

Idiopathic Acute Four-Compartment Syndrome of the Lower Leg

Zachary Jodoin^{1*}, Spencer Sims¹, Timothy Petsche²

¹College of Human Medicine, Michigan State University, East Lansing, MI, USA;

²Fox Valley Orthopaedics, Geneva, IL (USA)

*Corresponding Author: Zachary Jodoin; jodoinza@msu.edu

Case: We present a case of acute idiopathic four-compartment syndrome of the leg, treated by four-compartment fasciotomy, and wounds left to heal by secondary intention due to persistent edema following surgery.

Conclusion: This case highlights the importance of maintaining a high level of clinical suspicion for idiopathic spontaneous compartment syndrome presentation. This case also illustrates the variability of compartment syndrome treatment and recovery. The standard treatment for compartment syndrome is fasciotomy with delayed primary wound closure, but the patient elected to heal by secondary intention. The patient's long-term follow-up results showed positive outcomes.

Keywords: Compartment Syndrome; Orthopedics; Wound; Wound Complication; Wound Closure; Idiopathic Compartment Syndrome

INTRODUCTION AND PATIENT PROFILE

Compartment syndrome is a surgical emergency commonly associated with a traumatic event.¹ The pathophysiology involves increased fluid and tissue volume within a rigid fascial-contained compartment leading to tissue hypoxia and necrosis.² Compartment syndrome typically presents with pain, pallor, paresthesia, pulselessness, and paralysis. Diagnosis is difficult, and misdiagnosis can lead to significant morbidity and mortality.³ Intracompartmental pressure monitoring can confirm compartment syndrome, but this is invasive and painful.^{4,5} Treatment is straightforward with fasciotomy and delayed primary re-approximation.⁶ The most difficult aspect of compartment syndrome is swift diagnosis due to the rapidly progressive nature of its pathophysiology. By maintaining a high degree of clinical suspicion, a physician can decrease the odds of making a potentially fatal misdiagnosis. Here we present a case of acute idiopathic compartment syndrome of the leg, treated by four-compartment fasciotomy and wounds left to heal by secondary intention due to persistent postoperative edema. Long-term follow-up showed great patient satisfaction, improved muscle function and strength, and return to normal ambulation.

INTERVENTIONS AND OUTCOMES

A 32-year-old woman with a medical history of rheumatoid arthritis and sarcoidosis presented to an outside emergency department due to persistent right foot pain and numbness that woke her from sleep. She was evaluated in the emergency department (ED) and found to have mild swelling and tenderness in the right lower leg. She received an ultrasound that was negative for deep vein thrombosis (DVT), and she was discharged home with crutches and without orthopedic consultation.

Due to persistent pain, swelling, numbness, and weakness in the right leg, the patient returned to the emergency department 8 hours later. She reported no recent history of trauma or unusual physical exertion. Workup included X-rays, blood work, repeat venous Doppler ultrasound, and computed tomography (CT) scan. X-rays of the tibia and fibula were unremarkable. CT scan of the right leg showed diffuse, four-compartment edema with no obvious source. Orthopedic consultation was obtained to work-up suspected compartment syndrome. Upon orthopedic physical exam, the right leg showed no active muscle activity below the right knee, passive range of motion produced significant discomfort, distal pulses were diminished, and the compartments were tense to palpation.

Anterior and posterior intracompartmental pressures were measured at 55 and 52 mmHg confirming the diagnosis of compartment syndrome. The patient was diagnosed with idiopathic acute right leg compartment syndrome, and informed consent was obtained to perform emergency fasciotomy.

In the operating room, the patient was given general anesthetic, she was prepped and draped in routine fashion, a four-compartment right lower extremity fasciotomy was performed, and the wounds were packed with saline-soaked gauze and loosely wrapped to prevent further increased compartment pressures (Fig. 1).

Following surgery, the patient had bounding dorsalis pedis pulse, persistent loss of sensation to light touch, and complete loss of motor function below the knee.

The patient was discharged the following day with close outpatient follow-up.

Two weeks post-op: The right leg below the knee showed persistent edema and significant granulation tissue in the wounds. There were no signs of active infection. The patient had significant improvement to light touch sensation of the right foot, but no motor activity was observed. Due to persistent edema, referral to a wound clinic and re-evaluation for future skin grafting were ordered.

Four weeks post-op: The patient returned with significantly improved edema. The wounds had excellent closure and granulation. Delayed wound closure versus

healing by secondary intention was discussed. The patient elected for wound healing by secondary intention (Fig. 2). Use of an ankle-foot orthosis (AFO) was prescribed to keep the ankle in a neutral position when resting.

Three months post-op: The wounds were healed. She had active eversion of the foot and active flexion and extension of the toes. She could not significantly dorsiflex. She had intact sensation to light touch from the knee all the way down to the ankle. Below the ankle, she had light touch sensation in patchy distributions.

Six months post-op: The patient had a similar clinical picture with improved dorsiflexion.

Twenty-eight months post-op: The patient had weakness and limited range of motion in her right ankle. Her prior AFO broke, and the patient had not been using it for some time. At this time, treatment with physical therapy was started to improve ROM and strength in the ankle. Physical therapy evaluation showed full strength to plantar flexion, inversion, and eversion of the right ankle when compared to the left. The patient still had 2/5 strength to dorsiflexion in the right ankle.

Thirty months post-op: She ambulated with a normal gait and had improved dorsiflexion from the prior visit. The ankle rested in a neutral position. The patient was satisfied with her progress and was prescribed a new AFO and encouraged follow-up as needed.



Figure 1. Surgical image of a fasciotomy to decrease compartment pressures. (This is an open access photo with no citation required.)

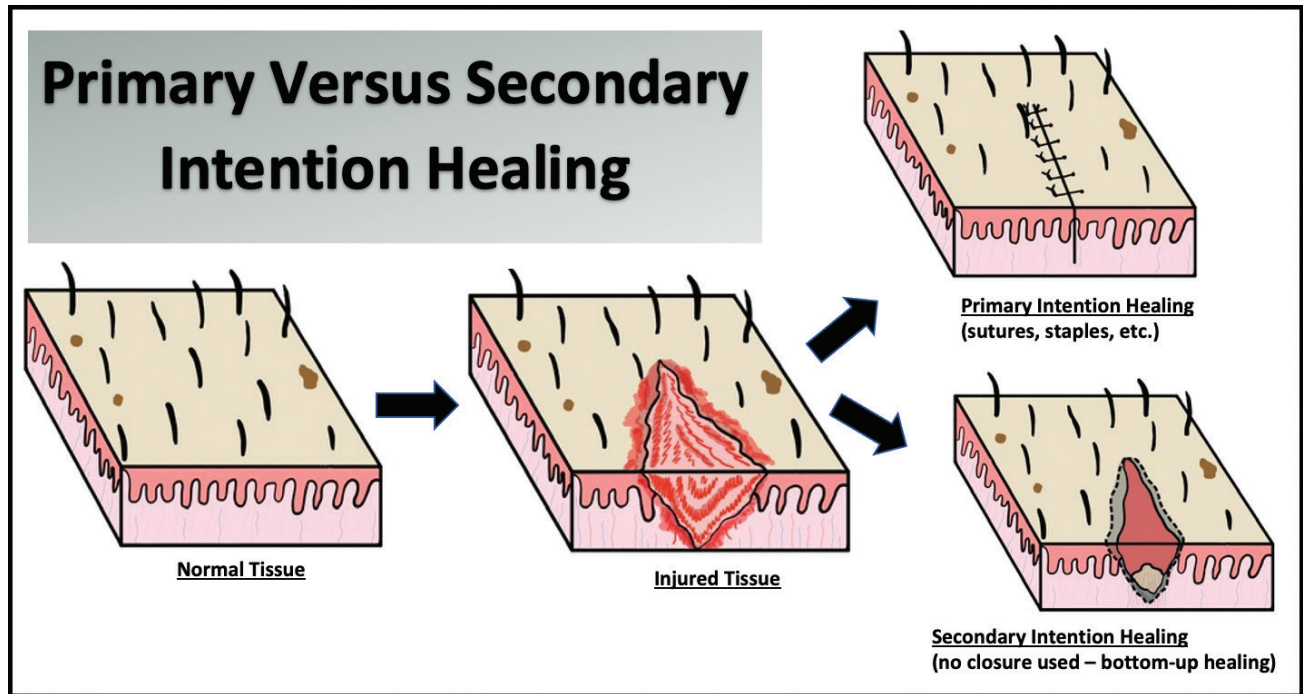


Figure 2. Illustration of primary versus secondary intention healing. (This is an image created by the author, Zachary Jodoin.)

DISCUSSION

This patient's plan of care had two unique components: the idiopathic presentation and the secondary intention healing.

Lower leg compartment syndrome is almost always associated with a high-energy trauma.¹ A study of 164 patients with acute compartment syndrome linked 69% of the cases to a fracture.⁷ It can also be seen with surgical insult such as total knee arthroplasty or coronary arterial bypass grafting. Other rarer causes of compartment syndrome include systemic diseases such as diabetes mellitus, HIV, hypothyroidism, hematogenous malignancy, systemic sclerosis, and neuroleptic psychiatric or statin medications.^{1,8,9} Our patient had a history of sarcoidosis with resulting rheumatoid-like arthropathy but given the patient's mild nature of these conditions and lack of evidence relating to the cause of compartment syndrome, it is unlikely the inciting factor in this patient's case. The fact that idiopathic acute spontaneous compartment syndrome has no real intrinsic or extrinsic trigger makes it very difficult to consider as a diagnosis. This, combined with the morbid and even fatal consequences of missing the diagnosis is what makes this case so important. The patient's discolored

muscle, visualized after fasciotomy, echoes the importance of maintaining a high level of clinical suspicion for compartment syndrome. Further delay of this patient's care could have led to amputation, renal failure, or death. Although other case reports have been published pertaining to acute idiopathic compartment syndrome of the lower leg, this report gives readers more insight to postoperative recovery and follow-up.^{11,12}

Closure of post-fasciotomy wounds is often challenging due to soft tissue edema and protrusion of muscles through the wounds.¹³ Due to the likely increased risk of infection, research into wound closure by secondary intention has been limited. In a 1992 study evaluating wound closure techniques in patients with traumatic compartment syndrome, wound complication rate was observed in 51% of wounds with delayed primary closure or wounds left to heal by secondary intention. This was compared to 5% with post-fasciotomy skin-grafting.¹⁴ Long-term follow-up, however, has found increased morbidity with skin grafting.¹³ Multiple post-fasciotomy wound closure techniques have been described. Kakagia reviewed techniques to achieve delayed primary closure, including the use of dynamic dermatotraction mechanical devices to provide

negative pressure therapy to the wound.¹³ It should be noted that persistently increased compartment pressures have been observed in premature primary closure of wounds, increasing the risk for recurrent iatrogenic compartment syndrome.¹⁵ Due to the significant risk of complications with premature closure, it is important to evaluate each patient individually when deciding which wound closure technique to perform. Although delayed primary closure is the prevailing method of wound closure in the literature, our patient was unique due to her late presentation, idiopathic nature of her compartment syndrome, and persistent edema following fasciotomy. Despite this unique wound closure plan, the patient's long-term follow-up showed promising results in terms of ambulation and physical abilities, as well as a high level of patient satisfaction.

Bulleted Learning Points

- Compartment syndrome basics.
- Recognizing compartment syndrome with high clinical suspicion.
- Wound closure options in compartment syndrome.

STATEMENT OF INFORMED CONSENT

The patient was informed and consented that data concerning the case would be submitted for publication.

Acknowledgments

This manuscript was previously accepted and presented at the 13th Annual MSU COGS Graduate Academic Conference 2021, Michigan Family Medicine Research Day, Michigan American College of Physicians Residents Day/Medical Students Day, and the Grand Rapids Spectrum Student Research Day.

Conflict of interest and funding

The authors have no conflicts of interest to declare. All co-authors have seen and agree with the contents of the manuscript. There is no financial interest to report.

REFERENCES

1. Tillinghast CM, Gary JL. Compartment syndrome of the lower extremity. In: Mauffrey C, Hak DJ, Martin MP III, eds. *Compartment syndrome*. New York: Springer; 2019, pp. 67–81.

2. Olson SA, Glasgow RR. Acute compartment syndrome in lower extremity musculoskeletal trauma. *J Am Acad Orthop Surg* 2005; 13(7): 436–44. doi: 10.5435/00124635-200511000-00003

3. Smith WR, Stahel PF, Suzuki T, Gabrielle P. Musculoskeletal trauma surgery. In: Skinner HB, McMahon PJ, eds. *Current diagnosis & treatment orthopedics*. 5th ed. New York: McGraw-Hill Education; 2013.

4. McQueen MM, Duckworth AD, Aitken SA, Court-Brown CM. The estimated sensitivity and specificity of compartment pressure monitoring for acute compartment syndrome. *J Bone Joint Surg*. 2013; 95(8): 673–7. doi: 10.2106/JBJS.K.01731

5. Valdez C, Schroeder E, Amdur R, Pascual J, Sarani B. Serum creatine kinase levels are associated with extremity compartment syndrome. *J Trauma Acute Care Surg* 2013; 74(2): 441–7. doi: 10.1097/TA.0b013e31827a0a36

6. Whitesides TE Jr, Heckman MM. Acute compartment syndrome: update on diagnosis and treatment. *J Am Acad Orthop Surg* 2013; 4(4): 209–18. doi: 10.5435/00124635-199607000-00005

7. McQueen MM, Gaston P, Court-Brown CM. Acute compartment syndrome. Who is at risk? *J Bone Joint Surg* 2013; 82-B(2): 200–3. doi: 10.1302/0301-620X.82B2.0820200

8. Wang J, Duan Q, Sun X, Mou X, Song B, Yuan, H. Acute compartment syndrome of the leg due to infection following an insect bite A case report. *Medicine (Baltimore)* 2018; 97(30): e11613. doi: 10.1097/MD.00000000000011613

9. Naidu KS, Chin T, Harris C, Talbot S. Bilateral peroneal compartment syndrome after horse riding. *Am J Emerg Med* 2009; 27(7): 901.e3–901.e5. doi: 10.1016/j.ajem.2008.11.009

10. Vajapey S, Miller TL. Evaluation, diagnosis, and treatment of chronic exertional compartment syndrome: a review of current literature. *Phys Sportsmed* 2017; 45(4): 391–8. doi: 10.1080/00913847.2017.1384289

11. Shaikh N, Barry M. Presentation of compartment syndrome without an obvious cause can delay treatment. A case report. *Acta Orthopædica Belgica* 2003; 69(6): 566–7.

12. Matziolis G, Erli HJ, Rau MH, Klever P, Paar O. Idiopathic compartment syndrome: a case report. *J Trauma* 2002; 53(1): 122–4. doi: 10.1097/00005373-200207000-00025

13. Kakagia D. How to close a limb fasciotomy wound. *Int J Low Extrem Wounds* 2014; 14(3): 268–76. doi: 10.1177/1534734614550310

14. Johnson SB, Weaver FA, Yellin AE, Kelly R, Bauer M. Clinical results of decompressive dermatomy-fasciotomy. *Am J Surg* 1992; 164(3): 286–90. doi: 10.1016/S0002-9610(05)81089-X

15. Hussmann J, Kucan J, Zamboni W. Elevated compartmental pressures after closure of a forearm burn wound with a skin-stretching device. *Burns* 1997; 23(2): 154–6. doi: 10.1016/S0305-4179(96)00090-3