Painless, Atraumatic, Isolated Lateral Compartment Syndrome of the Leg: An Unusual Triad of Atypical Findings

Luke S. Oh, MD, Paul B. Lewis, MD, Mark L. Prasarn, MD, Dean G. Lorich, MD, and David L. Helfet, MD

Abstract

Compartment syndrome can be a devastating complication with significant morbidity when not recognized or treated expediently. Among the classic pentad of signs and symptoms associated with compartment syndrome, pain that is out of proportion to the injury is often cited as the earliest and most sensitive.

We present a case report of an atypical presentation of compartment syndrome of the leg in which a patient taking lithium for bipolar disorder did not report pain out of proportion to the injury mechanism. Lithium has been implicated in altering pain perception and increasing the tolerance and threshold for pain, but this has not been widely reported in the orthopedic literature.

In addition to compartment syndrome that was painless, the patient presented with 2 additional atypical findings. She presented with compartment syndrome that was atraumatic and isolated to only 1 out of the 4 compartments of the leg. A compartment syndrome that is painless, atraumatic, and isolated to a single compartment represents an unusual triad of atypical findings that has not been previously reported.

With unusual presentations of compartment syndrome, there is an increased risk of late or unrecognized diagnosis, consequently increasing the likelihood of significant nerve damage or muscle necrosis. Clinicians have historically applied a higher level of scrutiny to patients who were deemed "obtunded," that is, those in whom an assessment of pain cannot be reliably determined. In the past, obtunded patients have included intubated or comatose patients, infants and children, mentally disabled patients, and patients with altered mental status, nerve injury, or distracting injuries. Based on evidence from the psychiatry and anesthesia literatures, we propose that patients taking lithium should be added to this list of "obtunded" patients in whom a reliable assessment of pain may not be possible.

Dr. Oh is Sports Medicine Fellow, Department of Orthopaedic Massachusetts General Hospital, Boston, Massachusetts.

Dr. Lewis is Orthopaedic Surgery Resident, Rush Medical College, Rush University Medical Center, Chicago, Illinois.

Dr. Prasarn is Orthopaedic Trauma Fellow, Dr. Lorich is Associate Director, Orthopaedic Trauma Service, and Dr. Helfet is Director, Orthopaedic Trauma Service, Hospital for Special Surgery and New York Presbyterian Hospital, New York, New York.

Address correspondence to: David L. Helfet, MD, Hospital for Special Surgery, 535 East 70th Street, New York, NY 10021 (tel, 212-606-1888; fax, 212-628-4473; e-mail, HelfetD@hss.edu).

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cute compartment syndrome is a potentially devastating condition in which the pressure within an osseofascial compartment rises to a level that decreases the perfusion gradient across tissue capillary beds, leading to cellular anoxia, muscle ischemia, and necrosis. Diagnosis is primarily clinical, supplemented by compartment pressure measurements. The clinical diagnosis of acute compartment syndrome is typically made on the basis of a classically known pentad of symptoms: pain, pallor, paresthesia, paralysis, and pulselessness. 1,2 Among the "5 P's" of compartment syndrome, pallor, paralysis, and pulselessness are considered to be late findings.^{1,2} It is well established that pain with passive stretch and pain that is out of proportion to the injury are the earliest, most sensitive, and reliable findings in diagnosing compartment syndrome. 1,2

We present a case report of an atypical presentation of compartment syndrome in which a patient taking lithium for bipolar disorder did not report pain with passive stretch or pain out of proportion to the injury mechanism. In the psychiatry and anesthesia literatures, there are reports suggesting that patients taking lithium have an altered perception of pain with often a higher threshold for pain.^{3,4}

In addition to compartment syndrome that was painless, this patient presented with 2 additional atypical findings. She presented with compartment syndrome that was isolated to a single compartment in the leg and without any history of trauma. Compartment syndrome that is painless, atraumatic, and isolated to a single compartment represents an unusual triad of atypical findings that has not been previously reported.

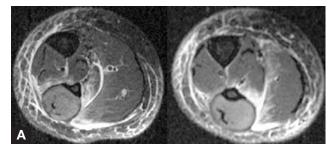
With unusual presentations of compartment syndrome, there is an increased risk of late or unrecognized diagnosis, consequently increasing the likelihood of significant nerve damage or muscle necrosis.⁵⁻²⁸ The goals of this case report are to examine the possible role that lithium may play in altering the pain threshold and to reinforce the need for a high index of suspicion in the diagnosis of compartment syndrome, especially in the setting of an atypical presentation. The authors have obtained written informed consent from the subject of this case report for the print and electronic publication of the text and accompanying images.

CASE REPORT

An obese woman in her mid-30s with bipolar disorder presented to the emergency room after developing a right footdrop 1 day prior to presentation. The patient recalled that her ankles were swollen and sore after wearing highheeled shoes 4 days earlier. She did not sustain any trauma and attributed her ankle discomfort to the fact that she wears high-heeled shoes infrequently and often feels uncomfortable after wearing these types of shoes. Despite her ankle soreness that lasted 48 hours, the patient was able to ambulate independently and did not take any narcotic pain medications. After the initial 48 hours, she reported a pain-free period but subsequently developed a footdrop. What prompted her to come to the emergency room was her footdrop, not her pain. There was no history of decreased sensation or paresthesia. At the time of the initial evaluation by the emergency room staff, the patient described an aching pain in the ankle that was 3/10 in severity. She attributed her ankle pain to a fall that she sustained a few hours earlier as a result of tripping over her "floppy foot."

On physical examination, the patient was observed to ambulate with a footdrop but without any pain. She demonstrated erythema and warmth over the anterior and lateral compartments of her leg with a superficial abrasion noted over the anterior compartment. This raised suspicion of possible early cellulitis. The patient reported that she sustained this "scratch" when she tripped a few hours earlier as a result of her footdrop. Palpation of the lateral compartment demonstrated only a slightly more swollen compartment compared with the contralateral leg. The anterior compartment was remarkably soft and nontender. Only a minimal increase in pain was reported with deep palpation and passive stretching of the tibialis anterior and peroneals. The patient had full strength of the gastrocnemius-soleus complex but little to no dorsiflexion or foot eversion. Sensation to light touch was intact over the plantar and lateral surfaces of the foot but decreased within the first dorsal webspace. Dorsalis pedis and posterior tibial pulses were readily palpable, and capillary refill was less than 2 seconds. Plain radiographs of the right lower extremity demonstrated no fracture or dislocations, and doppler examination revealed no evidence of deep vein thrombosis. Laboratory studies were unremarkable except for a significantly elevated creatine kinase (CK) level of 10,801 IU/L (normal range for a woman, 20-170 IU/L). To investigate a possible hematoma or infiltrate, the medical team ordered a computed tomography (CT) scan of the right lower extremity; this demonstrated a questionable abscess. At this time, the patient was admitted to the medicine service for treatment of cellulitis and/or abscess of her leg as well as further workup for her unexplained footdrop.

The patient's physical exam remained unchanged for the next 48 hours until she demonstrated decreased sensation over the distribution of the superficial peroneal nerve and increased swelling of the lateral compartment (all other compartments remained soft). Despite a palpable swelling of the lateral compartment, the patient had no complaints



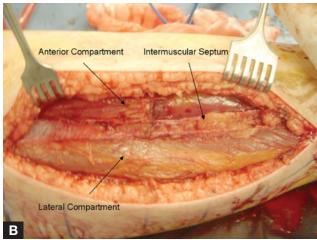


Figure. (A) Axial magnetic resonance imaging of the right leg clearly demonstrates an isolated tense lateral compartment. (B) Intra-operative photograph of the dusky, necrotic tissue in the lateral compartment compared with the healthy and normal anterior compartment muscles.

of increased pain with passive stretch of the lateral compartment. Magnetic resonance imaging was ordered by the medical team to further evaluate the questionable abscess noted on the CT scan, and it demonstrated increased signal intensity throughout the lateral compartment suggestive of muscle necrosis (Figure 1A). The orthopedic service was consulted regarding the development of a footdrop and increased swelling over the lateral side of the lower leg. At this time, the patient's lateral compartment was measured with a Stryker Intra-Compartmental Pressure Monitor (Stryker Corporation, Kalamazoo, Mich). The lateral compartment pressure was 92 mmHg and her diastolic pressure was 54 mmHg. Compartment pressures in the other compartments were normal. A repeat CK level was found to be 5,920 IU/L (normal range, 20-170 IU/L).

After the diagnosis of compartment syndrome was made, there were several important clinical considerations regarding the timing and utility of a fasciotomy at this time. Given the overlying cellulitis, there was a high risk for postoperative infection. Moreover, it was likely that the patient had a missed compartment syndrome with a potentially already necrotic compartment. The morbidity of an infected fasciotomy wound is significant, including amputation in cases of gangrene and multi-organ failure and sepsis. After weighing the risk and benefits as well as alternative treatment options, the patient provided informed consent and underwent an anterior and lateral

compartment fasciotomy. The anterior compartment demonstrated normal musculature and soft tissue, while significant muscle necrosis was found in the lateral compartment (Figure 1B). All nonviable tissues were débrided from the lateral compartment and the wound was covered with a vacuum-assisted closure (VAC) dressing. Postoperatively, intravenous clindamycin, zosyn, and vancomycin were administered after consultation with the infectious diseases team. A repeat irrigation and débridement was performed 4 days later. Plastic surgery was then consulted to evaluate the fasciotomy wound for primary closure versus the need for a soft-tissue flap. VAC dressing changes and frequent irrigation and débridment procedures resulted in sufficient wound granulation, and the patient avoided a need for skin graft or soft-tissue flap. The patient was discharged to a rehabilitation facility after a 40-day hospitalization.

DISCUSSION

Compartment syndrome is an orthopedic emergency that can result from a variety of causes, the most common being trauma. 1,2,29,30 Rarely, it develops without any history of trauma, and several etiologies for atraumatic compartment syndrome have been described.^{7,11,12,15,17,22,25,31,32} Compartment syndrome with an atypical presentation can result in a delay in diagnosis and treatment; therefore, a high index of suspicion needs to be maintained in these cases. 5,6,24,33 Hope and McQueen¹⁶ demonstrated that in cases without a fracture, there was a significantly greater delay to fasciotomy compared with those with a fracture. At fasciotomy, 20% of patients without a fracture had muscle necrosis requiring débridement compared with 8% of patients with a fracture. 16 Early fasciotomy not only improves patient outcome but also is associated with decreased indemnity risk.³³ Management of compartment syndrome in the modern era involves not only avoiding the sequelae of a missed diagnosis but also minimizing the risk of a malpractice claim. Bhattacharyya and Vrahas³³ reported that increasing time from the onset of symptoms to the fasciotomy was linearly associated with an increased indemnity payment. A fasciotomy performed within 8 hours after the first presentation of symptoms was uniformly associated with a successful defense.³³

The most common etiology for an atraumatic compartment syndrome is strenuous activity that may result in chronic exertional compartment syndrome (CECS). 34-37 In CECS of the leg, the anterior and deep posterior compartments are primarily affected. 34-37 However, there have been a few cases reported in the literature in which only the lateral compartment was involved. 8 In the majority of these cases, there was significant strenuous activity or an associated traumatic event, such as an inversion injury, fracture, or peroneal muscle rupture. 8,28 Our patient presented with a rare scenario in which increased pressures were isolated to the peroneal compartment without a strenuous or traumatic event. This combination of an atraumatic, nonexertional compartment syndrome isolated to the peroneal compartment is unusual and has not been reported previously.

When classic signs and symptoms are absent, there is an

increased risk for delayed diagnosis and treatment. 5,6,24,33 The presentation of our patient's compartment syndrome was unusual because not only was it confined to the lateral compartment of the leg and without history of trauma, but the most reliable signs of pain with passive stretch and pain out of proportion were not present. To our knowledge, there are no previous reports in the orthopedic literature of a compartment syndrome that lacked pain as a symptom, except in cases where an anesthetic nerve block or a patient-controlled analgesia pump was administered. 21,38,39 At first, the suggestion that an acute compartment syndrome can be painless seems heretical. However, a careful review of this patient's clinical presentation and psychiatric medication usage offers a possible explanation for her muted pain symptoms. Given that the patient arrived in the emergency room already having developed a footdrop, this case was likely a delayed presentation of a missed compartment syndrome.^{5,40-42} One may encounter absence of significant pain as a sequela of a late compartment syndrome.^{5,40-42} Therefore, the patient's rather mild symptomatology during her hospitalization is consistent with a late compartment syndrome. What is unique in this case, however, is that the patient denies ever having had pain that was out of proportion in the days preceding the emergency room visit. The patient recalls having had bilateral ankle swelling and soreness after wearing high-heeled shoes 4 days prior to hospital admission. She did not sustain any trauma and attributed her ankle discomfort to the fact that she wears high-heeled shoes infrequently and often feels uncomfortable after wearing these types of shoes. Despite her ankle soreness, she was able to ambulate independently and did not take any narcotic pain medications.

Since the patient had a long-standing history of bipolar disorder, it seems plausible that her psychiatric condition altered her pain perception and muted her pain symptoms. There are several reports in the literature of increased pain threshold in patients with psychiatric illnesses. 4,15,43-47 Studies have demonstrated that a patient with depression may respond differently to pain in different settings. 43,44 For example, in response to experimental pain such as thermal or electrical pain, patients with depression have been shown to demonstrate decreased sensitivity.⁴³ However, with regard to ischemic muscle pain, a study that performed pain testing (both threshold and tolerance) on 30 patients with major depression and matched controls demonstrated that patients with depression had an increased sensitivity to ischemic muscle pain.⁴³ The physiological basis of this phenomenon is unclear, but this study suggests that although depressed patients may demonstrate a high threshold and tolerance to heat and electrical pain, they will demonstrate a low threshold and tolerance for the type of pain that one may experience in compartment syndrome. 43 These results offer conflicting explanations and do not suggest that our patient's psychiatric condition per se was necessarily responsible for her muted pain symptoms.

Although our patient's psychiatric diagnosis and condition may not be directly associated with her altered pain

sensation, the medication that she was taking for her psychiatric condition may have had a significant effect. Several psychiatric medications, including lithium, have been implicated in increasing the pain threshold. 15,45 Our patient had taken moderate to high doses of lithium daily for the management of her bipolar disorder. In the psychiatry and pain management literatures, lithium has been known to alter pain perception and increase the tolerance and threshold for pain.^{3,4} In a clinical trial performed in the 1970s that would be difficult to replicate, Tosca and colleagues⁴ evaluated the pain threshold and pain tolerance to electrical stimulation in a group of 15 patients hospitalized for affective disorder and in a group of healthy hospital volunteers. There were two testing sessions performed 28 days apart. During the first testing session, all subjects were drug-free. For the second testing session, all subjects were treated with lithium and blood levels were measured. The results revealed that depressed in-patients had decreased pain perception at baseline compared with healthy hospital volunteers.⁴ After lithium administration, however, both groups demonstrated an increased tolerance to pain compared with their respective baseline. 4 The analgesic effect of lithium has also been demonstrated in studies performed in animal models.³ Unfortunately, a serum lithium level was not checked during our patient's hospital admission since the medication was previously always within therapeutic range, and the association with decreased pain threshold was discovered following discharge. These findings emphasize the need for greater scrutiny in patients taking lithium when evaluating them for possible compartment syndrome.

Given that the morbidity associated with a missed compartment syndrome is significant, clinicians have historically applied a higher level of scrutiny to patients who were deemed "obtunded," that is, those in whom an assessment of pain cannot be reliably determined. 1,2,29, 30,48 In the past, obtunded patients included intubated or comatose patients, infants and children, mentally disabled patients, and patients with altered mental status, nerve injury, or distracting injuries. 1,2,29,30,48 It seems that, now, patients taking lithium should be added to this list of "obtunded" patients. All of these patients represent a vulnerable group whose inability to demonstrate the hallmark symptoms and signs of the syndrome puts them in jeopardy of a late diagnosis of a compartment syndrome and its potentially devastating sequelae.5,40-42 Recognizing compartment syndromes requires having and maintaining a high index of suspicion, particularly in cases with a non-classic presentation. Diligent and frequent serial examinations are important for these types of patients, and there should be a lower threshold for measuring intracompartmental pressures and/or for performing fasciotomy. 1,2,29,30,48

CONCLUSIONS

The diagnosis of compartment syndrome in a non-classic presentation can be elusive. Our patient presented with an unusual triad of atypical findings of compartment syndrome: 1) increased pressure isolated to the lateral compartment, 2) no reported trauma prior to presentation, and 3) the absence of many of the classically known signs and symptoms of compartment syndrome.

There should be a high index of suspicion for compartment syndrome when paresthesia or loss of function is present, regardless of pain assessment. 4,15,45 Patients taking lithium should be considered "obtunded," similar to intubated or comatose patients, infants and children, mentally disabled patients, and patients with altered mental status, nerve injury, or distracting injuries. Additionally, when an orthopedic service is consulted to rule out a suspected compartment syndrome, the use of compartment pressure monitoring should be used more liberally. Early fasciotomy not only improves patient outcome but also is associated with decreased indemnity risk.³³

AUTHORS' DISCLOSURE STATEMENT

The authors report no actual or potential conflict of interest in relation to this article.

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