SCIENTIFIC LETTER

EXERCISE-INDUCED ILIAC ARTERY FIBROSIS

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ABSTRACT

Exercise-induced iliac endofibrosis (IE), initially known as cyclist iliac endofibrosis, is a low prevalence disease that presents characteristic symptoms of ischemia in patients without classical risk factors. We present a 45-year-old triathlete female patient. She consulted for intermittent claudication at 100 m in the lower right limb for about one year. She presented to the consultation with right external iliac artery thrombosis. Endarterectomy and plastic with vein patch were performed with favorable evolution. Iliac endofibrosis is a rare disease but it is one of the diagnoses to consider when dealing with young athlete patients with symptoms of lower limb ischemia.

Keywords: *Iliac endofibrosis, acute ischemia, exercise-induced endofibrosis, ciclyst endofibrosis*

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INTRODUCTION

Cases of young patients with no cardiovascular risk factors and clinical symptoms of arterial ischemia, be it acute or chronic, are rather unusual and frequently lead to a delayed, difficult and inappropriate diagnosis. In particular this is due to the fact that symptoms do not usually appear at rest or are not very severe, and all pulses may be palpable. These patients tend to be evaluated by different specialists and undergo multiple studies and physical therapy. Some of the pathologies to be ruled out (without limitation) are popliteal trapping, chronic compartment syndrome, adventitial cystic disease and other less frequent presentations such as peroneal, pedal or tibial trapping.

In spite of this, the diagnosis becomes a firm suspicion when taking note that the patient is a high-endurance athlete with many years of practice and many kilometers of cycling or long-distance running.

CLINICAL CASE

A female, 45-year old patient who complains of intermittent claudication at 100 meters in the right lower limb but regularly exercises in the bicycle without difficulty. The patient reports to have experienced an episode of pain in that limb during a road cycling session that was relieved in time and with analgesics. The physical examination revealed absence of pulses on the right side and all pulses present with good amplitude in the left lower limb. The right ABI was 0.3 at rest and the left ABI 1.

The Doppler ultrasound scan revealead thrombosis of the right iliac artery all along its extension, except for the first 3 cm of the proximal segment, where parietal thickening could be seen with significant stenosis and the common femoral was patent after the inferior epigastric and iliac circumflex arteries. The angioCT confirmed the diagnosis of iliac endofibrosis of the external iliac, complicated with local thrombosis (C0F5E0) of the Chevallier[1] classification and no signs of distal embolism could be observed (Figure 1). Not significant contralateral stenosis was also observed (probably inicipent endofibrosis).

It was decided to perform external iliac endarterectomy all along its extension and patch plastic thrombectomy of the homolateral saphenous vein through a transaponeurotic retroperitoneal approach (Figures 2 and 3). The pathological anatomy of the endarterectomy revealed fibrosis and chronic inflammation of the intima with thrombosis in organization. After 2 days of hospitalization the patient was discharged. She recovered all pulses after surgery, and the ABI in this limb at rest was 0.0. All flows measured by Doppler ultrasound were triphasic with preserved velocity.

During the first three months she could take walks without any inconvenience and as from the fourth month she reinitiated sports practice without claudication (she was recommended not to go back to cycling).

DISCUSSION

The first cases of cyclist iliac endofibrosis (exercise-induced) were described in the ´80s, which means that this pathology has been recently identified (probably associated to the cycling boom during

FIGURE 1. Stenosis and thrombosis of the external iliac artery

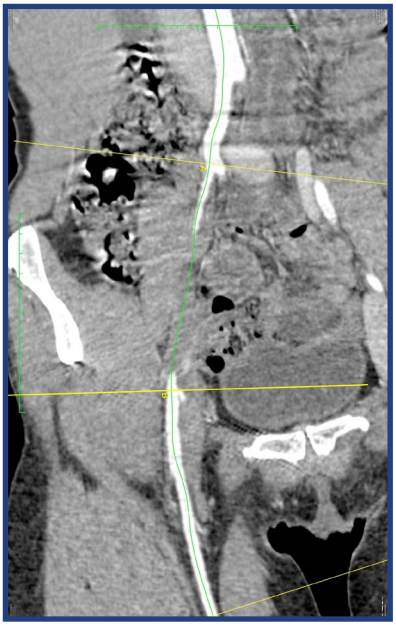


FIGURE 2. Intraoperatory image of iliac endofibrosis and distal thrombosis

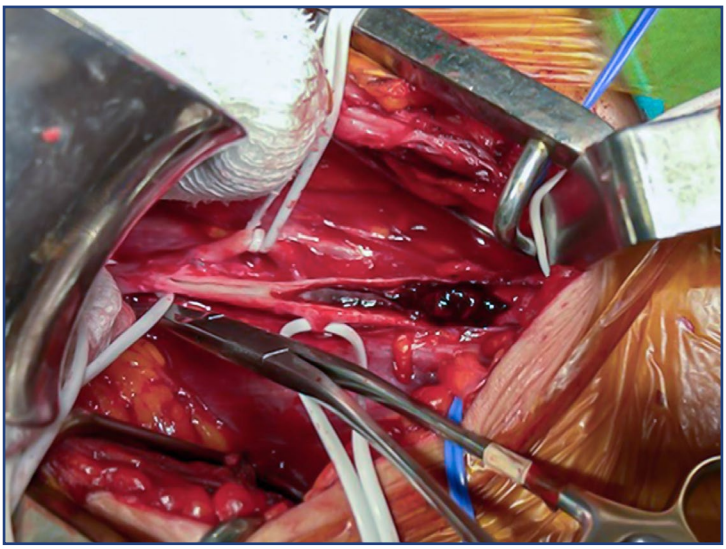
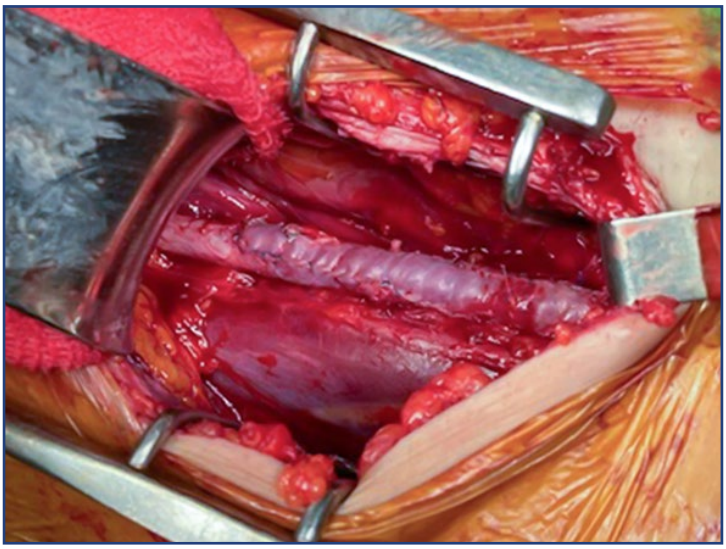


FIGURE 3. Saphenous vein patch



the ´70s and ´80s, and it is not surprising that it was first described in France, a country well-known for the practice of road cycling and in a cycling rehabilitation center)[1-3]. Although it is known as endofibrosis of the external iliac artery (94.9%), it should be considered that fibrosis may also present itself at the level of the common iliac artery (14.4%), the common femoral artery (0.5%) and the profound femoral artery (7.2%), in the latter particularly as stenosis in the form of ostial diaphragm[1]. This pathology has been described mainly in high-endurance long-term cyclists, but it has also been found in runners, triathletes, rowers, body builders and others. In the vast majority these are young patients with many years of high-endurance sports practice. Symptoms may be seen as from the first 5 years of initiation of sports and at 50.000 km run; however, those that require surgical treatment usually are 14 years and older and have run over 130,000 km[1].

This presentation is clearly a cause of non-atherosclerotic stenosis, but its pathophysiology and etiopathogenesis is still unknown. Subintimal thickening takes place with collagen, elastin and smooth muscle cells depositions; and in cases that get complicated dissection and/or localized thrombosis can be seen[4].

Its etiology is rather uncertain. It has been proposed that the shearing stress produced by hyperflow may stimulate fibrosis, which would lead to endothelial malfunction[5]. The cyclist´s position may produce kinking, with the consequent increase in flow velocity and parietal stress, apart from lengthening the external iliac artery, which would in turn increase the degree of kinking. It is not very clear whether the same factors may act in fixed or mobile segments of the arteries[1]. Psoas hypertrophy upon fixation of the external iliac artery by the feeding collaterals of the psoas has been mentioned as an anatomic reason that

might play a role[6]. Localized vasospasm has been identified in some cases, also of an unknown origin (perhaps due to the endothelium-induced arterial dilation)[7]. Vasospasm may be visualized by means of image studies (Doppler ultrasound, CT or MRI) pre and post exercise[7-10].

Suspected initial diagnosis is based on the history of high-endurance sports practice, but other differential diagnosis of non-atherosclerotic arterial disease must be ruled out in individuals without the classical risk factors. Patients frequently present all palpable pulses, thus the importance of performing the ABI at rest and post-exercise (until onset of symptoms). A drop greater than 0.3 is considered the cut-off value. Then, it is possible to continue the evaluation with image diagnostic methods. The Chevalier group usually performs diagnostic angiography and it is important to stress that this must include a lateral projection of the iliac-femoral axis in flexion to identify kinks, stenosis and extrinsic compressions. Based on the angiography, they propose a classification of the pathology assessing the status of the primitive, external, common femoral and profound arteries. They stress that the degree of stenosis is not determinant, since, as has been previously described, the problem has dynamic characteristics (vasospasm, kinking, extrinsic compression, lengthening of the external iliac artery) and they have also seen that the vast majority of treated patients present less than 50% stenosis[1].

As for the treatment, once the diagnosis is confirmed, cycling or the predisposing activity must be discontinued. This is one of the vascular diseases for which surgical treatment continues to be definitive and the first choice. Chevalier et al., since their initial description, suggested endofibrosectomy of the affected area, shortening of the external iliac, reimplantation and plastic with vein patch[1-3]. The same authors perform intraoperatory angioscopy to confirm the diagnosis, and even prefer this option over intravascular ultrasound. Peak et al. suggest the use of bovine pericardium patch so as to preserve the saphenous vein and avoid possible aneurismatic dilation in future[11]. In complicated cases with dissection or extensive thrombosis, iliofemoral bypass may be performed as a first attempt (with vein, Dacron prosthesis or ePTFE). After surgery, most authors agree that the patient must continue antiaggregation treatment with aspirin only (75-100 mg/day). Doppler ultrasound is performed after surgery to ensure patency and confirm that there are no major issues.

Mid-term results (50 m) in most groups is over 90% primary patency and 100% assisted/secondary patency. There are not many references regarding long-term results.

There are a few described cases of endovascular treatment using balloon and drug-coated balloon with unfavorable results that show early symptoms relapse[12,13].

It is suggested to reinitiate sports activity at 3 months, and of course not returning to cycling, although professional cyclists who have resumed have not required reintervention[1].

The consensus of the international group for the study and identification of endofibrosis (INSITE) was published in 2016, suggesting surgery as the treatment of choice (endofibrosectomy whenever possible and eventually bypass in case of extensive occlusions), diagnosis with ABI (to be complemented with image studies) and resuming sports activity at 6-8 weeks[14].

CONCLUSION

Exercise-induced iliac endofibrosis (IE), is a low prevalence vascular disease. It must be initially suspected in patients with typical symptoms of arterial claudication with a background of intense physical activity (particularly cycling) without typical cardiovascular risk factors. Surgical treatment has low morbidity since these are mostly healthy patients and the surgical intervention is not technically demanding. Mid-term results are also promising and provide absolute resolution of the symptoms in more than 90% of patients.

Conflicts of interest

The authors have no disclosures to declare.

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