[ Primary Care ]

# External Iliac Artery Vasospasm in an Elite Female Runner

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A 33-year-old elite female runner presented to a tertiary care sports medicine clinic with a 2-year history of progressive anterior thigh and circumferential leg pain with associated foot paresthesias brought on by high-intensity running. She had both external iliac artery vasospasm and chronic exertional compartment syndrome. External iliac artery vasospasm is a rare cause of exertional leg pain, particularly in the running population. This case highlights the unique features of this condition, addresses the multidisciplinary approach that led to the accurate diagnoses, and demonstrates that more than 1 etiology for exertional leg pain can coexist in an athlete.

Keywords: iliac artery; intermittent claudication; running; compartment syndrome

Exertional leg pain is a potentially disabling problem faced by athletes, particularly runners. The differential diagnosis of exertional leg pain is broad, including musculoskeletal, vascular, and neurologic etiologies. Typically, reports that identify the external iliac artery (EIA) as a source of exertional leg pain describe endofibrosis as the pathophysiologic disturbance within the artery as opposed to vasospasm.[8,11-13] Also, competitive cyclists are the most common athlete population with symptoms of EIA pathology. However, EIA can present in any endurance athlete involved in a sport that requires repetitive hip flexion.[5,6]

## CASE REPORT

A 33-year-old elite runner presented with a 2-year history of unilateral exertional leg pain that progressively limited her competitive performance. She had circumferential left leg pain that was intermittently accompanied by left anterior thigh pain. She experienced associated paresthesias on the dorsum and plantar aspect of her foot that would develop approximately a half mile into her run. Her symptoms only occurred with high-intensity running, worsened by sprinting up hills, and was absent at rest. She transitioned to cycling, which did not improve her symptoms. Despite her symptoms, she remained highly competitive while intermittently stopping during a race to "shake out her leg," which would temporarily reduce her symptoms. She was also able to train for and participate in a full-distance Iron Man triathlon, but had symptoms when training and racing during running and cycling but not while swimming. She pursued multiple conservative interventions but found no improvement with these treatments. Her past medical history included bilateral inguinal hernia repairs at age 2 and a strained left hip flexor 3 years prior to presentation. Her local workup included a lumbar spine magnetic resonance imaging (MRI) study, which demonstrated mild spinal stenosis without neuroforaminal narrowing. She had received no prior electrodiagnostic or vascular studies.

On physical examination, the patient was a thin, athletic-appearing female. Her neurologic examination, including strength, sensation, and reflexes, was normal. Posterior tibial and dorsalis pedis pulses were intact and did not change with passive ankle dorsiflexion or resisted ankle plantarflexion with an extended knee. There was no clubbing, cyanosis, pallor, or edema of the lower extremities. Gait was nonantalgic with no myelopathic features. There was no tenderness to palpation over the left lower extremity musculature.

Because of her normal neurologic examination, the primary differential diagnoses included chronic exertional compartment syndrome (CECS) or vascular etiologies (eg, popliteal artery entrapment syndrome [PAES] or EIA endofibrosis) of exertional leg pain. The workup began with an exertional leg pain MRI of her left leg. This protocol is 96% sensitive and 87% specific for anterior leg compartment CECS and can simultaneously evaluate for PAES and musculoskeletal causes of leg pain (eg, tibial stress

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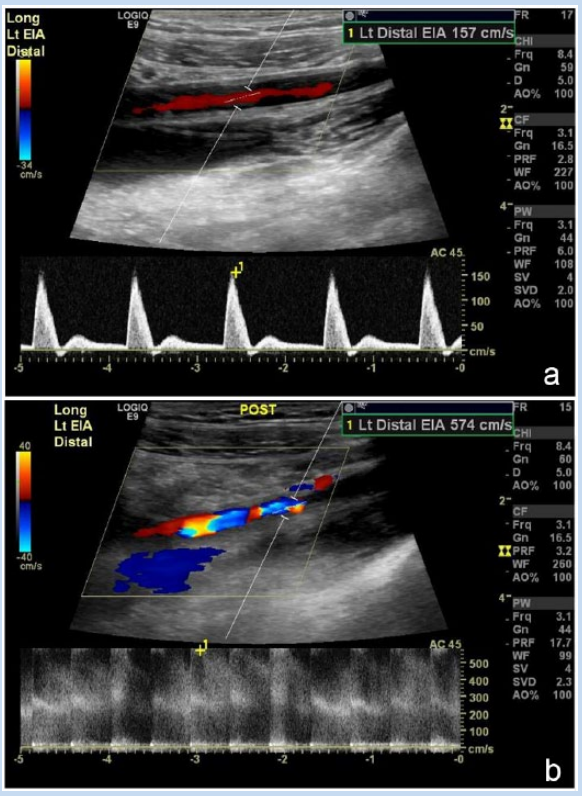


Figure 1. (a) Pre- and (b) postexercise vascular ultrasound images of the external iliac artery (EIA) with respective flow velocities. Note significant postexercise vessel luminal narrowing, increased flow velocity, and mosaic Doppler pattern indicative of turbulence. These findings, which resolved with rest, are consistent with the diagnosis of EIA vasospasm.

fracture).[10] The MRI demonstrated a peak ratio signal intensity of 1.63 in the anterior leg compartment musculature after resisted isometric ankle dorsiflexion exercise (normal, ≤1.54). These findings were consistent with anterior leg CECS. The MRI did not identify PAES or musculoskeletal abnormalities.

Because of the thigh symptoms, she was referred to the vascular surgery department to evaluate for a proximal vascular lesion. Their workup included a preexercise vascular ultrasound, which demonstrated normal common and EIA artery wall thickness, vessel diameter, and blood flow (Figure 1a). No evidence of EIA endofibrosis was found. Pre- and postexercise (treadmill running) ankle brachial indices (ABIs) were subsequently obtained. The speed of her run was at 6.5 miles per hour at a 10% grade, and exercise was terminated at 3 minutes because of thigh, calf, and anterior leg tightness and foot numbness. Her left ABI was normal at rest (normal, 0.9-1.2) with normal Doppler signals. Postexercise, there was an abnormal waveform in the left common femoral artery, and her ABI dropped to 0.21 one minute after exercise. Her left foot was pale and took 8 minutes to return to normal. Her ABI returned to normal (0.90) 15 minutes postexercise.

Owing to the markedly abnormal postexercise ABI, she was reexercised and a vascular ultrasound of the iliac vasculature revealed a marked postexercise increase in blood flow velocities (preexercise, 175 cm/s; postexercise, 574 cm/s), turbulence, and a 3-cm region of luminal narrowing (Figure 1b) that returned to normal 7 minutes postexercise. These findings were consistent with left EIA vasospasm.

Since her MRI suggested CECS in the anterior compartment but had not been validated for CECS in the other leg compartments, pre- and postexercise compartment pressure testing was performed in the left lateral and superficial and deep posterior leg compartments using a compartment pressure monitor (Stryker). Testing was negative (Table 1). Thus, her final diagnosis was EIA vasospasm and anterior leg compartment CECS.

Conservative and surgical management strategies were discussed with the patient. Since diagnostic testing cannot determine which etiology was responsible for her anterior leg pain (or if both etiologies were responsible), the risks and benefits of performing a concomitant anterior leg compartment fasciotomy was discussed. After weighing her options, the patient chose to pursue nonoperative measures; however, she remained symptomatic at her 6-month follow-up.

## DISCUSSION

This case highlights the importance of including EIA vasospasm in the differential diagnosis of exertional leg pain in elite athletes. EIA pathology causing exertional leg pain in high-level running athletes has been described in a few case reports; however, unlike this case, endofibrosis was identified in all cases.[5,7] One case series of 8 female endurance athletes with exertional leg pain identified EIA vasospasm in 2 patients. However, EIA endofibrosis was also present in both patients.[14] The presentation of EIA vasospasm was nearly indistinguishable from symptoms described for EIA endofibrosis. EIA endofibrosis is typically seen in high-level competitive cyclists (1 in 5) but can be seen in other endurance sports at an age when atherosclerosis is uncommon.[4,9] Typical presentation is unilateral lower extremity pain, muscle distension, or weakness that occurs in more than 3 compartments (including the thigh) of the lower extremity during maximal or near maximal exercise involving repetitive hip flexion that is absent at rest or during submaximal exercise. There is a 79% positive predictive value for EIA endofibrosis as the etiology of exertional leg pain when femoral bruits are present at rest combined with the above symptoms.[8,9] The EIA travels along the anterior portion of the psoas muscle. With hip flexion, the EIA becomes functionally lengthened leading to a serpentine course because of the decreased distance it has to travel across the anterior hip.[3,9] This can result in kinking of the EIA during hip flexion. With

Table 1. Pre- and postexercise compartment pressures of the superficial posterior, deep posterior, and lateral leg compartments[a]

| Compartment | Baseline (mm Hg) | 1 Minute Postexercise (mm Hg) | 5 Minutes Postexercise (mm Hg) |
|---|---|---|---|
| Superficial posterior | 13 | 13 | NT |
| Deep posterior | 10 | 19 | NT |
| Lateral | 10 | 11 | NT |

[a]The anterior compartment was not assessed secondary to the positive results on the magnetic resonance imaging exertional protocol. NT, not tested due to the low compartment pressure values 1 minute postexercise.

repetitive hip flexion, the theory is that increased turbulence in the artery leads to intimal hyperplasia of the arterial wall (endofibrosis).[3] The absence of endofibrosis in this case raises the clinical question as to whether vasospasm is a precursor to endofibrosis or whether it exists as its own entity.

Conservative treatment options included modifying the aggravating activities, avoiding activities with repetitive hip flexion, adopting a more upright cycling posture, and decreasing exercise volume and intensity. Symptoms worsen in approximately 80% of patients who continue to participate in the aggravating activities without modification.[9,12] The efficacy of medications for the treatment of EIA vasospasm is unknown. Calcium channel blockers used to treat elderly patients with atherosclerotic lesions have not been effective for EIA endofibrosis found in the high-level athlete. This may be because endofibrosis and atherosclerosis have entirely different pathophysiologic mechanisms and histopathologic composition. Histologic studies comparing endofibrosis versus atherosclerosis in the lumen of diseased EIA specimens found that calcium deposition and inflammatory markers were rarely present in endofibrosis yet observed in about 80% of atherosclerotic lesions.[15] The surgical treatment for EIA vasospasm involves an endarterectomy and saphenous vein patch. This surgery has been quite successful but has only been performed in EIA endofibrosis. One study involving 36 treated endurance athletes found 77% were symptom-free or could perform at their desired level of activity with minimal complaints at 29-month follow-up.[2]

This case also highlights the importance of a thorough systematic approach to the diagnostic evaluation of athletes with exertional leg pain. If a patient presents with unilateral claudicatory symptoms in the thigh after high-intensity endurance activities requiring repetitive hip flexion, a vascular evaluation is imperative. When preexercise vascular ultrasound testing identifies a normal-appearing vessel with no wall thickening or endofibrosis, provocative vascular exercise testing should be performed, as vasospasm of the artery during and after exercise may exist. This evaluation should include pre- and postexercise ABIs. A decrease in the 1-minute postexercise ABI relative to the preexercise ABI of 0.66 is highly suggestive of a vascular cause for exertional leg pain.[1] Since this patient also experienced intermittent anterior leg pain without thigh symptoms, and more than 1 etiology for exertional leg pain can coexist, evaluation for alternative causes of exertional leg pain (eg, PAES and CECS) was warranted. By identifying all of the potential causes of a patient's exertional leg pain, appropriate treatment options can be offered to the patient to fully address the pathology. A staged surgical approach might be indicated. If, for instance, vascular surgery did not completely resolve her anterior leg symptoms, the second etiology (anterior leg CECS) likely would be responsible for the persistent symptoms.

## CONCLUSION

A thorough knowledge of the various etiologies for exertional leg pain assists the clinician with the diagnostic work-up and treatment. By identifying the offending etiology (or etiologies), an appropriate treatment can be initiated leading to the successful treatment of this complex problem.

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