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Case Report

Spontaneous compartment syndrome of all limbs: A case report

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ABSTRACT

Compartment syndrome is a serious condition that results from increased pressure in a closed muscle compartment segment, which affects tissue perfusion and leads to necrosis. It can be acute because of fracture and crush injury or chronic due to recurrent transient increases in the intra-compartmental pressure that occurs during exertional activities. Rhabdomyolysis is a rare cause of compartment syndrome; clinicians should be aware of different atypical presentations of compartment syndrome and should have a low threshold for such a case. Early diagnosis and intervention may lead to better clinical outcome and may prevent any possible complication or future morbidity and disabilities. We are presenting a rare case of spontaneous atraumatic four-limb compartment syndrome in a patient post-minimal exertion, which was managed by urgent surgical fasciotomy and compartment decompression. 29 months follow-up; patient regained full function of both upper extremity without residual motor or sensory deficit, while he had bilateral foot drop without sensory deficit, and he was walking using bilateral ankle foot orthotic (AFO). We hypothesize that his compartment syndrome developed due to rhabdomyolysis, which was extenuated by dehydration.

Keywords: Compartment syndrome, Non-traumatic, Physical exertion, Rhabdomyolysis, Spontaneous, Surgical decompression

INTRODUCTION

Acute compartment syndrome is a surgical emergency that can be due to a traumatic fracture incident, crush injury, or any condition that creates acute edema or swelling, which in turn increases the compartmental pressure, the increase in pressure may lead to tissue hypo-perfusion leading to necrosis of muscles.[1] There are reported cases in the literature of acute compartment syndrome occurring outside of trauma scope and in the context of isolated medical conditions. These conditions included diabetes mellitus, the use of anticoagulants, hypothyroidism, leukemic infiltrations, and nephrotic syndrome.[2,5,7] These chronic conditions can cause edema and swelling in a given compartment, leading to compartment syndrome development.[3] There are some theories in multiple case reports correlating compartment syndrome with the use of medications such as non steroidal anti-inflammatory drugs (NSAIDs) (Diclofenac).[4] It is suggested that the impaired hemostasis and platelet dysfunction resulting from ingesting NSAIDs increases the incidence of spontaneous intramuscular bleeding and increased compartmental pressure.[4]

Compartment syndrome can present with different atypical presentation such as, Rhabdomyolysis. Clinicians should be aware of such presentations and should have a low threshold for such cases.

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Early diagnosis and continuous monitoring of compartment syndrome are of great importance in the management of pending compartment syndrome.

On the other hand, chronic compartment syndrome is mainly caused by recurrent transient increases in the intra-compartmental pressure that occurs during exertional activities and is relieved by rest. This presents with more vague symptoms, which poses a challenge for timely diagnosis and results in delayed diagnosis of such condition. [5] Treatment is usually surgical through compartment pressure release by fasciotomy of the affected compartment. [6]

In this study, we aim to discuss a rare presentation of compartment syndrome involving all four limbs compartments for a patient that presented to our institution in Doha, Qatar.

CASE REPORT

A 39-year-old male presented to the emergency department (ED) with a 2-day history of widespread body cramps and generalized weakness that started shortly after walking more than 2 kilometers outdoors in hot sunny weather. He stated that when he went for his walk and did not hydrate well. Shortly after he started walking, he developed severe bilateral leg and forearm cramps, which increased gradually until he was unable to bear weight on his feet. He noticed his urine became darker and decreasing in frequency since his complaints started. He did not have any history of trauma. Consequently, he presented to the ED seeking urgent medical advice. The patient's past medical history was significant for hypertension for 3 years, managed by Nifedipine.

On examination, the patient was found to be hypertensive, with a blood pressure of 175/95 mmHg that increased gradually after receiving 3.5 L of 0.45% normal saline at a rate of 125 ml per hour, reaching up to 210/110 mmHg. Both legs were tender, mildly to moderately swollen, and tense. The legs were elastic in consistency at the distal half of both calves (more so on the anterolateral aspect bilaterally). Pain could only be elicited with passive ankle dorsiflexion, but not with passive flexion/extension of his toes. The patient had an intact sensation in both legs, palpable dorsalis pedis artery bilaterally, and good capillary refill. Bilateral pulse oximetry showed an SPO_2 of 99%. The upper limb examination showed mild bilateral forearm swelling and mild tenderness.

Hematological Investigations showed hyperalbuminemia (56.0 gm/L, normally 3.4 to 5.4 g/dL), High Creatine Kinase level (19032 U/L, normally 22 to 198 U/L), Creatinine level of 142 umol/L (normally 74.3 to 107 umol/L), Myoglobin level of 5791 ng/mL (normally 25 to 72 ng/mL), as well as elevated liver enzymes. These results confirmed the diagnosis of Rhabdomyolysis for which treatment was started, and raised the suspicion of compartment syndrome of both lower limbs. The decision was taken to begin treatment for rhabdomyolysis and to keep him under close observation for compartment syndrome in all four limbs. This constituted limb elevation, ice pack application, and serial compartments assessment. A few hours later, the patient was reassessed after he started complaining of increasing pain in both forearms. He was unable to move his toes bilaterally, and his forearms were tense on compartment palpation. This was accompanied by difficulty in actively moving his fingers bilaterally. At that point, urgent fasciotomy and compartment decompression of the four limbs were carried out by the team as limb-saving procedures.

Intraoperatively, all four compartments of both legs were released through dual medial-lateral 10 cm long incisions [Figure 1]. The muscles bulged through the deep fascia as soon as they were released. Both forearm compartments were released through an incision that was made over the medial border of each ulna until all volar and dorsal compartments were ensured to be soft.

In the postoperative period, the patient was shifted to the ICU unit for follow-up. Postoperatively, clinical examination revealed palpable distal pulsations in all his limbs except the left dorsalis pedis artery; however, it was detectable using bedside Doppler. SPO2 of all limbs was 99%, with the exception of the left lower limb, which was 90%. Loss of active dorsiflexion of both feet was noticed. Both hands showed contracture-like stiffness (more so on the left hand), with painful active and passive movements of the fingers.

Serial examinations of all limbs around 6 hours following the surgery showed decreasing pain with passive movements and improved active finger movement with mild weakness in extension of the fingers remaining and persistent loss of active movements of his feet bilaterally.

Two days later, the patient was taken to the operative room for wound debridement and removal of necrotic tissue. four second look debridements were carried out over the following days, excising most of his anterior leg compartment bilaterally.

Twelve days later, all wounds were healing well except for the one on the right leg, which required secondary closure

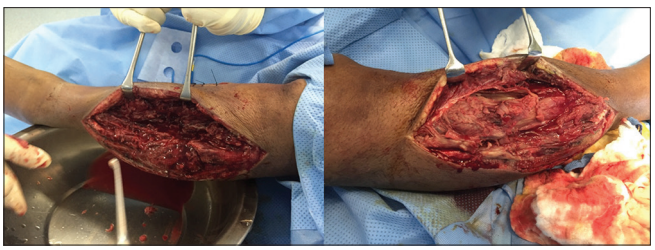

Figure 1: Intraoperative findings of bilateral lower extremity compartments.

Figure 2: Bilateral lower extremity compartments and right forearm compartment after a serial of debridement sessions.

[Figure 2]. At 29 months follow-up; he regained full function of both upper extremity without residual motor or sensory deficit. He had bilateral foot drop without sensory deficit, which is explained by loss of the anterior compartment and he was walking using bilateral ankle foot orthotic (AFO) [Figure 3].

DISCUSSION

We presented a case of a unique, spontaneous atraumatic four-limb compartment syndrome, which was explained by rhabdomyolysis. Overheat was the most likely cause of developing rhabdomyolysis; also, rapid rehydration of the patient may have contributed to the development of the compartment syndrome. There have been case reports in the literature describing similar presentations.

Riolo *et al.* in 2015 have reported a case of four-limb compartment syndrome in 49-year-old with a history of hepatitis B, chronic hepatitis C, and epilepsy; the patient developed four-limb compartment syndrome, and they concluded that it was due to rhabdomyolysis caused by multiple factors, the patient underwent urgent four-limb fasciotomy. [7] Also, Khan *et al.* had reported a similar case (only in one limb) of 41-year-old medically free lady who suffered from rhabdomyolysis and acute renal failure simultaneously, and the cause was attributed to inadvertent posturing while sleeping. [8] The patient developed bilateral spontaneous compartment syndrome in the leg, which was treated by fasciotomy. One of the most important steps in the treatment of rhabdomyolysis is the administration of intravenous fluids to avoid long-term damage to the kidneys, which may lead to fluid retention and increase interstitial edema. This may increase intra-compartmental pressure likely through further edema, ischemia and fluid sequestration. Furthermore, this, in turn, may lead to a closed cycle of increasing edema, and more muscle damage and necrosis. [7]

Dalton *et al.* described a case of acute spontaneous compartment syndrome in both forearms in a 31-year-old lady with no history of trauma presented to the ED with acute compartment syndrome in the forearm. Her past

Figure 3: The patient condition at final follow-up.

medical history was only positive for hypertension and coeliac disease. She underwent bilateral forearm fasciotomy, and her condition improved. [9] Although our patient had a history of recent hypertension for the past three years, it was most likely not contributing to his recent condition.

Davidson *et al.* reported a 31-year-old human immunodeficiency virus (HIV) positive male who presented to the ED complaining of bilateral leg pain, which was later diagnosed as bilateral acute compartment syndrome in the leg. [10] The patient underwent a two-incision leg fasciotomy in each leg that improved his condition, which was explained by the development of myositis caused by his use of antiretroviral medications.

A case of bilateral gluteal compartment syndrome in a 52-year-old patient following his right total knee arthroplasty was reported by Osteen *et al.* The patient underwent fasciotomy for both glutei, and later it was concluded that the cause of his condition was possibly due to the intraoperative length and positioning, with simvastatin-induced myositis. [11]

Interestingly, there is a case that was reported by Rohman *et al.* of a patient who suffered from recurrent episodes of compartment syndrome following blunt trauma to his thigh. The 31-year-old patient was treated for a femoral fracture when he was 18-year-old using an external fixator. He suffered from an incident of compartment syndrome

Table 1: All reported cases of multiple spontaneous compartments in the literature.

Case	Cause	Treatment
Sofat *et al*.[13]	Alcohol intoxication (bilateral legs)	Bilateral fasciotomies
Lynch *et al*.[14]	Exercise-induced (bilateral legs)	Bilateral fasciotomies
Ramdass *et al*.[15]	Simvastatin, thyroxine induced (bilateral legs)	Bilateral fasciotomies
Lu *et al*.[16]	Vibrio vulnificus infection (bilateral legs)	Bilateral fasciotomies
Figueras Coll *et al*.[17]	Methanol poisoning (bilateral legs)	Bilateral fasciotomies
Naidu *et al*.[18]	Horse riding (bilateral legs)	Bilateral fasciotomies
Ballesteros *et al*.[19]	Prolonged kneeling position during spinal surgery (bilateral legs)	Bilateral fasciotomies
Chin *et al*.[20]	Prolonged lithotomy position (bilateral legs)	Bilateral fasciotomies
Simon *et al*.[21]	Four-limb compartment syndrome associated with the systemic capillary leak syndrome	Four-limb fasciotomy
Khan *et al*.[8]	Inadvertent posture during sleep (bilateral legs)	Bilateral fasciotomies
Osteen *et al*.[11]	Prolonged intraoperative position and simvastatin use (bilateral glutei)	Bilateral fasciotomies
Davidson *et al*[10]	Antiretroviral myositis (bilateral legs)	Bilateral fasciotomies
Dalton *et al*.[9]	Hypertension and telmisartan use (bilateral forearms)	Bilateral fasciotomies
Riolo *et al*. (2015)[7]	Spontaneous four limb compartment syndrome	Four-limb fasciotomy
Current case	Spontaneous compartment syndrome of all limbs	Four-limb fasciotomy

nine months later and then suffered from another ten compartment syndrome incidents in the same thigh over the next ten years. He was treated with fasciotomies each time. [12] Table 1 includes all cases found in the literature with bilateral spontaneous compartment syndrome.

Acute compartment syndrome is a surgical emergency. The outcome of its management depends on multiple factors; the severity of the condition, duration of ischemia, pre-injury status and accompanying comorbidities, and, most importantly, the time of surgical intervention. [22] Fasciotomy that is delayed by more than eight hours from presentation usually accompanied by irreversible myoneural damage and late intervention in these cases may result in more harm for the patient than early intervention, as it increases the risk of developing multi-organ failure. [23] Additionally, fasciotomy delayed by more than 12 hours is associated with a higher infection rate. [24] Finally, fasciotomy wounds themselves have many complications like; pruritus, wound discoloration, recurrent ulceration, tethered tendons, muscle herniation, chronic venous insufficiency, and altered sensation within the margins of the wound. [25]

CONCLUSION

There are multiple cause which can lead to the development of compartment syndrome, clinicians should be aware of such different presentations and have a low threshold for such cases, as Early diagnosis and intervention may lead to better clinical outcomes and avoid any possible complications or future morbidity and disabilities. Rehydration in case of rhabdomyolysis should be done cautiously as it might lead to increase the risk of volume overload, leading to the possible development of compartment syndrome due to tissue edema. Monitoring of compartment syndrome is of great importance in the management of pending compartment syndrome. As in agreement with the literature, we believe that prompt surgical fasciotomy is the treatment of choice to improve outcomes and decrease morbidity and mortality.

AUTHORS' CONTRIBUTIONS

AK conceived the aims of this report and performed the decompression surgery and gave input into the final draft. AA and HN wrote the initial draft. IS wrote the final draft.

All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

ETHICAL APPROVAL

No ethical approval was required for the publication of this case report. The patient has provided his written consent for this case to be published.

Declaration of patient consent

The authors certify that they have obtained all appropriate patients consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published, and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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Conflict of interest

None of the authors authors have any conflicts of interest.

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