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Pathophysiology of an adolescent with compartment syndrome: a case report and review of the literature

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Abstract

Background Acute compartment syndrome occurs because of increased pressure within the fascial compartment, resulting in tissue ischemia. This surgical emergency, usually resulting from severe injury, is characterized by rapidly developing pain and swelling of the extremities. This case report aims to raise awareness among physicians by emphasizing the importance of high clinical sense in the diagnosis of compartment syndrome.

Case presentation This report describes a 25-year-old Saudi male who presented to the emergency department with a 1-week history of left leg pain. The pain suddenly worsened to severe anterior leg pain and swelling after he rapidly climbed multiple stairs. Acute compartment syndrome in the anterior compartment with rhabdomyolysis was diagnosed. Urgent fasciotomy was performed and the patient recovered gradually.

Conclusion This report presents an exceptional case of acute myonecrosis in a patient with non-sickle cell disease, involving acute compartment syndrome in the anterior compartment, and rhabdomyolysis, which led to compartment syndrome.

Keywords Case report, Emergency department, Orthopedic, Compartment syndrome, Acute myonecrosis, Fasciotomy

Introduction

Acute compartment syndrome (ACS) is a critical emergency in orthopedics and traumatology. It occurs due to increased interstitial pressure, also known as intracompartmental pressure (ICP), within a closed osteofascial compartment leading to pain by obstructing local circulation [1]. The legs are the most prevalent sites of ACS, followed by the forearm, arm, thigh, foot, gluteal area, hands, and abdomen [2].

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The incidence of ACS has been reported as 7.3 per 100,000 males and 0.7 per 100,000 females, with trauma accounting for the majority of cases. ACS is most frequently caused by tibial shaft fracture, which has a frequency of 1–10%. Young males under 35 years of age are at higher risk, likely owing to greater relative intracompartmental muscle mass and a higher likelihood of high-energy trauma. In addition, individuals with bleeding and hematological disorders, such as sickle cell disease (SCD) and hemophilia, are more susceptible. In pediatric leukemia, ACS cases without acute-triggering trauma have been documented [3–8].

This report presents an exceptional case of atraumatic acute myonecrosis in a patient without SCD, involving ACS in the anterior compartment and rhabdomyolysis, which led to compartment syndrome. Previous publications have reported similar presentations, primarily in

patients with SCD or in those without SCD but with ACS in more than one compartment. To our knowledge, few cases have been reported combining non-traumatic isolated compartment syndrome in patients without SCD.

Case presentation

A 25-year-old Saudi male with a known medical history of juvenile myoclonic epilepsy, for which he takes valproic acid, presented to the emergency room with a 1-week history of left leg pain that suddenly worsened to severe anterior leg pain and swelling.

The patient had been in his usual state of health until 1 week prior when he rapidly climbed multiple stairs at his university, triggering bilateral leg pain. Three days later, the pain was localized to the left leg, specifically over the shins, and rated a severity of 10/10. The pain was associated with notable swelling and inability to bear weight. There was no history of pain in other parts of the body, fever, change in urine volume or color, cough, runny nose, sore throat, shortness of breath, orthopnea, paroxysmal nocturnal dyspnea, chest pain, recent seizure episodes, trauma, or recent travel. The patient denied drug use.

On examination, the patient appeared well, with a thinly built body, but in severe pain. Vital signs were within normal limits. Inspection of the left leg revealed swelling over the leg and foot along with an erythematous rash compared with the right leg. Palpation of the left leg and foot revealed severe tenderness. Pitting edema was present over the left leg up to the midshaft. Sensation and vascularity were intact, confirmed by Doppler ultrasonography. There was no evidence of pallor, extensive erythema, swelling, blisters, calf swelling, tenderness over the deep vein territories, or joint effusion in the knee or ankle. While the range of motion was normal, pain was noted during dorsiflexion. Weight-bearing was possible but painful.

Laboratory investigations revealed elevated inflammatory markers with an elevated erythrocyte sedimentation rate (ESR) of 32 mm/hour (normal range: 0–13 mm/hour) and a C-reactive protein (CRP) level of 242.8 mg/L (normal range: 0–5 mg/L). Creatine kinase (CK) levels were elevated at 15,384 U/L (normal range: 26–174 U/L). Other laboratory results, including complete blood count, electrolyte levels, renal function, and coagulation profile, were within normal limits.

A bedside deep vein thrombosis ultrasound study of the lower limbs yielded negative results. Radiography of the left leg revealed no acute osseous abnormalities. The soft tissues were unremarkable, and alignment and joint spaces were maintained. Contrast-enhanced computed tomography (CT) of the left leg showed changes suggestive of underlying edematous alterations in the anterior compartment. No definite gas foci, vascular stenosis, or thrombosis was detected (Figs. 1 and 2).

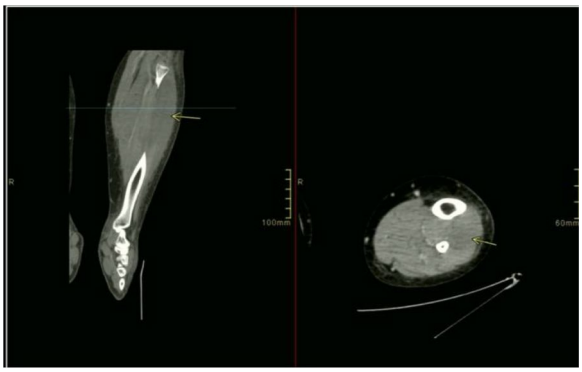

Fig. 1 An enhanced computed tomography scan of the left leg revealed diffuse enlargement of the anterior compartment muscle associated with decreased muscle attenuation (yellow arrow) likely indicative of underlying edematous changes

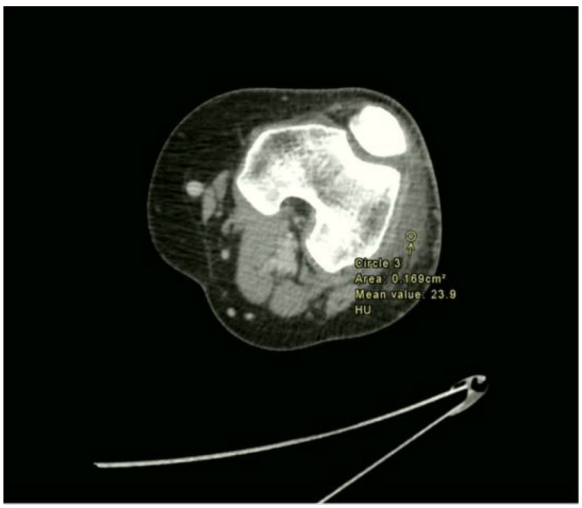

Fig. 2 Surrounding subcutaneous fatty stranding and free fluid (HU = 23.9 indicating fluid content)

Initially, the patient was treated for rhabdomyolysis with intravenous fluids and strong analgesia. Despite the intravenous administration of multiple doses of morphine, the pain continued to increase. Given the complexity and unclear etiology of the patient's symptoms, an orthopedic surgeon was consulted to facilitate a comprehensive evaluation and establish a diagnosis. The impression was left leg compartment syndrome, and an immediate fasciotomy was performed. Under general anesthesia, a 40-cm lateral longitudinal incision was made 3 cm posterior to the lateral edge of the tibia. All muscles in the anterior compartment were dusky, dark in color, not contracting, with no bleeding, and firm in

consistency, indicating muscle necrosis. The tibialis posterior, extensor digitorum longus, and extensor hallucis longus muscles were debrided and nearly completely removed. The wound was closed primarily without tension after 1 day, and the patient was discharged 2 days after the initial surgery.

After the procedure, the patient remained conscious with stable vital signs. He was able to walk with the assistance of a walker frame and was partially able to bear weight on the leg. Neurovascular status was normal and intact. The patient was referred for physical therapy and follow-up appointments for wound care and drainage.

One week after the fasciotomy closure, the wound on the left leg was examined and found to be clean and dry. The patient was able to walk with a cast boot and tolerably bear full weight on the leg. The neurovascular status of the leg was normal and intact. One month after the procedure, the patient was instructed to continue daily home exercises. Unfortunately, the patient did not continue follow-up, and no further information could be obtained.

Discussion

The pathophysiology of compartment syndrome is complex and several explanations have been proposed. One proposed mechanism suggests that intense eccentric exercise can damage muscle fibers, leading to the release of protein-bound ions such as CK, which increases osmotic pressure within the compartment. This causes muscle fibers to swell and increases the blood volume in the compartment. Consequently, rising pressure impedes blood flow, creating a cycle in which ischemia-related edema further increases compartment pressure, ultimately resulting in muscle necrosis. The common end result is cellular anoxia, thought to be due to compromised blood flow caused by elevated compartmental pressure, thereby affecting organ perfusion [9].

Pain is the most common symptom of compartment syndrome. It is considered the earliest and most sensitive sign, which is most likely preceded by a traumatic event [10]. However, ACS may also occur after minor trauma or non-traumatic events due to muscle necrosis [11]. Myonecrosis in compartment syndrome has been described in 16 reported cases in the literature, but all those patients had a medical history of SCD [12]. Few reported cases have extensively demonstrated compartment syndrome in patients without SCD. The clinical presentation of this patient was consistent with myonecrosis cases reported in the literature. The biopsy specimen obtained from the patient showed necrotic muscle tissue.

The lower leg is a common site of compartment syndrome and is composed of four compartments. The anterior compartment is the most frequently affected [13]. Conversely, the lateral compartment is rarely affected and is usually misdiagnosed as a muscle strain or contusion [14].

Isolated compartment syndrome is uncommon, irrespective of the anatomical location. Lateral compartment syndrome has been documented in only 15 cases in the literature [15], with normal CK levels reported in all but 1 case (1798 U/L) [16]. Table 1 lists the cases of healthy individuals who developed unilateral ACS following exercise, two of which were similar to our case. Our patient also had underlying rhabdomyolysis, evident by the high creatinine kinase (CK) level; this elevation is not usually seen in myonecrosis in previous literature.

Furthermore, anterior compartment syndrome can occur in the thigh, although less frequently than in the lower leg. The quadriceps muscles, including the rectus femoris, vastus lateralis, vastus medialis, and vastus intermedius, are located in the anterior compartment of the thigh. Increased pressure within this compartment can compress muscles, blood vessels, and nerves, resulting in tissue injury similar to anterior compartment syndrome in the lower leg [26]. Any of the three thigh fascial compartments may develop elevated pressure and muscle degeneration, leading to serious compartment syndrome of the thigh. The most frequent causes are vascular injuries with ischemic reperfusion, physical trauma with or without fracture, and spontaneous bleeding into myofascial spaces. Few cases of anterior compartment syndrome of the thigh have been reported in athletes, and it rarely occurs in inactive individuals, as in this case [21, 22, 25, 27–29].

The diagnosis of compartment syndrome with this unusual combination of myonecrosis, high CK level, atraumatic, and isolated compartment syndrome is challenging and can often go undiagnosed, given the atypicality of such a presentation [30].

Compartment syndrome is primarily a clinical diagnosis and requires a high index of suspicion, based on history and examination. Intracompartmental pressure measurement is the gold standard for diagnosis and can be used to aid in diagnosis when uncertain [13]. Given this patient's atypical signs and symptoms, a contrast-enhanced CT scan of the leg was performed.

Laboratory tests are not required to establish a diagnosis of compartment syndrome. However, a retrospective study conducted over a period of 10 years found that a CK level greater than 4000 U/L was 100% associated with compartment syndrome [31]. This finding was consistent with the elevated CK level in this case, thus establishing underlying rhabdomyolysis.

There is no standard management for this group of patients, given the rarity of their presentations. However,

Table 1 Summary of cases from the literature of unilateral acute compartment syndrome in healthy individuals following exercise

Ref	Age	Ssx	Hx	Compartment involved	Operation finding	Associated rhabdomyolysis; CK level if mentioned	Outcome	F/u
Nagy *et al.* [17]	26 M	Pain and numbness in the lateral left leg	Rugby training	Isolated peroneal lateral compartment	Viable muscles; bleeding and contracting on pinching	Yes; 4659 U/L	Pain improved, sensation return to normal, CK dropped	Complete return to normal sports activities in 9 months
Basnet *et al.* [18]	23 M	Left lower leg pain, numbness and swelling	Running a 5 km race	Left anterior and lateral compartments	No muscle rupture was found	Yes; 15,988 U/L	Ssx and CK level improved	NA
Griffith *et al.* [19]	50 M	Walking event during an army physical fitness test	Anterior leg pain with paresthesia to the dorsum of the right foot	Anterior and lateral compartments	No myonecrosis	NA	Ssx improved	5/5 strength but the same swelling and residual superficial peroneal neuritis after 1 year
Griffith *et al.* [19]	30 F		Right lower leg pain	Anterior and lateral compartments	Viable muscles	Yes; NA	Multiple repeated debridement almost losing 90% of the musculature	4/5 dorsiflexion strength to the left ankle and returned to physical activity, but required the IDEO brace at all times
Griffith *et al.* [19]	19 M	Ruck run	Heat injury with loss of consciousness	Anterior and lateral compartments	Myonecrosis	Yes; NA	Foot drop with 0/5 strength to the tibialis anterior	Back to physical activity, had 4/5 dorsiflexion strength, but required the use of an IDEO brace at all times
Shaikh *et al.* [20]	40 M	NA	Left lower leg pain	Isolated anterior compartment	Pink amorphous substance and no muscle necrosis	NA	NA	Pain free and improved sensation with a power of 4/5
McKinney *et al.** [21]	17 M	Athlete with strenuous workout	Right leg pain	Isolated anterior compartment	Muscle necrosis	Yes; 19,000 U/mL	Tibialis anterior longus (0/5), EHL (0/5)	After 4 months: improved tibialis anterior with dorsiflexion (4/5) only, with normal sensation
Jimenez *et al.* [22]	13 F	Athlete lacrosse player	Right leg pain	Anterior and lateral compartments	Thick fascia only	NA	Improvement in foot dorsiflexion and EHL strength	Symptom-free with full strength and no neurologic symptoms
Guenther *et al.* [23]	22 M	Military physical fitness test	Right leg pain	Four compartments	Nonviable anterior tibialis otherwise unremarkable	Yes; 9785 U/L	Severe pain, sensory loss and decreased strength, intact vascularity	NA

Table 1 (continued)

Ref	Age	Ssx	Hx	Compartment involved	Operation finding	Associated rhabdomyolysis; CK level if mentioned	Outcome	F/u
Paul *et al.* [24]	50 M	Played football	Right leg pain	Isolated anterior compartment	Bulged, dark brown with sluggish reaction on electrical stimulation	NA	Swollen dilated veins	Performing routine activities without complaints after 1 year
Yeom *et al.** [25]	48 F	NA	Leg lower leg pain	Isolated lateral compartment	Brownish and necrosis with the loss of activity of the muscles, compressed left common peroneal nerve	Yes; 56.9 ng/mL	NA	Pain free, and recovered function of the motor power of the left lower leg after 1 month

* This case involves the rare presentation of isolated compartment syndrome combined with rhabdomyolysis and muscle necrosis, which is similar to our case

Ref Reference, *Ssx* signs and symptoms upon presentation, *Hx* history, *CK* creatine kinase, *F/u* follow-up, *M* male, *F* female, *NA* not available, *EHL* extensor hallucis longus

immediate surgical fasciotomy is required to reduce ICP and prevent irreparable ischemic damage to muscles and peripheral nerves. Fasciotomy is not recommended 36 hours after injury, and ideally, it should be performed within 6 hours of injury [13, 32]. Data show that one-third of patients will regain normal limb function if the operation is performed within 12 hours of injury [13].

Conclusion

This report described a rare case of isolated ACS in the leg. Physicians should be aware that ACS can present atypically and that a high index of suspicion is crucial for timely diagnosis. Therefore, careful physical examinations, particularly when symptoms such as disproportionate pain, swelling, or neurological deficits are present, along with compartment pressure measurements when clinically indicated, are essential. Interdisciplinary collaboration among emergency physicians, orthopedic surgeons, and other specialists is important to ensure rapid intervention and optimal patient outcomes, highlighting the importance of team-based approaches in managing atypical ACS presentations. Further studies are required to elucidate the pathophysiology of myonecrosis in these patients.

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Author contributions
MA and NA designed and conceived the study. NA guided the development of the study. RA and TA contributed to drafting the manuscript. NA and MA undertook critical revision of the manuscript. All authors read and approved the final manuscript.

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Availability of data and materials
The datasets used during the current study are available from the corresponding author on reasonable request.

Declarations

Ethical approval and consent to participate
The study was approved by King Fahad Specialist Hospital—Saudi Arabia, and the patient's written consent was obtained.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests
The authors declare that they have no competing interests.

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