

# Nonsurgical treatment of cystic adventitial disease of the popliteal artery caused by functional popliteal artery entrapment syndrome

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Lower extremity claudication in young men can be caused by popliteal artery entrapment and vascular pathology. The case reported here describes a young male wrestler who presented with unilateral calf pain with activity. Imaging studies at rest demonstrated cystic adventitial disease of the popliteal artery. Stress imaging showed severe functional popliteal artery entrapment. This patient was treated with an imaging-guided botulinum toxin injection of the muscles at the site of arterial compression. The patient was asymptomatic 6 weeks after treatment. Repeat stress imaging demonstrated complete resolution of the entrapment and nearly complete resolution of the popliteal artery cystic adventitial disease. (*J Vasc Surg Cases* 2015;1:28-31.)

Cystic adventitial disease (CAD) of the popliteal artery is a rare cause of lower extremity claudication. CAD was first described in the external iliac artery by Atkins and Key in 1947.<sup>1</sup> The disease is most often unilateral, affecting the popliteal artery (85%).<sup>2</sup> Young men, otherwise healthy, are most commonly affected and often present with intermittent claudication.<sup>3</sup> The prevalence is one of every 1200 cases of calf claudication.<sup>3</sup>

Several etiologies of CAD have been proposed, including repeated microtrauma with intramural bleeding, synovial tracking, and true synovial cysts.<sup>3-6</sup> Treatment of CAD can be challenging. Current treatment methods include image-guided cyst aspiration, surgical cystectomy, or resection of the affected segment of blood vessel with placement of a graft.<sup>7,8</sup> There have also been rare cases of spontaneous cysts regression and treatment with percutaneous transluminal angioplasty with limited success.<sup>9-13</sup> Written consent was obtained from the patient's parents to publish this study.

## CASE REPORT

A 17-year-old male, right side-dominant wrestler, presented with right lower extremity pain exacerbated during wrestling practices and competitions for the past 7 months. The patient described severe calf and ankle pain of 10 of 10 along with some

degree of numbness and tingling in his foot. The patient is otherwise healthy and is a nonsmoker, with no history of vascular disease, traumatic injury, or prior surgery. Vital signs were normal, and the patient's history was otherwise noncontributory.

On physical examination at rest, the patient's pulses were symmetric in the lower extremity. There were no areas of discoloration or venous varices. The calf muscles were well developed bilaterally. The patient had normal strength, range of motion, sensation, and reflexes. Bilateral plantar flexion against resistance at 63.5 kg using the MVP FLEX (MVP Innovations, Casper, Wyo) resulted in recreation of the patient's symptoms in the right leg. There was also mild pallor of the distal toes of the right foot. The left foot was normal in appearance.

Noncontrast magnetic resonance imaging demonstrated abnormal cystic change in the right popliteal artery (Fig 1). A subsequent computed tomography (CT) angiogram was performed at rest and with plantar flexion against resistance at 63.5 kg using the MVP FLEX (Fig 2). The CT scan demonstrated a cyst measuring 10 × 16 × 48 mm in the wall of the right popliteal artery. In addition, with stress maneuvers, the imaging demonstrated severe bilateral functional popliteal artery entrapment syndrome (fPAES) due to compression by the gastrocnemius muscle (Fig 1).

A treatment was proposed to the patient using an imaging-guided botulinum toxin injection of the muscle at the site of vascular compression. A small area of muscle was targeted in the bilateral leg using ultrasound and CT guidance. Five 25-gauge needles were placed into the gastrocnemius muscle directly adjacent to the sites of vascular compression (Fig 2). After confirmation of needle placement, 200 units of Botox (Allergan, Irvine, Calif) reconstituted in normal saline were injected under ultrasound guidance into each leg.

The patient returned to physical activity 2 days after the procedure. By 1 month after treatment the symptoms in the right leg had resolved. The patient returned for follow-up imaging at 6 weeks. A repeat CT angiogram of the legs was performed at identical levels of plantar flexion force (63.5 kg) using the MVP Flex. Imaging studies demonstrated complete resolution of the functional popliteal artery entrapment bilaterally and nearly complete resolution of the CAD of the right popliteal artery (Fig 3). The cyst now measured 3 × 5 × 36 mm.

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Author conflict of interest: J.C.M. has a pending patent for the MVP Flex used in this study for diagnostic purposes.

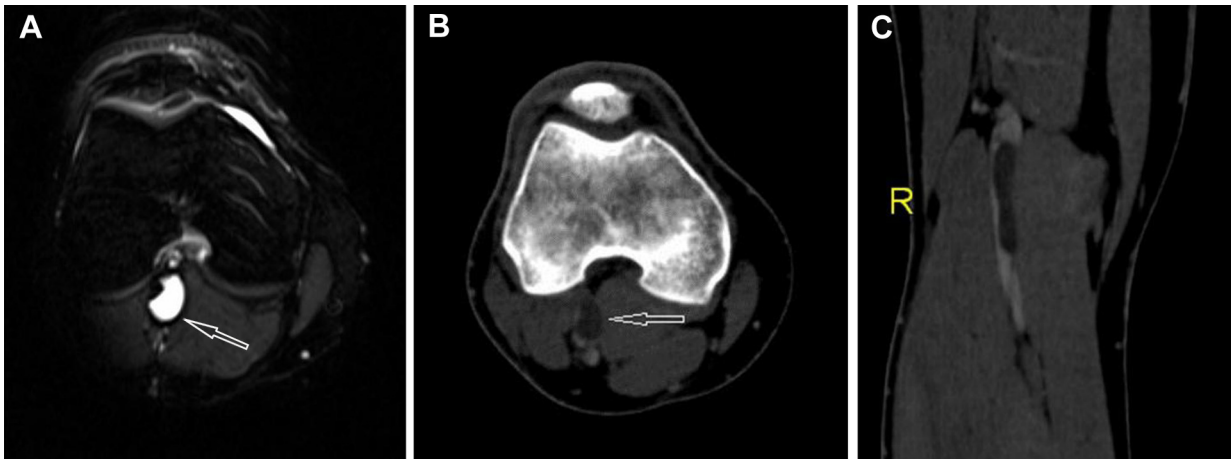
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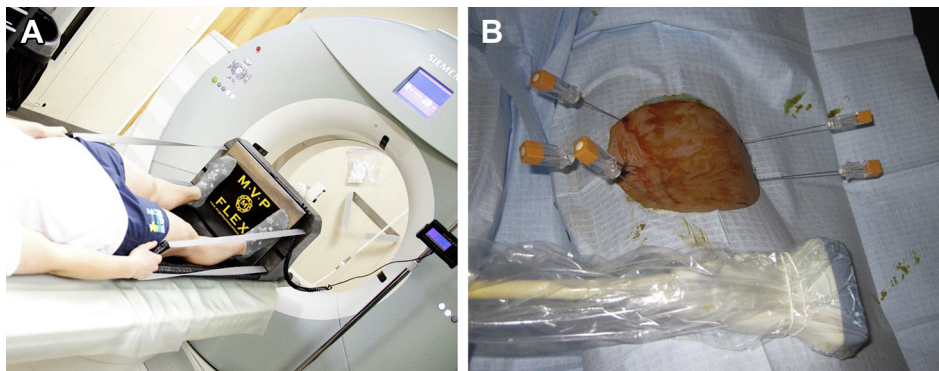
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**Fig 1.** A, Resting axial short-tau inversion recovery magnetic resonance image of the right popliteal fossa demonstrates cystic change in the popliteal artery (*arrow*). B, Axial stress computed tomography (CT) angiographic image of the right leg at the level of the popliteal fossa demonstrates significant arterial compression and cystic adventitial disease (CAD) of the popliteal artery (*arrow*). C, Coronal stress CT angiographic image demonstrates the extent of cystic change along the popliteal artery.



**Fig 2.** A, Patient positioned in the computed tomography (CT) scanner on the MVP FLEX (MVP Innovations, Casper, Wyo) for stress imaging. The patient's plantar flexion force against resistance was monitored on the adjacent screen. B, Needle placement in the popliteal fossa for injection.

The patient was asymptomatic at 5 months after treatment. He will continue to be monitored with planned in-office follow-up and stress imaging at 6 months, 1 year, and 2 years. We also plan on retreating the affected areas at ~6 and 15 months after the initial treatment to promote local muscle atrophy and muscle retraining to prevent recurrence of the vascular compression.

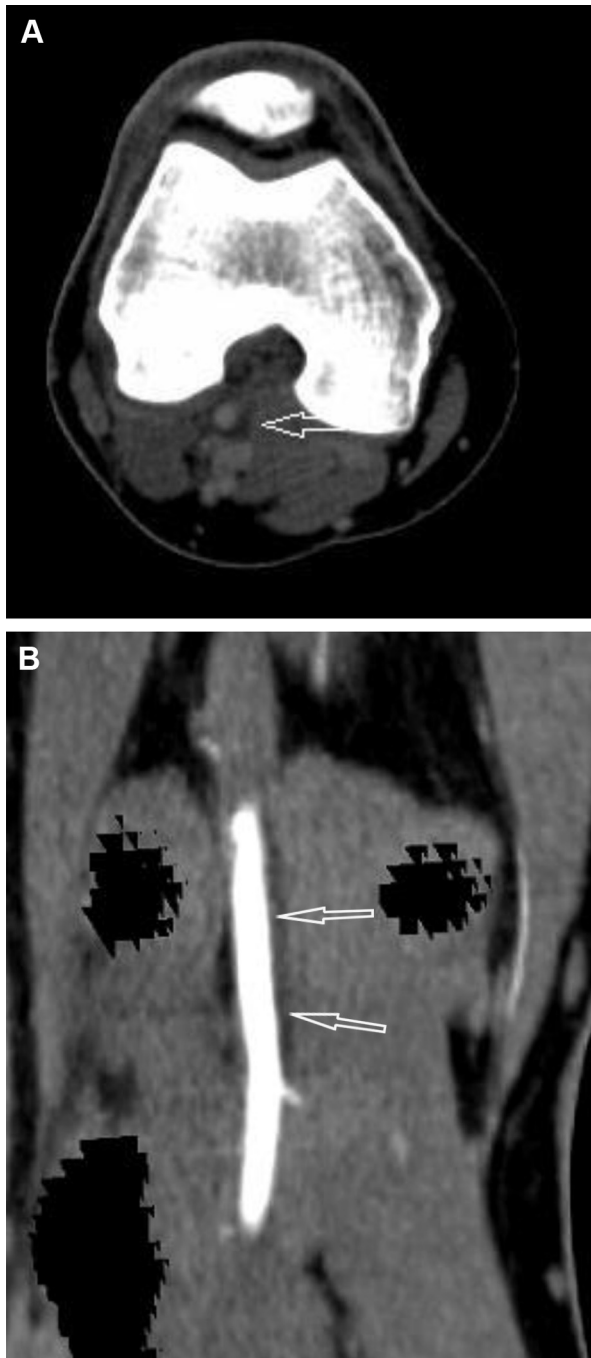
## DISCUSSION

Lower extremity vascular disease in young healthy men is a rare medical condition. The case presented here describes a healthy young man with fPAES and CAD of the popliteal artery. This case outlines a nonsurgical approach to treating CAD likely caused by fPAES.

CAD is more common in men in the fourth or fifth decades of life. Cystic enlargement can produce mass effect on the adjacent flow lumen, thereby decreasing blood flow to the lower extremity. Proposed causes of CAD

include repeated microtrauma, ectopic aganglionosis, systemic connective tissue disease, developmental abnormalities, or synovial herniation.<sup>14</sup> Pathology demonstrates a collection of gelatinous material containing mucoproteins and mucopolysaccharides located between the adventitia and middle layer of the vessel wall.<sup>12,15</sup>

The diagnostic evaluation includes a thorough history and physical examination. Most patients are asymptomatic at rest, and the physical examination is normal. With stress maneuvers, including passive dorsiflexion or active plantar flexion of the foot, symptoms may be reproduced, including pain, paresthesia, and pallor, within the lower extremity. Imaging studies include vascular ultrasound, magnetic resonance angiography, or CT angiography. Stress maneuvers should be performed during imaging examinations to assist in identifying functional causes of vascular injury, including PAES.



**Fig 3.** **A**, Axial stress computed tomography (CT) angiogram 6 weeks after treatment demonstrates a significant reduction in the cyst size (*arrow*) compared with the pretreatment image. **B**, Coronal stress CT angiogram 6 weeks after treatment demonstrates significant reduction in the cyst size compared with the pretreatment image (*arrow*).

PAES is one potential cause of CAD due to repeated microtrauma. Arterial compression can result in early arteriosclerosis and thrombus formation with associated distal ischemia. In addition, the abnormal blood flow

mechanics can result in post-stenotic ectasia or aneurysm formation.

Treatment of CAD primarily involves surgical resection of the diseased segment with graft interposition, with varying degrees of success.<sup>7,8</sup> Less invasive techniques, including cyst aspiration and cyst resection, have also been described with moderate success.<sup>16</sup>

The patient described above presented with severe fPAES and CAD resulting in unilateral lower extremity claudication. Treatment of fPAES in this patient resulted in nearly complete cyst resolution and complete symptom resolution by 6 weeks after treatment. The patient is currently 5 months after treatment and remains asymptomatic and active in competitive sports. The technique used is minimally invasive and targets the cause of the pathology, which in this individual was likely functional popliteal artery compression due to overdeveloped calf muscles in the dominant leg. Examination and imaging using reproducible forces of plantar flexion against resistance resulted in a direct nonsubjective comparison of the muscles and vasculature before and after treatment.

My experience using this diagnostic and treatment protocol in other patients with fPAES has the following average results: (1) patients typically require three treatments over a 2-year period, (2) localized atrophy of the treated muscle was demonstrated on subsequent imaging, and (3) no significant side effects were noted, but occasional transient muscle soreness and weakness have been reported lasting 2 to 3 weeks.

## CONCLUSIONS

CAD is a rare entity with limited treatment options. The case presented suggests fPAES is one possible cause of CAD. This patient was successfully treated using a minimally invasive, imaging-guided intervention focally targeting the site of dynamic vascular compression. The patient returned to sports without limitations or side effects from the treatment. Imaging studies were performed with controlled, reproducible, levels of muscle engagement allowing direct comparison of before and after treatment outcomes. The patient continues to remain asymptomatic 5 months after treatment.

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