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Artery Endofibrosis

## CASE REPORT

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# Iliac Artery Endofibrosis in a Middle-Aged Female Long-Distance Runner

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## ABSTRACT

Exercise-induced iliac artery endofibrosis is a recently described abnormality of the external iliac artery that typically affects younger, healthy endurance athletes. Characteristic of the initially termed cyclist's iliac syndrome is lower limb pain during exercise with rapid recovery after exercise. This clinically complicated case describes an older female long-distance runner in whom an incorrect diagnosis of fibromuscular dysplasia was originally made when she presented with claudication and thrombosis of the right external iliac artery. A thrombectomy and bilateral balloon angioplasty were performed; however, her symptoms persisted. Four months later, she unexpectedly complained of dual calf claudication, a diagnosis of exercise-induced iliac artery endofibrosis was made, and a bilateral prosthetic graft bypass procedure was performed, which resulted in a good outcome.

Key Words: Arterial Endofibrosis, External Iliac Artery, Endurance Athlete, Exercise-Induced, Vascular Stenosis

**E**xercise-induced iliac artery endofibrosis (IAE) is a recently described symptomatic arterial flow restriction seen in young male and female endurance athletes of all disciplines. Publications in the 1980s reported highly trained cyclists complaining of lower leg pain during training.<sup>1,2</sup> Vascular problems in young healthy athletes are unusual and are easily overlooked, often leading to diagnostic delays as symptoms are commonly attributed to musculoskeletal or neurologic causes.<sup>3,4</sup>

Young athletes present with symptoms due to arterial flow limitation in the lower limb, which progressively worsen during exercise, disappearing when training is stopped.<sup>5</sup> Indicators may include vague lower limb pain, cramps, numbness, weakness, claudication, and unexplained performance deterioration.<sup>4,6</sup> Pain often affects the anterior thigh during physical performance exercises, radiating into the buttocks and calf.<sup>4</sup>

IAE commonly causes narrowing of the first few centimeters of the external iliac artery, with 10% of the cases affecting multiple artery sections.<sup>6</sup> The common iliac artery, femoral artery, or the profunda femoris artery may be involved.<sup>7</sup>

There seems to be a left iliac artery predominance<sup>3,5,6</sup>; however, in 15% of the cases, presentation is bilateral, and in rare cases, arterial dissection is founded.<sup>6</sup> Histologic findings indicate intimal and medial artery wall fibrosis, with no evidence of vasculitis or atherosclerosis.<sup>5</sup> Arterial kinking without the occurrence of internal narrowing can also occur.<sup>8</sup>

Most athletes with flow limitations in the iliac arteries seem to be young, highly trained athletes<sup>9,10</sup>; therefore an unexpected complex clinical case of IAE in a more mature runner is reported. After unsuccessful angioplasty procedures, she suddenly presented with dual calf claudication and responded well to a bilateral common iliac to common femoral prosthetic graft bypass procedure.

## CASE REPORT

A 53-yr-old woman, and avid long-distance runner for more than 15 yrs, presented with acute pain in the right leg. The pain was described as deep in the muscles of the posterior aspect of the right lower leg and, to a much lesser degree, the lower left leg. Being a passionate runner, she typically averaged 40 to 80 km a week in the years before the event and also completed ultramarathons and trail challenges. For the past 5 yrs, she had been aware of pain on the start of a run, which became so bad at 800 m that she had to stop and wait for it to subside, after which she could continue for 20 to 25 km. She typically finished a 42-km marathon in less than 4 hrs; however, her effort tolerance slowly deteriorated, and eventually, she was not able to finish the event. Two weeks before consultation, she participated in the Otter Challenge, a 42-km extreme trail run that is run in 1 day as opposed to the hike, which is done in 5 days. A sudden, marked pain in the right calf was experienced on any exertion, progressing to the upper leg. No pain was ever experienced during rest and symptoms had always been minimal on the left.

A lumbar magnetic resonance imaging scan before the first consultation identified no spinal canal stenosis or pressure effect on the existing nerve roots or obvious renal or pelvic abnormality. Sacroiliac and hip joints appeared morphologically normal.

On clinical examination, she was slender, with a height of 1.65 m and body weight of 52 kg. Her blood pressure was 120/75 mm Hg; heart rate, 76 bpm; and total blood cholesterol, 4.2 mmol/l. She had no known vascular risk factors, is a nonsmoker, takes no alcohol, uses no medication, and has no history of trauma to her right leg.

Resting duplex Doppler ultrasonography examination revealed the right external iliac artery totally occluded and filled with hypoechoic thrombus. The common iliac arteries, right internal iliac artery, and both left internal and external iliac arteries were patent with normal triphasic flow. The common femoral artery had poor residual flow of 23 cm/sec peak systolic velocity with posterior deviation seen particularly on the left side with hip flexing.

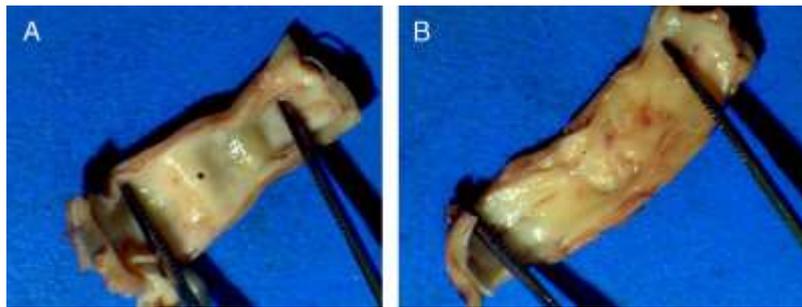
Vascular angiography computed tomography scan demonstrated thrombosis of the right external iliac artery, with a considerable network of collateral vessels indicating some chronicity. Irregularities in the left external iliac and common femoral arteries were noted. Fibromuscular dysplasia was diagnosed and a thrombectomy and bilateral balloon angioplasty of the external iliac arteries were performed via an open approach through the femoral arteries. Clopidogrel bisulfate and aspirin were prescribed.

Immediately after operation, mobility was limited and exercise tolerance could not be tested. For a 4-mo period, she became more active and then suddenly became aware of bilateral calf claudication, which was now worse on the left than the right. Although symptoms were limited to the calves on running, pain was experienced in both thighs when cycling.

Further consultation revealed audible bruits over the pelvic fossa area, louder on the left than the right. The left femoral pulse and both popliteal pulses were weakly palpable, but the ankle pulses were absent bilaterally. Reasonable biphasic waveforms presented at the right ankle but very blunted monophasic signals on the left. Resting ankle pressure of 90 mm Hg on each side, compared with the upper limb pressure of 130 mm Hg indicated a 40 mm Hg pressure gradient across the iliac artery. After minor exercise, flow was virtually undetectable on the left with a fall in the ankle pressure to 50 mm Hg on the right. Color duplex Doppler demonstrated a significant right iliac artery stenosis situated proximally, as well as a distal left external iliac and common femoral artery stenosis.

The picture was compatible with either fibromuscular dysplasia or external IAE described in athletes; however, the poor response to balloon dilation and the patient's athletic history, although older, favored the diagnosis of IAE.

Formal catheter-directed angiography via the left brachial artery confirmed extensive irregular stenosis of the entire right external iliac artery and multiple stenoses bilaterally. With 90-degree hip



**FIGURE 1** Microscopy of the left iliac artery vessel (40 × 11 × 2 mm) (A) and right iliac artery vessel (50 × 10 × 3 mm) (B). Intima shows focal areas of smooth raised nodules. The intimal hyperplasia changes are consistent to the intimal end-stage disease seen in popliteal artery entrapment syndrome.<sup>11</sup>

flexion, a marked femoral artery abnormality was noted indicating a form of entrapment, supporting a diagnosis of athlete's IAE.

Vein patch angioplasty was the optimum procedure. A bilateral common iliac to common femoral prosthetic graft bypass was performed removing the diseased arteries, restoring blood flow to the legs to normal.

Histopathology examination of smooth muscle actin and muscle-specific actin immunohistochemical stains indicates that intimal thickening in the artery is due to hyperplasia of smooth muscle and fibrous tissue. A small number of cells in the thickened intima are macrophages (histiocytes), as indicated by the CD68-positive cells. This is also part of the reactive intimal response. Detailed descriptive clinical and histopathology report can be seen in Figures 1 and 2.

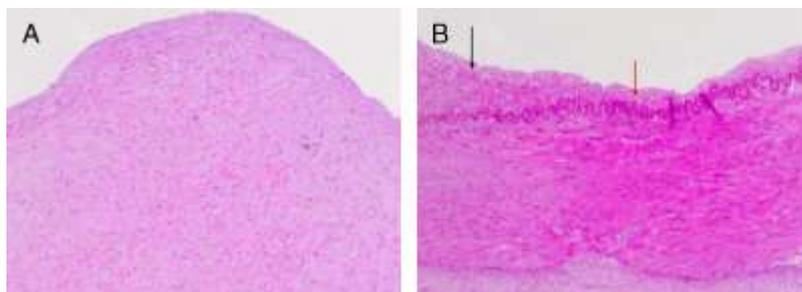
Clinical recommendations regarding return to training and competition were limited to gentle walking for a period of 6 wks. Thereafter, she could start with 5-km runs a day that she could build up as she felt comfortable. She had follow-up sonar evaluations at 3 and 6 mos after surgery, which presented normal. No restriction was placed on competition events, and no anticoagulation medication was prescribed. Six months after the procedure, the patient

has reported no recurrence of any symptoms on re-summing jogging and cycling exercises.

## DISCUSSION

To comprehensively understand why this impairment appeared only after a long sporting career is difficult and can only be speculated on.

Running biomechanics and, more specifically, the magnitude of actual hip joint flexion and extension movements with the repeated stretching and compressing of the external iliac artery may be a contributor of arterial flow limitation in this patient. She has run mostly hills during her training regimen, as a result of the hilly suburb she lives in, and has done spinning classes twice a week and cycling events during training. However, excessive hip flexion during training may not be considered as the only pointer. The acute episode could have resulted from her participation in the Otter Challenge 2 wks before the thrombosis, as enhanced shear forces in the presence of supraphysiologic blood flow has been suggested in the literature.<sup>12</sup> Other contributors that have been described include external compression by an enlarged psoas muscle, nutritional and hemorrheologic factors, a genetic predisposition, and hormonal changes.<sup>5</sup>



**FIGURE 2** Microscopy of the internal elastic lamina shows evidence of marked fibromuscular intimal thickening (A). Internal thickening varies from 0.1 to 0.2 mm (red arrow) to 1 mm (black arrow) (B). No evidence of atheromatous plaques or inflammation was seen.

In individuals with IAE, passionate to continue their sporting activities, surgery with endovascular intervention remains the treatment of choice.<sup>12</sup> Knowledge and appropriate imaging techniques are critical for diagnosis, assessment of location, posttreatment evaluation, and follow-up.<sup>10</sup> Endofibrossectomy with patch angioplasty is the suggested treatment method.<sup>5,7</sup> Excellent results were documented using reconstruction with either autologous or prosthetic interposition vein grafting techniques that demonstrated a 90% primary permeability and 99% return to sport, including high-level competition.<sup>7</sup> Preventing exercise-induced IAE development remains a challenge and requires related pathophysiologic knowledge. Isokinetic testing can be a valuable modality to include to facilitate the rehabilitation process. Attention is drawn to older patients presenting with IAE, bilateral manifestation, incorrect diagnosis, and clinical complications associated with exercise-induced IAE.

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