

CASE REPORT

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Chronic Exertional Compartment Syndrome: A Case Report About Claudication in a Healthy AdultAdrienne Mays-Kingston, MD¹, Jeremy Eckels, BS¹, Holly Farkosh, MD¹, Austin Nichols, MD², Paris Johnson, MPH¹, Adam M. Franks, MD¹**ABSTRACT**

Claudication occurs when the blood supply is insufficient to service the musculature in the body with oxygen and metabolic waste management. A clinical complaint of claudication is commonly seen in primary care among older patients with vascular risk factors. A young and healthy patient presenting with claudication is less common and often results in delayed diagnosis with numerous extraneous diagnostic studies. This case discusses a young, healthy male patient with lower extremity symptoms that got worse with exercise and better with rest. He had normal physical exam findings leading to multiple diagnostic studies and over 12 months between the onset of symptoms and his return to full activity. Claudication can result from rare conditions, such as chronic exertional compartment syndrome, popliteal artery entrapment syndrome, fibromuscular dysplasia, and cystic adventitial disease. Symptomatic individuals with chronic exertional compartment syndrome experience reversible muscular pain from exercise-induced pressure, which increases within the finite spaces of any muscular compartment. Understanding the pathophysiology of exertional compartment syndrome and its related diagnoses allows for an organized diagnostic approach to young, healthy patients with claudication symptoms. This organized approach allows timely care, which is imperative for primary care physicians to reduce the number of tests performed, decrease the time to diagnosis, and reduce both the anxiety and cost for the patient. The approach presented herein can serve as a reminder of a proper work-up in similar patients and allow practitioners to identify the conditions that require intervention to improve outcomes.

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INTRODUCTION

Claudication is caused by insufficient blood flow to muscles, most commonly affecting the distal lower extremity. Pain symptoms commonly affect the calf, thigh, and buttocks, typically occur with exercise, and are relieved with rest.¹ Claudication is traditionally seen in older patients, especially those with risk factors including cardiovascular risks of hyperlipidemia, hypertension, diabetes, and smoking. Claudication is uncommon in younger patients, especially those without risk factors for vascular disease. Providers are much less familiar with the differential for lower extremity claudication in a young, healthy athlete (Table 1), which includes

chronic exertional compartment syndrome (CECS), popliteal artery entrapment syndrome (PAES), cystic adventitial disease (CAvdD), and fibromuscular dysplasia (FMD).¹ As a group, these conditions are very rare, but CECS was most frequently seen, accounting for 33%.^{2,3} The incidence of PAES is 0.17%-3.5%, with 85% being male and occurring in the third decade of life.⁴ Also predominately in male athletes, CAvdD occurs in 1 out of 12,000 cases.^{5,6} Iliofemoral artery FMD has very few cases reported.⁷

Compartment syndrome occurs when increased pressure within a myofascial compartment leads to impaired blood flow and innervation to distal structures (Figure 1). Acutely, this surgical emergency



	Chronic Exertional Compartment Syndrome	Cystic Adventitial Disease	Fibromuscular Dysplasia (FMD)	Popliteal Artery Entrapment Syndrome
History of Present Illness	Exertional pain that is relieved with rest	Pain commonly affects the calf but can also be in the thigh and buttock	Exertional pain that is relieved with rest	Exertional pain that is relieved with rest
Physical Exam	Normal physical exam or muscle tension or tenderness palpated at the affected location.	May have diminished lower extremity pulses. May have numbness and tingling in the lower extremities.	Almost always have FMD of other vascular bed, commonly with kidney. Will likely have elevated blood pressure.	Moving the patients' lower extremities into positions such as planar flexion may illicit pain
Epidemiology	33% of athletes with chronic leg pain	Cause of chronic claudication in 1 out of 12,000 cases. Higher predominance in male athletes	Rarely occurs in iliofemoral artery. Few cases have been reported. More common in females.	0.17%-3.5%- Significant male predominance (85%)
Diagnostic Testing	T2 MRI can be used. Intracompartmental pressure (ICP) measured 1 min after exercise with a measurement >27.5 which is a gold standard diagnosis.	Ultrasound or magnetic resonance (MR) angiography	Can be detected with an ultrasound, computer tomography (CT), angiography, and arteriography	Combination of MRI and ultrasound of affected area typically during forced plantar flexion.

TABLE 1: Differential diagnosis for claudication in a young healthy patient.

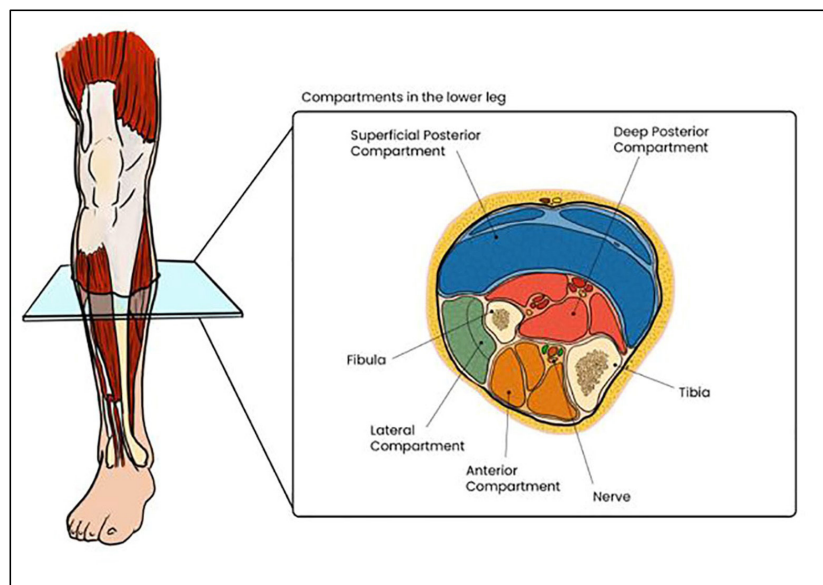


FIGURE 1: Anatomy of the Lower Leg demonstrating different compartment.

is most commonly due to high-impact trauma to the lower extremities but can also occur in the upper extremities and abdomen.⁸ Chronically, compartment syndrome occurs as pressures rise slowly during vigorous exercise. Symptoms include pain, tightness, cramps, muscle weakness, paresthesia, and foot/wrist drop.^{9,10} Up to 90% of patients diagnosed with CECS are affected in anterior or posterior compartments of the lower leg,¹¹ but lateral compartment and both thigh and

abdominal involvement does occur.¹¹⁻¹³ However, vigorous gripping movements such as rowing and motocross have led to CECS in the forearm.⁹ Although mostly seen in military and young athlete populations, it is also seen in older patients who experienced prior trauma or are insulin-treated diabetics.¹⁴ While all family physicians should feel comfortable diagnosing and treating claudication in an older patient with cardiovascular risk factors, not all are comfortable with the same in a young,

healthy patient. The potential delay in identification and treatment can be problematic. Below is a detailed evaluation of a young athlete with lower extremity claudication symptoms.

CASE PRESENTATION

A 34-year-old healthy male presented to his primary care physician complaining of a 4-month history of numbness/tingling in his right lower extremity. The patient also described a “pressure-like sensation, like my leg is going to fall asleep,” but denied pain. This sensation began in his toes, gradually involving the distal right lower leg, most apparent in the posterior region. An avid runner, the patient noted that his symptoms occurred when running or exercising, significantly limiting these activities. He denied any injury or trauma to the lower extremity.

Physical examination revealed a healthy-appearing man with normal vital signs and no obvious abnormalities. The skin of the RLE was normal, without pallor, bruising, rashes, or deformity. Both

lower extremities were non-tender to palpation, with full range of motion of knees and ankles, 5/5 strength, normal sensation, 2+ reflexes, and 2+ pulses. Initial laboratory workup included CBC with differential, electrolytes, peripheral blood smear, ceruloplasmin, rheumatoid factor, anti-CCP, ANA, acute hepatitis panel, protime and INR, and zinc. All were within normal limits. Mild elevations in liver functions aspartate aminotransferase (AST) and alanine aminotransferase (ALT) were observed. An x-ray of the lumbar spine showed multilevel degenerative changes, unlikely to explain the symptoms. Brain magnetic resonance imaging (MRI) was normal. A nerve conduction study displayed no evidence of radiculopathy, plexopathy, neuropathy, or nerve entrapment.

One month later (Figure 2), the patient complained that the symptoms occurred faster with exercise. Sensation began returning 5 minutes after exercise but took about 1-2 hours to return completely to “normal.” He was referred to a sports medicine specialist for further workup with the concern for exertional compartment syndrome. Bilateral

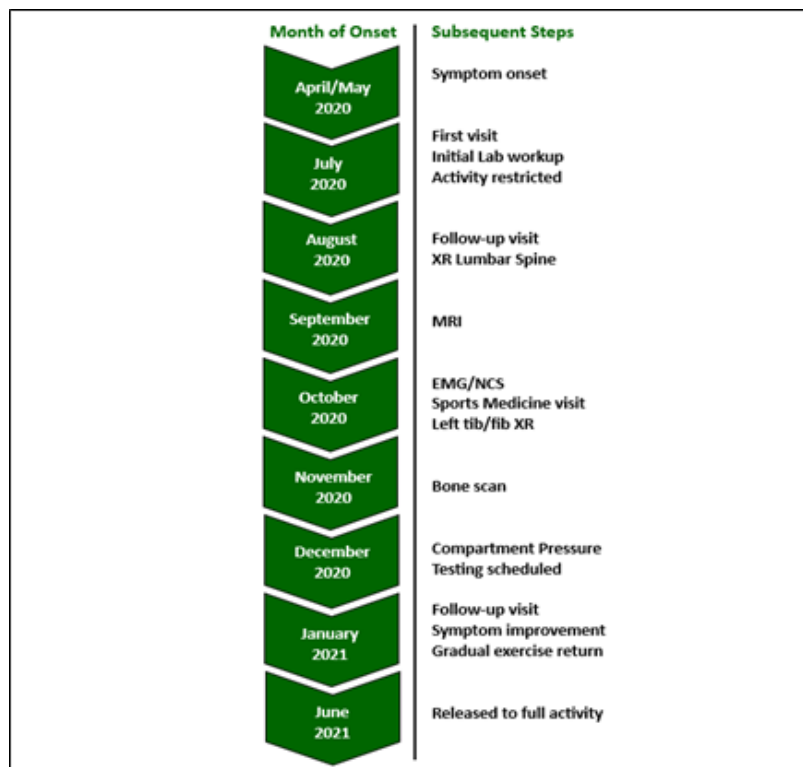


FIGURE 2: Timeline of symptom onset and subsequent steps in treatment until resolution of symptoms. **Compartment pressure testing was originally scheduled, but later cancelled as it is an invasive procedure which the patient chose to forgo as symptom improvement occurred.



tibia-fibula x-rays were unremarkable for acute bony abnormality, and a bone scan ruled out a stress fracture. Compartment pressure testing was ordered but was delayed for over 3 months by local unavailability. While awaiting the testing, the patient decreased his amount and intensity of running and exercising and limited his occupational physical activity. Ten months later, the patient felt better satisfied running every other day at a moderate pace and decreased weekly mileage. He was released back to work with no restrictions on physical duty. Since his symptoms have been well controlled, he declined to pursue compartment pressure testing.

DISCUSSION

Claudication symptoms occur due to occlusion of arterial blood flow to distal extremities. In compartment syndrome, this typically results from an acute increase of pressure within a muscular compartment confined by connected fascial planes, usually by trauma. Vascular occlusion occurs as the pressure of the compartment overcomes the pressure driving the blood flow.¹⁵ The exact physiological mechanism behind CECS is not known. It is hypothesized that a combination of microtrauma, myopathy, and muscle hypertrophy, along with a 20% expansion of muscular volume during exercise,¹⁶ competes against the inelastic fascia of myofascial compartments, leading to neurovascular compromise.¹⁷ Pain results from ischemia during periods of high pressure or pressure-induced myofascial compartment pain receptors.^{9,18} Chronically, repeated episodes of ischemia reduce muscular density and increase fat content when viewed by computed tomography.¹⁵ Muscle biopsy shows fewer type 1 muscle fibers.¹⁵ Prolonged ischemia also decreases mitochondrial function, leading to impaired metabolism. This inhibits skeletal muscle ATP generation, increasing reactive oxygen species and oxidative degeneration.¹⁵

Diagnosis of CECS is difficult due to the variety of conditions that present with similar symptomology (Table 1).^{10,11,16} A T2-weighted MRI can rule out other causes of intermittent non-claudication pain, such as stress fractures, thrombosis, nerve injury, or tendon injury.¹¹ Diagnosis of claudication should be suspected on clinical presentation. Dorsalis pedis

and/or posterior tibial pulses may be reduced, and a low ankle-brachial index (ABI) of <0.9 further supports the diagnosis of claudication.¹ Difficulty exists in catching these intermittent changes. Exclusion of other claudication syndromes in young patients is important (Figure 3). The diagnosis of PAES is suspected if plantar flexion of the affected lower extremity elicits symptoms and diminishes dorsalis pedis pulses and is confirmed with a combination of MRI and ultrasound.⁴ Magnetic resonance angiography diagnoses CADvD by revealing an “hourglass” appearance within the affected artery, indicating stenosis.^{5,6} Duplex ultrasound will also show an anechoic or hypoechoic structure within the affected artery or vein.⁵ Iliofemoral artery FMD can be seen by duplex ultrasound, CT angiography, and arteriography.⁷

In CECS, distal pulses or normal ABI do not rule out claudication, as this process is intermittent. A post-exercise ABI may be required to appreciate the decreased pressures upon symptom development.¹ While no specific guidelines exist to diagnose CECS,

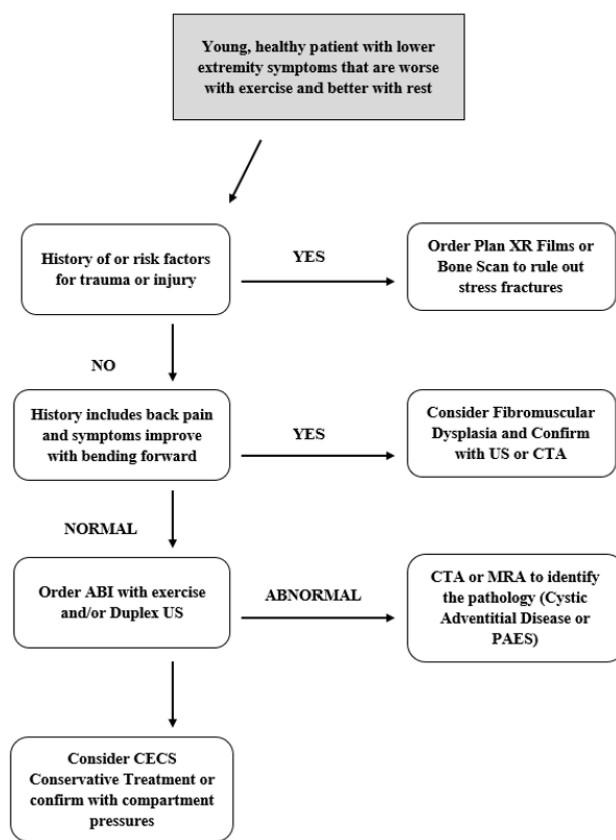


FIGURE 3: Suggested patient diagnostic workflow.



the current standard practice is intra-compartmental pressure testing by traditional manometry. Diagnosis of CECS using the Pedowitz criteria requires a pressure >15 mmHg at rest, > 30 mmHg after 1 minute of exercise,¹⁹ Less-invasive diagnostic techniques such as T2 MRI, infrared spectroscopy,^{11,18} vascular testing, bone scans, and laser Doppler flow have also been utilized.^{8,20}

Primary care physicians rely on epidemiology and exposure to develop a differential for patients' symptoms. It is only in retrospect that the diagnosis is clear. As a majority of patients with claudication symptoms have risk factors for vascular disease, an extensive evaluation was undertaken and found to be negative. With a normal physical exam and a lack of cardiovascular risk factors for vascular disease, the evaluation shifted away from traditional claudication-associated syndromes. Despite the lack of manometry testing, this patient clearly had CECS. His subjective symptoms were consistent with exercise-induced claudication and improved with conservative therapy. His lack of hypertension, family history, or supporting physical exam reduced the chance of FMD. Both CAdvD and PAES would have failed to improve with conservative treatment.

TREATMENT/PROGNOSIS

The treatment of claudication in a young, healthy adult differs based on the overall cause of the symptoms. Surgically releasing the popliteal artery is only beneficial for symptomatic patients with PAES, as asymptomatic individuals rarely progress to have symptoms.⁴ While surgical resection of the affected area and venous grafting is the definitive treatment for CAdvD,^{5,6} ultrasound-guided cyst aspiration has also been shown to improve symptoms.⁵ Stenting of the affected area, followed by anti-platelet therapy, is the treatment of choice for FMD.⁷

Treatment of CECS varies depending upon the patient's case. Fasciotomy of the affected compartment is invasive and should be reserved for severe cases.^{9,18,20} Conservative treatments such as gait modification, rest, activity modulation, chemodenervation, and physical therapy have shown success.²¹ Recovery depends upon the individual case, but while best outcomes are associated with

fasciotomies, surgical complications can be as high as 13%.¹⁰ Compared to age-matched peers, patients with intermittent claudication have an increased mortality rate of 12%/year²² due to heart disease (66%) and stroke (10%).¹ This risk is inverse to the patient's ABI. Serious adverse outcomes are related to cardiovascular risk factors like smoking and diabetes, with amputation rates between 1-7% at 5-10 years.¹

CONCLUSION

All primary care providers are familiar with identifying and managing claudication in older patients with cardiovascular risks. However, exposure to the same symptoms in young, healthy patients is rare. Unfamiliarity with the differential, evaluation, and treatment can lead to delays in management. An algorithmic approach available for primary care providers would allow them to more efficiently test, educate, and treat their young, healthy patients with claudication.

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