

Arterial endofibrosis in professional cyclists

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SUMMARY: Arterial endofibrosis in professional cyclists.

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External Iliac Artery Endofibrosis (EIAE) is an uncommon disease usually affecting young, otherwise healthy, patients. It usually involves cyclists but cases have been reported in other groups of endurance athletes. The external iliac artery is the most affected anatomical site but other locations are described too. The precise pathophysiology and long-term evolution of the disease still remain unknown. The diagnosis may be challenging and delayed as the patients usually present symptoms only in extreme conditions and physical and instrumental exami-

nations may be normal at rest.

We present two cases of young professional cyclists who suffered of exercise-induced leg pain which led them to reduce running. Both patients were firstly treated with balloon angioplasty that rapidly failed to improve their symptoms. The successive open surgery with endofibrotomy and autologous saphenous vein closure patch completely resolved physical limitations.

EIAE is a rare disease that can induce arterial stenosis, thrombosis, dissection and secondary atheroma. After-exercise ankle-brachial index represents a useful diagnostic criterion. Careful observation of angio-CT may strengthen the suspect. Knowledge of these features allows a better pre-operative assessment and an early effective treatment. Surgical revascularization remains the gold standard approach.

KEY WORDS: External iliac artery endofibrosis - Peripheral arterial disease - Non atherosclerotic arterial disease.

Introduction

External Iliac Artery Endofibrosis (EIAE) is an uncommon and rare disease usually affecting primarily young, otherwise healthy, athletes. The first description of the disease was by Chevalier in 1986 in cyclists (1) but cases have been reported in other groups of endurance athletes including triathletes, runners, cross-country skiers, rowers and rugby players (2).

The external iliac artery is the most affected site but other locations have been reported (3-5). The precise pathophysiology and long-term evolution are unknown. The diagnosis may be difficult and often delayed

due to the absence of signs and symptoms at rest.

We present two cases of young professional cyclists who suffered of exercise-induced leg pain significantly reducing their physical performance and we discuss on this entity whose knowledge is crucial for correct diagnosis and treatment.

Case 1

A 23-year-old professional cyclist was referred for muscle fatigue with significant cramping and pain of his right leg when attempting to run. He was in optimal general conditions with a complete negative medical history.

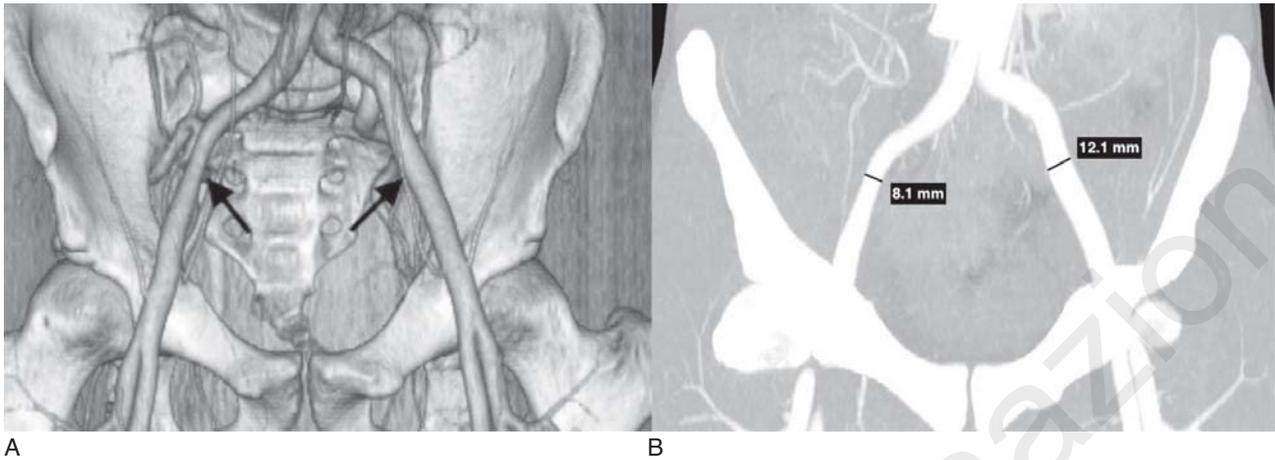
Clinical examination revealed that common femoral pulses were both palpable and strong; otherwise the distal pulses were clearly palpable only in the left leg. The skin was bilaterally warm without any pathological sign.

The ankle-brachial index (ABI) at rest and after exercise was performed. Resting-ABI was 1.02 on the right side and 1.04 on the left. After exercise right ABI drop to 0.61 while left ABI remained the same.

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A **B**
Fig. 1 A, B - Case 1: CT-angiography showing a minimal calibre reduction of the right external iliac artery compared to the left external iliac artery with no other evident lesions (A: 3D-lumen reconstruction, B: MIP-reconstruction with EIA diameter indicated).

The subsequent angio-CT documented a normal common iliac artery on the right; on the contrary, a minimal calibre reduction of the proximal tract of right external iliac artery (rEIA) was identified with no other significant radiological lesions (Figure 1).

An endovascular approach was attempted firstly: by means of a retrograde femoral access, an invasive pressure examination in right common iliac artery under and above the proximal segment of external iliac artery was obtained, without significant difference, even after papaverine intra-arterial injection. After the pressure study, we performed an angioplasty of the EIA with a 10-millimeter diameter non-compliant balloon for at least 3 minutes of inflation. Post-procedural period was regular.

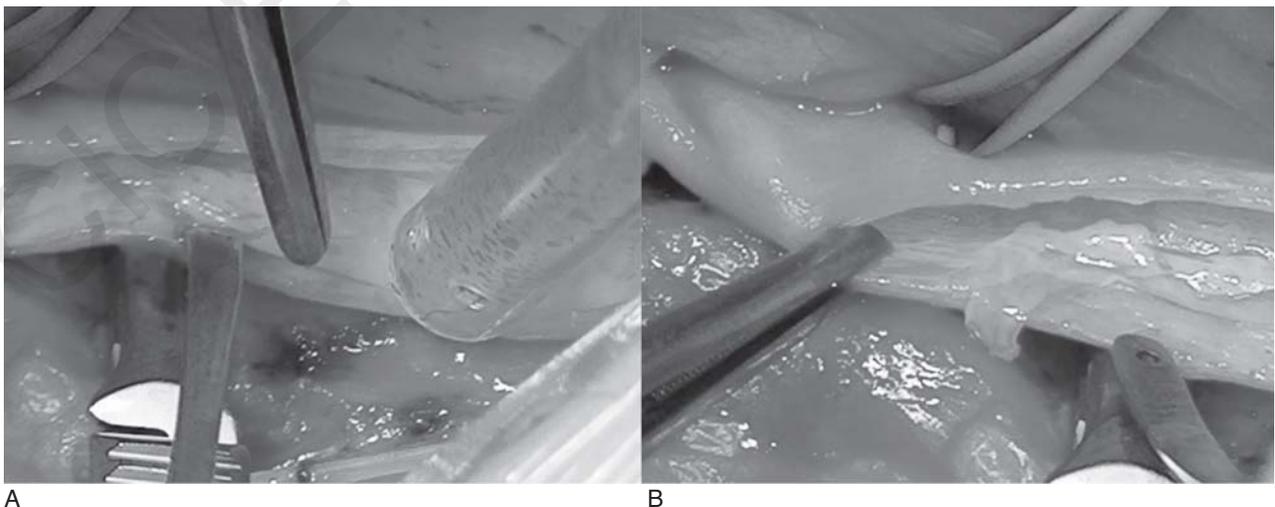
After discharge, the patient did not refer any clinical improvement and an open surgical repair was planned.

The right iliac vessels were exposed by mean of a right pararectal extra-peritoneal approach. Longitudinal arteriotomy of rEIA at site of lumen reduction revealed a significant endothelial hyperplasia. An endofibrosectomy (Figure 2) was performed and the artery was closed with an autologous great saphenous vein patch.

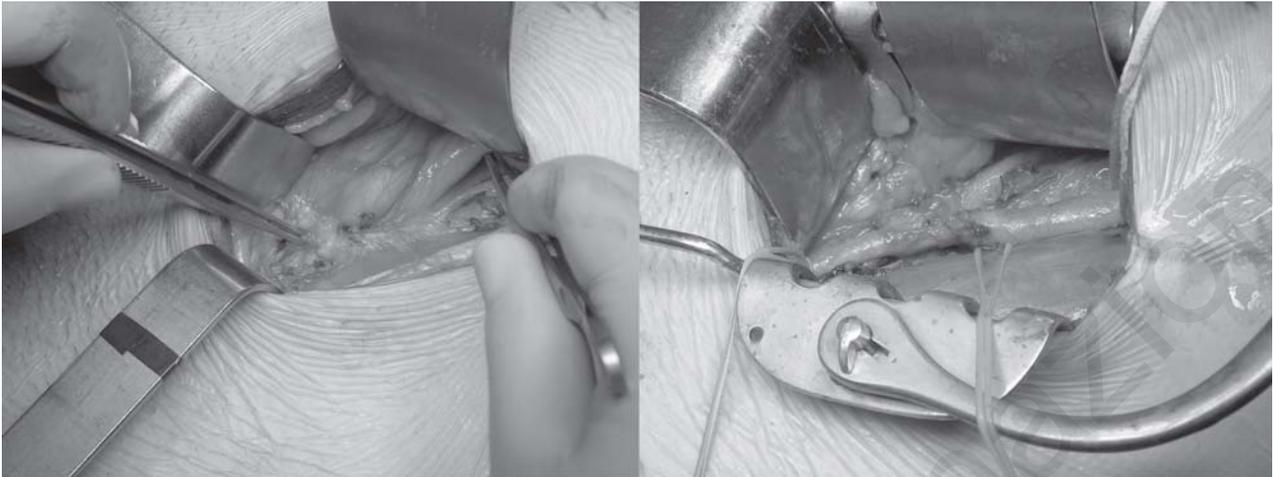
Histological analysis revealed marked fibrosis of the tunica media. The subsequent hospital stay was uneventful. The patient was discharged at fourth post-operative day.

Two months later the patient returned to ride the bike with no more symptoms and after 6 months he won a professional cyclist race.

One-year angio-CT follow up confirmed the regular patency of the rEIA lumen.



A **B**
Fig. 2 A and B - Case 1: endofibrosectomy of the origin of the right external iliac artery.



A
B
Fig. 3 A, B - A) Case 2: it is evident the narrowing of the origin of the external iliac artery due to the severe hypertrophy of the left psoas muscle. B) The left iliac vessels after complete separation from the psoas muscle.

Case 2

A 25-year-old professional cyclist presented a clinical history very similar to the case one.

The CT-scan revealed a narrowing at the origin of the left external iliac artery (IEIA), in contact to the left psoas muscle that appeared hypertrophic.

At another Institution, a balloon angioplasty was firstly attempted, followed by laparoscopic debridement of the IEIA from the left psoas muscle. After these two attempts, he presented at our Vascular Institution referring an immediate recurrence of disabling symptoms. The clinical examination at rest did not reveal any alteration with normal peripheral pulses. Anyway ABI examination before and

after exercise revealed a significant drop on left side (from 1 to 0.60). In consideration of previous failed miniminvasive approach, we managed for open revascularization.

The left iliac vessels were exposed by means of a left pararectal extra-peritoneal approach. Tough adhesions between the artery and the hypertrophic psoas muscle were identified, narrowing the origin of the external iliac artery (Figure 3 A). After lysis of the adhesions and a complete dissection of the iliac vessels from neighbouring tissue (Figure 3 B), an endofibrosectomy of the first tract of the IEIA and an autologous great saphenous vein closure patch was performed (Figure 4).

Even in this case, the histological examination revealed a severe fibrosis of the tunica media.



Fig. 4 - Case 2: autologous saphenous vein closure patch.

The postoperative course was uneventful and the patient was discharged at fifth post-operative day.

After the treatment, the patient's symptoms immediately disappeared and after two months he returned to ride a bike at professional levels.

Eight-month angio-CT showed restoration of the IEIA lumen with no vascular complications.

Discussion

External Iliac Artery Endofibrosis (EIAE), although uncommon, represents an important clinical entity affecting cyclists and other endurance athletes otherwise healthy. The prevalence of the disease is unknown but in professional cyclists it can be estimated to account up to 20% of all overuse leg injuries (3). EIAE has also been reported in other endurance sports such as long-runners, triathletes, speed skating and more (4-6). Although the external iliac artery is mainly affected, the common iliac and the common femoral arteries may also be involved (3-5).

Certainly different from the atherosclerosis, the aetiology of EIAE still remains unclear. In histological specimens there is a thickening of the vessel intima due to subendothelial accumulation of loose connective tissue containing variable amounts of collagen, elastin and smooth muscle cells, as shown also in our cases. Differently to the atherosclerosis mechanism, the collagen fibres are densely packed and calcification is typically widespread (7-9).

The pathophysiology has been investigated in the cyclist patients and some factors have been postulated. First of all, the position: in order to obtain the maximal aerodynamic benefit, they force an hyperflexion of the hip joint that may alter the iliac artery anatomy inducing a chronic friction and stimulating the endofibrosis (9). A significant hypertrophy of the psoas muscle may worsen the mechanical stress: the muscle pinches the artery, which is often fixated to it by the fascia and its collateral branches, provoking a continuous arterial traction and a subsequent endofibrosis (9, 10). This mechanism was particularly evident in our second case, where a thigh adhesion between artery and muscle was found.

The diagnosis may be difficult and delayed. Usually, symptoms are unilateral and they appear only at near-maximal exercise. In that situation, muscle cramp is the most frequent symptom followed by feeling of swelling, numbness or pain that develops in the calf, thigh or buttock of the affected side (5, 11, 12). At rest, physical examination is generally completely negative and peripheral pulses are present with absence of the traditional signs of arterial disease. Different dynamic instrumental investigations (i.e. ABI measurement, pulse volume recording and papaverine-assisted mean pressure gradient) have been proposed to help in the diagnosis. In our ex-

perience, the ABI measurement at rest and after exercise resulted positive (at least 0.4 of difference) and it clarified many diagnostic doubts. It probably represents one of the easier, cheaper and more effective methods to investigate this rare arterial disease, especially when associated to uncertain clinical or radiological finds (6-8). The predictive value of Duplex, in fact, remains low even in trained hands and a normal Duplex should not exclude the diagnosis. Angio-CT well reveals stenosis, dissection or arterial lumen narrowing but it requires careful attention because of arterial defects may be minimal, as in our first case. The role of MR-angiography is limited to those patients without endovascular lesions whose symptoms may be due to arterial kinking. Digital subtraction angiography provides anatomic details before surgical intervention and allows location of the stenosis and quantification of the excessive length of the artery (2, 13).

Surgery is the best treatment in individuals who want to continue their sporting lifestyle. The most common method of repair is endofibrossectomy with patch angioplasty (1). In more complex cases reconstruction with either autologous 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 (2).

Transluminal balloon angioplasty and stenting techniques have both been proposed but they are not considered appropriate for treating endofibrotic lesions. Unlike atherosclerotic stenosis, these lesions are elastic and tend to re-coil or dissect within days from the procedure (4). Moreover, a stent placed in the external iliac artery would be subject to the same forces and deterioration of the native artery if the patient return to the same level of activity. Potential fracture, plicature or migration of the stent were all concerns. Long-term outcome of stent angioplasty in such condition remains debatable (9).

Conclusions

EIAE is a rare disease which could give to the clinicians both diagnostic and therapeutic problems. Clinical history, physical examination in conjunction with pre- and post-exercise ABI and careful observation of angio-CT represent the fundamental criteria to establish the correct diagnosis. Once diagnosed, surgical endofibrossectomy with arterial patch closure is the best treatment for obtaining a definitive clinical benefit.

Conflict of interest

The Authors declare that there is not conflict of interest.

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