

Statement of Purpose

To report a rare presentation and treatment of chronic, atraumatic exertional compartment syndrome (ECS) in the bilateral foot.

Literature Review

Acute, atraumatic cases of exertional compartment syndrome (ECS) of the lower extremity are unusual. Most commonly reported cases involve the anterior and/or lateral compartments of the leg following intense exercise such as running or spinning [1-3]. Noted in these presentations is the concern that in addition to the localized compromise to neuro-vascular structures, that resulting muscular damage can produce rhabdomyolysis that can be life-threatening in terms of causing acute kidney injury, cardiac dysrhythmias, and disseminated intravascular coagulopathy [4].

Even more uncommon are the reporting of atraumatic ECS that localize within the foot itself. Previously, acute cases of atraumatic ECS have been noted to the author's knowledge on five other occasions, once, following spinal surgery and thought to be caused by neuro-stimulation related to the surgery [5], and four other times in the acute setting, three requiring fasciotomies [6-8], one following only a conservative course [9].

However, no prior study has reported a case of chronic, atraumatic ECS isolated to the foot. We report here the case of a young, athletic female who develops progressive bilateral, chronic atraumatic ECS in the initial setting of running that progresses to symptomatic levels with simple, short distance walking over a period of approximately two years.

Case Study

History of Presenting Illness

A 19-year old female patient presents stating that about two years prior, she had been running in a 5K race and she began to develop numbness in the medial, plantar arch of both feet. She continued to run over the next few months, but she noticed that the numbness was re-occurring at progressively short distances, with her most recent run having produced the same numbness after running 0.1 mile.

Case Study (Continued)

At the time of presentation, the patient was a university student at a major school, which required walking 5-10 minutes between buildings for classes. She noted that she frequently needed to stop to rest because of the pain generated from these brief periods of brisk walking. The patient was no longer able to run at the initial presentation due to the pain generated during exertion. She denied having problems with pain radiating proximally above the ankle, symptoms of calf or buttock claudication.



Figure 1: Clinic treadmill testing produced palpable bulge of the medial foot after running 0.5 mile.

Physical Exam: Prior to Exertion

A comprehensive lower extremity exam was conducted, which included having the patient run on a clinic treadmill. Prior to the run, the patient's lower extremity was examined, noting no open skin lesions, no signs of trauma. Dorsalis pedis and posterior tibial arterial pulses were weakly palpable and confirmed to have a biphasic pattern via hand-held doppler.

While at rest, patient was noted to have intact muscle strength and pain could not be elicited from the feet from passive and active range of motion involving rearfoot and forefoot joints, Pratt's testing, heel rise testing, palpation of the soleal sling, common fibular nerve, medial tarsal tunnel.

Neurologically, the patient had normal sensory responses to Wartenberg wheel (all dermatomes), two-point discrimination, vibration, light touch, proprioception, contrast temperature differentiation, Achilles and patellar reflexes.

Physical Exam: Treadmill Running

Patient was placed on a treadmill in the clinic and allowed to self-select a comfortable running speed, settling at 5.5 mph. **Table 1** records specific distances and patient's symptoms as they relate to the above history. After reaching 0.52 miles (mi), the patient noted that her pain was too severe to continue to run and stopped to help relieve the pain.

Case Study (continued)

Table 1: Patient response during Treadmill Running

Distance (mile)	Symptom	Location
0.25	Numbness	Right > Left medial, plantar midfoot
0.46	Mild achy pain	Bilaterally equal, same distribution
0.52	Severe achy pain	Bilaterally equal, same distribution

Pre-operative Imaging

Patient was sent for bilateral MRI to determine if there were impingement near the level of the tarsal tunnel based upon findings of the treadmill running test. **Figure 2** illustrates MRI findings of the left foot, which were also similarly found on the right foot. Noted in these cross-sections are multiple tortuous blood vessels coursing into the porta pedis region with an enlarged abductor hallucis muscle belly.

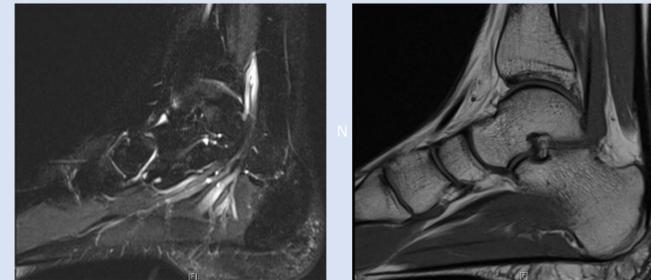


Figure 2: Pre-operative MRI of the left foot demonstrating several branching blood vessels and enlarged abductor hallucis at the porta pedis.

Pre-operative Vascular Testing

As noted previously, the patient had dopplerable pulses. In order to demonstrate decrease blood flow to plantar foot following exercise, the patient had segmental pressures taken after the patient had performed 40 toe-up exercise motions. Seen in **Figure 3** is the notable decrease in waveform quality and pressures in the plantar foot.

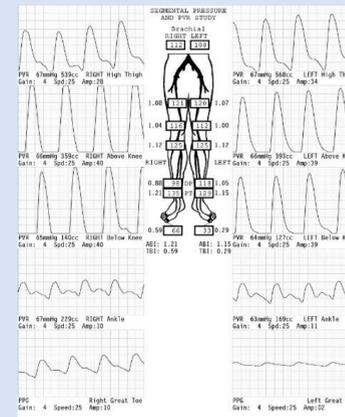


Figure 3: Segmental pressures of the lower extremity after 40 toe-up exercise motions.

Case Study (continued)

Surgical Intervention

Following pre-operative testing, patient was taken to surgery on two separate occasions for external neurolysis of the tibial nerve and its branches distal and proximal to the medial malleolar axis. Shown in **Figure 4** illustrates intra-operative NIMS monitoring set-up.



Figure 4: NIMS set-up used in right foot surgery.

Following surgical release of the fascia, multiple aberrant vascular branches, and debulking of the abductor hallucis entrapping the medial foot as seen in **Figure 5**, the patient experienced baseline voltage reading increases from <300 μ V pre-release to >3300 μ V post-release.



Figure 5: Intra-operative image of the right foot immediately prior to surgical release. (Left) Demonstrates the engorged nature of the vascular supply immediately adjacent to the tibial nerve as it courses into the plantar foot. (Right) Illustrates bulking of the abductor hallucis, which was noted to entangle the tibial nerve and its branches.

Analysis & Discussion

Post-Operative Period

Within 4 months of both tibial nerve neurolysis surgeries, patient had been able to return to running activities with no pain occurring at any long distance patient attempted to run (tested at over 5 miles at one time). At the 17 months follow-up visit, patient has maintained full activity with no symptomatic re-occurrence.

Case Pearls

This case report is the first to report chronic, atraumatic exercise induced compartment syndrome isolated to the foot. Because of the progressive worsening of symptoms over the two year period that the patient reports problems, it is hypothesized that the enlarged abductor hallucis would engorge with blood with physical activity. This engorgement of the abductor hallucis lead to tibial nerve entrapment and vascular compromise. With each episode, the oxygen starved plantar foot may be further stimulated vascular collateral growth, facilitating the presentation of the aberrant, tortuous multiplication of vessels in the region – further entrapping the regions nervous tissue in a progressive manner.

Key pearls in this cases' investigation were as follows:

- Re-production of patient's compartment syndrome with treadmill running.
- Capturing evidences of the abherent anatomy present in the medial foot (i.e. abductor hallucis enlargement, tortuous blood vessels)
- Characterization of exercise effects of blood flow with segmental limb pressures following exercise
- Demonstration of improved nerve signaling intra-operatively with the NIMS device

References

1. McKinney, B., C. Gaunder, and R. Schumer, *Acute Exertional Compartment Syndrome with Rhabdomyolysis: Case Report and Review of Literature*. Am J Case Rep, 2018. 19: p. 145-149.
2. Schwartz, A., C. Poole, and C. Schlieien, *Characterization of the Development of Acute-on-Chronic Exertional Compartment Syndrome A Case Report of Symmetric Compartment Syndromes and Review of the Literature*. Bull Hosp Jt Dis (2013), 2017. 75(2): p. 148-152.
3. Kim, D., et al., *Spinning-induced Rhabdomyolysis: Eleven Case Reports and Review of the Literature*. Electrolyte Blood Press, 2015. 13(2): p. 58-61.
4. Jordan, K.S. and S.E. Mannel, *A Case of Exertional Rhabdomyolysis: When Exertion Exceeds Capacity*. Adv Emerg Nurs J, 2017. 39(2): p. 106-113.
5. Stotts, A.K., et al., *Medial compartment syndrome of the foot: an unusual complication of spine surgery*. Spine (Phila Pa 1976), 2003. 28(6): p. E118-20.
6. Myerson, M.S. and B.I. Berger, *Isolated medial compartment syndrome of the foot: a case report*. Foot Ankle Int, 1996. 17(3): p. 183-5.
7. Miozzari, H.H., et al., *Acute, exertional medial compartment syndrome of the foot in a high-level athlete: a case report*. Am J Sports Med, 2008. 36(5): p. 983-6.
8. Blackledge, D.K., et al., *Acute exertional compartment syndrome of the medial foot*. J Foot Ankle Surg, 1996. 35(1): p. 19-22.
9. Kelsey, N.R., L.D. Edmonds, and D.M. Biko, *Acute exertional medial compartment syndrome of the foot in a teenager*. Radiol Case Rep, 2015. 10(2): p. 1092.