIAE:

1. Posture: The aerodynamic position in cycling and skating requires hip hyperflexion, which may cause repetitive stretching and deformation of the iliac arteries.[7]
2. Vessel length: Abnormally long iliac vessels increase the likelihood of kinking during hip flexion. Kinking might be a predisposing factor for endofibrosis, but it in itself is a cause of decreased blood flow without endofibrosis.[27,28]
3. Arterial fixation: The external iliac artery might be immobilized by surrounding soft tissue or collateral branches to the iliopsoas muscle. The fixation at one point might increase the probability of the artery to kink and causes tethering of the central portion of the artery.[21]
4. Psoas hypertrophy: A hypertrophied psoas muscle increases the stretching of the external iliac artery during hip flexion[7] and increases mechanical stress, which may lead to

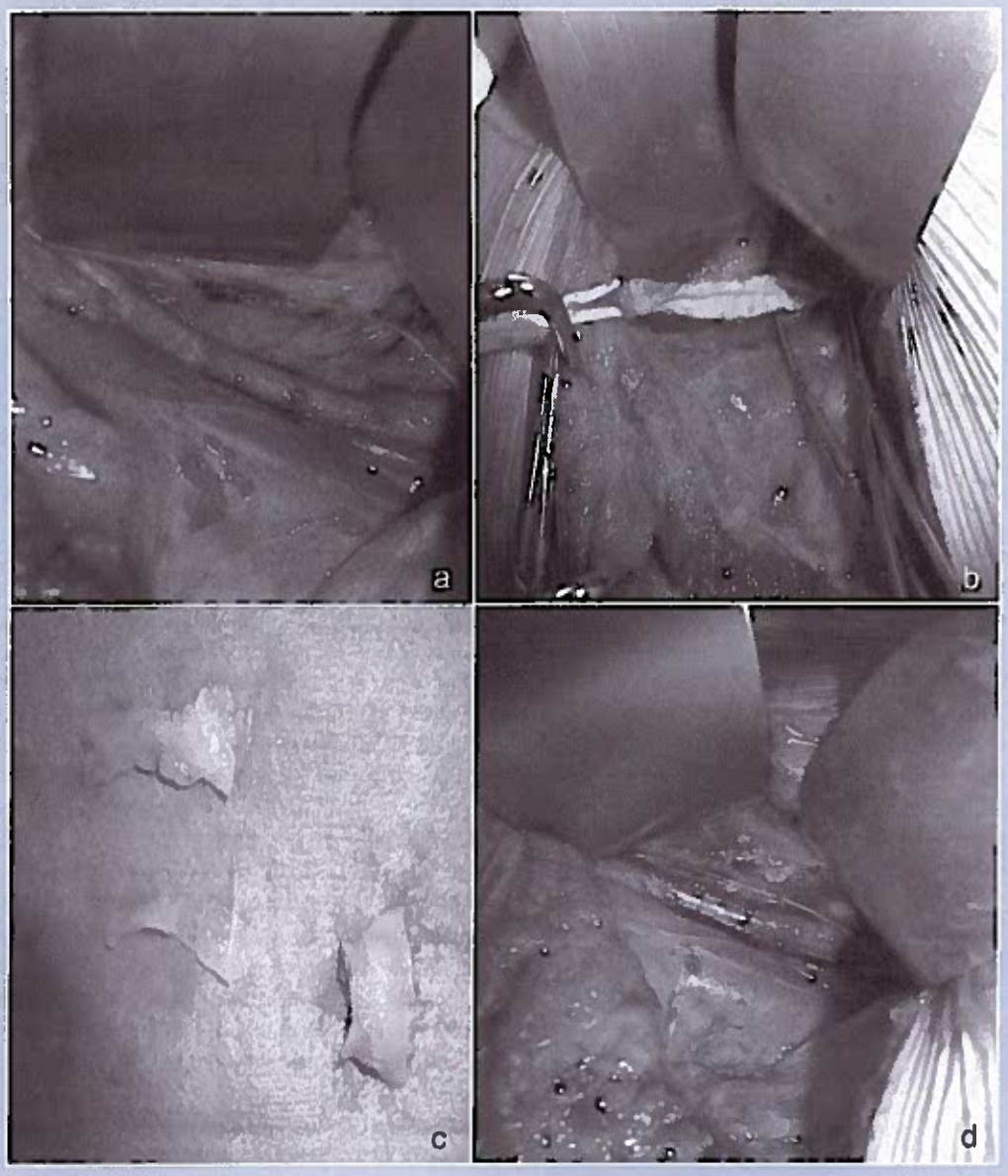

Figure 3. Intraoperative images. (a) Exposure of the diseased left external iliac artery; (b) opening of the diseased artery; (c) endofibrotic lesions removed from the left external iliac artery; and (d) repaired artery with autologous vein patch.

endofibrosis. In addition to creating endofibrosis, hypertrophy of the psoas exacerbates kinking of the external iliac artery.

5. Systemic factors: The role of systemic factors in the development of EIAE has not been investigated extensively. Feugier and Chevalier[12] found that 75% of their patients who had undergone surgical intervention for symptomatic EIEA had abnormalities in methionine metabolism. Although this presents a weak connection, it warrants further investigation with regard to the role systemic factors play in the development of EIAE.

Clinical Presentation

Patients are usually asymptomatic while resting or in submaximal exertions.[16] At maximal or near maximal exercise, symptoms will start to appear. They include muscle cramping, parasthesia, weakness, numbness, swelling of the thigh, and an unexplained deterioration in the athlete's abilities.[11,13,15,31] The pain usually radiates to the thigh but might also radiate to the calf and buttock. The symptoms usually resolve on cessation of maximal exertion, but as the disease progresses, they might appear with submaximal exertion as well.

Because of the unspecific nature of the clinical presentation, diagnosis is usually delayed between 12 and 41 months.[20] Physical examination is usually unremarkable, peripheral pulses are intact, and capillary refill is not elongated. In 44% of patients, there is an audible bruit over the femoral artery,[1,13,15] which might be better detected after exercise or with the hip flexed. Combining the clinical history with a specific vascular questionnaire, the finding of an audible femoral bruit and normal back mobility facilitated identification of a vascular cause in 79% of cases.[20,29]

To improve the accuracy of the clinical history and physical examination, provocative tests are added, of which the ankle-brachial pressure index (ABPI) is most important. ABPI might give normal results when tested at rest or by standard protocols; however, using bicycle-specific protocols allows identification of up to 85% of patients with flow limitations.[5,30]

The next step toward definite diagnosis is imaging. The best standard for assessing vascular obstruction due to atherosclerosis is static digital subtraction angiography; however, it has not been evaluated in the diagnosis of endofibrotic lesions. The first imaging modality used is Doppler ultrasound. The test should be performed in hip flexion, isometric contraction of the hip flexors, or after exercise. In this manner, the test will detect flow limitations in up to 85% of cases,[3,4,25] and combined with clinical history, physical examination, and ABPI will reach an accuracy of more than 90%.[24] Doppler allows measurement of peak systolic velocity, which is sensitive for endofibrotic stenosis. Doppler is a noninvasive, cost-effective way to assess for endofibrotic lesions; however, it is highly operator dependent, and high sensitivity has been found in patients presenting with severe disease. Because of these limitations, additional imaging tests are recommended, such as CT angiography or static magnetic resonance angiography (MRA). CT angiography allows for 3-dimensional reconstructions; however, it exposes the patient to additional radiation without offering advantage over MRA combined with Doppler.[32,36] MRA allows for assessment of vessel length, stenosis, and kinking. The test should be performed with the hips flexed since the majority of cases do not kink unless in hip flexion. Despite all the advantages of MRA, it should be performed in addition to Doppler because movement artifact might lower the MRA sensitivity.

Treatment

Conservative treatment should be attempted prior to surgical intervention. This should include positional changing, which avoids both extreme hip flexion and maximal exertions. In cases of recreational athletes, when conservative treatment does not resolve the symptoms, they should be encouraged to change their sport. Competitive cyclists should be advised to raise the handlebar and bring the saddle forward. In doing so, they are reducing hip flexion; however, changing cycling position might not be feasible for professional cyclists.

A connection between the development of endofibrosis and atherosclerosis has not been found; however, it has been implied that continued stenosis of the artery might lead to development of arthrosclerosis later in life.[22] Therefore, risk factors should be assessed, such as lipid and cholesterol profile, smoking, and a family history of atherosclerosis, and if found, should be addressed.

Surgical intervention is warranted whenever a patient who has demonstrated clear arterial pathology accounting for his or her symptoms has failed conservative treatment and is unwilling or unable to change his or her sport. Open vascular surgery is currently the preferred surgical option. Several reports have shown endovascular techniques to be unsuitable for this pathology[14,35] because of high rates of recurrence, stent placement leading to intimal hyperplasia,[26] and dissection of the endofibrotic segment.[10,12]

Surgical treatment allows many athletes to return to high-level competition; however, there are no studies, to our knowledge, comparing the different techniques and their long-term effects.

CONCLUSION

External iliac artery endofibrosis is an underrecognized cause of leg pain in endurance athletes. Because of the nature of activity and pain, the athlete will usually seek medical attention from an orthopaedic sports physician first. Therefore, awareness and understanding of this pathology is crucial to facilitate timely and patient-specific care.

REFERENCES

1. Abraham P, Bickert S, Vielle B, Chevalier JM, Saumet JL. Pressure measurements at rest and after heavy exercise to detect moderate arterial lesions in athletes. *J Vasc Surg*. 2001;33:721-727.
2. Abraham P, Bouye P, Quere I, Chevalier JM, Saumet JL. Past, present and future of arterial endofibrosis in athletes: a point of view. *Sports Med*. 2004;34:419-425.
3. Abraham P, Leftheriotis G, Bourre Y, Chevalier JM, Saumet JL. Echography of external iliac artery endofibrosis in cyclists. *Am J Sports Med*. 1993;21:861-863.
4. Alimi YS, Accrocca F, Barthelemy P, Hartung O, Duluc M, Boufi M. Comparison between duplex scanning and angiographic findings in the evaluation of functional iliac obstruction in top endurance athletes. *Eur J Vasc Endovasc Surg*. 2004;28:513-519.
5. Arko FR, Harris EJ, Zarins CK, Olcott C 4th. Vascular complications in high-performance athletes. *J Vasc Surg*. 2001;33:935-942.
6. Bender MH, Schep G, Bouts SW, Backx FJ, Moll FL. Endurance athletes with intermittent claudication caused by iliac artery stenosis treated by endarterectomy with vein patch—short- and mid-term results. *Eur J Vasc Endovasc Surg*. 2012;43:472-477.
7. Bender MH, Schep G, de Vries WR, Hoogeveen AR, Wijn PF. Sports-related flow limitations in the iliac arteries in endurance athletes: aetiology, diagnosis, treatment and future developments. *Sports Med*. 2004;34:427-442.
8. Bray AE, Lewis WA. Intermittent claudication in a professional rugby player. *J Vasc Surg*. 1992;15:664-668.
9. Chevalier JM, Enon B, Walder J, et al. Endofibrosis of the external iliac artery in bicycle racers: an unrecognized pathological state. *Ann Vasc Surg*. 1986;1:297-303.
10. Cook PS, Erdoes LS, Selzer PM, Rivera FJ, Palmaz JC. Dissection of the external iliac artery in highly trained athletes. *J Vasc Surg*. 1995;22:173-177.
11. Del Gallo G, Plissonnier D, Planet M, Peillon C, Testart J, Watelet J. Dissecting aneurysm of the external iliac artery. An unusual course of endofibrosis in an athlete [in French]. *J Mal Vasc*. 1996;21:95-97.
12. Fetiger P, Chevalier JM. Endofibrosis of the iliac arteries: an underestimated problem. *Acta Chir Belg*. 2004;104:635-640.
13. Ford SJ, Rehman A, Bradbury AW. External iliac endofibrosis in endurance athletes: a novel case in an endurance runner and a review of the literature. *Eur J Vasc Endovasc Surg*. 2003;26:629-634.

14. Giannoukas AD, Berczi V, Anoop U, Cleveland TJ, Beard JD, Gaines PA. Endofibrosis of iliac arteries in high-performance athletes: diagnostic approach and minimally invasive endovascular treatment. *Cardiovasc Intervent Radiol.* 2006;29:866-869.
15. Kral CA, Han DC, Edwards WD, Spittell PC, Tazelaar HD, Cherry KJ Jr. Obstructive external iliac arteriopathy in avid bicyclists: new and variable histopathologic features in four women. *J Vasc Surg.* 2002;36:565-570.
16. Lim CS, Gohel MS, Shepherd AC, Davies AH. Iliac artery compression in cyclists: mechanisms, diagnosis and treatment. *Eur J Vasc Endovasc Surg.* 2009;38:180-186.
17. Maree AO, Ashetqul Islam M, Snuderl M, et al. External iliac artery endofibrosis in an amateur runner: hemodynamic, angiographic, histopathological evaluation and percutaneous revascularization. *Vasc Med.* 2007;12:203-206.
18. Masmoudi S, Frikha I, Hadjkacem A, et al. Clinical case of the month. Iliac artery endofibrosis in a soccer player [in French]. *Rev Med Liege.* 2002;57:135-137.
19. Nakamura KM, Skeik N, Shepherd RF, Wennberg PW. External iliac vein thrombosis in an athletic cyclist with a history of external iliac artery endofibrosis and thrombosis. *Vasc Endovascular Surg.* 2011;45:761-764.
20. Peach G, Schep G, Palfreeman R, Beard JD, Thompson MM, Hinchliffe RJ. Endofibrosis and kinking of the iliac arteries in athletes: a systematic review. *Eur J Vasc Endovasc Surg.* 2012;43:208-217.
21. Pillet J, Chevalier JM, Rasomanana D, et al. The principal artery of the psoas major muscle. *Surg Radiol Anat.* 1989;11:33-36.
22. Rousselet MC, Saint-Andre JP, L'Hoste P, Enon B, Megret A, Chevalier JM. Stenotic intimal thickening of the external iliac artery in competition cyclists. *Hum Pathol.* 1990;21:524-529.
23. Scavee V, Stainier L, Deltombe T, et al. External iliac artery endofibrosis: a new possible predisposing factor. *J Vasc Surg.* 2003;38:180-182.
24. Schep G, Bender MH, Kaandorp D, Hammacher E, de Vries WR. Flow limitations in the iliac arteries in endurance athletes. Current knowledge and directions for the future. *Int J Sports Med.* 1999;20:421-428.
25. Schep G, Bender MH, Schmikli SL, Wijn PF. Color Doppler used to detect kinking and intravascular lesions in the iliac arteries in endurance athletes with claudication. *Eur J Ultrasound.* 2001;14:129-140.
26. Schep G, Bender MH, van de Tempel G, Wijn PF, de Vries WR, Eikelboom BC. Detection and treatment of claudication due to functional iliac obstruction in top endurance athletes: a prospective study. *Lancet.* 2002;359:466-473.
27. Schep G, Kaandorp DW, Bender MH, et al. Excessive length of iliac arteries in athletes with flow limitations measured by magnetic resonance angiography. *Med Sci Sports Exerc.* 2002;34:385-393.
28. Schep G, Kaandorp DW, Bender MH, Weerdenburg H, van Engeland S, Wijn PF. Magnetic resonance angiography used to detect kinking in the iliac arteries in endurance athletes with claudication. *Physiol Meas.* 2001;22:475-487.
29. Schep G, Schmikli SL, Bender MH, Mosterd WL, Hammacher ER, Wijn PF. Recognising vascular causes of leg complaints in endurance athletes. Part I: validation of a decision algorithm. *Int J Sports Med.* 2002;23:313-321.
30. Taylor AJ, George KP. Ankle to brachial pressure index in normal subjects and trained cyclists with exercise-induced leg pain. *Med Sci Sports Exerc.* 2001;33:1862-1867.
31. Taylor AJ, Tennant WG, Batt ME, Wallace WA. Traumatic occlusion of the external iliac artery in a racing cyclist: a cause of ill defined leg pain. *Br J Sports Med.* 1997;31:155-156.
32. Venstermans C, Gielen JL, Salgado R, Bouquillon P, Lauwers J. Endofibrosis of the external iliac artery. *JBR-BTR.* 2009;92:184-185.
33. Vink A, Bender MH, Schep G, et al. Histopathological comparison between endofibrosis of the high-performance cyclist and atherosclerosis in the external iliac artery. *J Vasc Surg.* 2008;48:1458-1463.
34. Walder J, Mosimann F, Van Melle G, Mosimann R. Iliac endofibrosis in 2 cycling racers [in French]. *Helv Chir Acta.* 1985;51:793-795.
35. Wijesinghe LD, Coughlin PA, Robertson I, Kessel D, Kent PJ, Kester RC. Cyclist's iliac syndrome: temporary relief by balloon angioplasty. *Br J Sports Med.* 2001;35:70-71.
36. Willson TD, Revesz E, Podbielski FJ, Blecha MJ. External iliac artery dissection secondary to endofibrosis in a cyclist. *J Vasc Surg.* 2010;52:219-221.