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Spontaneous bilateral compartment syndrome of the legs: A case report and review of the literature

T. Khan^{a,*}, G.H. Lee^a, A. Alvand^b, J.S. Mahaluxmivala^a^a Princess Alexandra Hospital, Harlow, Essex, UK^b University of Oxford, UK

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ABSTRACT

INTRODUCTION: Bilateral acute compartment syndrome of the legs is a rare presentation requiring emergent surgical intervention.**PRESENTATION OF CASE:** We report the case of 41-year-old woman who presented with acute bilateral compartment syndrome of the legs, complicated by rhabdomyolysis and acute renal failure.**DISCUSSION:** There are very few previously reported cases of bilateral compartment syndrome of the legs. In the present case, despite any clear causative factor, we suggest that the aetiology is related to inadvertent posture during sleep.**CONCLUSION:** The diagnosis of acute bilateral compartment syndrome of the legs requires a high index of suspicion, particularly in the absence of obvious aetiology. A successful outcome can be achieved with early diagnosis, prompt surgical intervention and a multidisciplinary approach.

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1. Introduction

Compartment syndrome is defined as increased tissue pressure in a closed facial space leading to progressive circulatory compromise of the nerves and muscles within the involved compartment.¹ Common causes of compartment syndrome include open or closed fractures, osteosynthesis, arterial injury, temporary vascular occlusion/revascularisation, burns, drug overdose and viral myositis.²

Bilateral compartment syndrome of the legs is a rare presentation with potentially catastrophic complications. The few cases reported in the literature all have a clear aetiology based on the patient history. We describe a unique case of spontaneous bilateral compartment syndrome of the legs, complicated by rhabdomyolysis and acute renal failure.

2. Case report

A forty-one year old woman presented to the Accident and Emergency Department with a 12-h history of bilateral leg swelling. She described awaking with difficulty in walking due to pain in both legs and had noted the passage of dark urine since earlier that day. There was no history of injury, alcohol or substance abuse. She had

no other co-morbidities, was not taking any regular medications including oral contraceptives and was a non-smoker.

Initial observations included a temperature of 36.6 °C, a respiratory rate of 21/min with 97% oxygen saturations on air, heart rate 95/min and blood pressure 149/106 mmHg. Examination of the legs revealed bilateral swelling with blisters below the knees. She had tense compartments on palpation, but passive movement of the ankles and dorsiflexion of halluces did not aggravate her pain. She described a subjective reduction in sensation below the knees bilaterally. Both dorsalis pedis and posterior tibial pulses were palpable and clearly audible on Doppler examination.

The initial impression of the emergency doctor was an allergic reaction due to the presence of blisters on both legs. However, upon review of the blood results and discussion with a senior colleague, a more sinister picture was suspected.

Initial blood results were: haemoglobin 19.3 g/dl, white cell count $19.9 \times 10^9/l$, creatinine 143 $\mu\text{mol/l}$, sodium 130 mmol/l, urea 10.7 mmol/l. Arterial blood gases showed pH 7.37, pCO₂ 4.50, pO₂ 12.74, base excess -6.4, lactate 3.25. The serum creatine kinase level was 374,800 IU/l.

After review by the orthopaedic team, a diagnosis of bilateral compartment syndrome of the legs with rhabdomyolysis and acute renal failure was made. The patient was taken to theatre immediately and underwent bilateral four compartment fasciotomies using two incisions on each leg. There was evidence of muscle necrosis and a thorough debridement was performed. The wounds were dressed appropriately. She was transferred to the Intensive Care Unit and required haemofiltration for worsening acute

* Corresponding author at: Department of Trauma & Orthopaedic Surgery, Princess Alexandra Hospital, Harlow, Essex, UK. Tel.: +44 1279827406; fax: +44 0186573764.

E-mail address: tanvirkhan@doctors.org.uk (T. Khan).

Table 1
Case reports of bilateral compartment syndrome of the legs.

Cases	Cause	Treatment
Chin et al. (2009) ⁵	Prolonged lithotomy position	Bilateral fasciotomies
Figueras Coll et al. (2008) ¹⁰	Methanol poisoning	Bilateral fasciotomies
Lu et al. (2008) ¹¹	Vibrio vulnificus infection	Bilateral fasciotomies
Lynch et al. (2006) ¹²	Exercise-induced	Bilateral fasciotomies
Ramdass et al. (2007) ¹³	Simvastatin, thyroxine induced	Bilateral fasciotomies
Naidu et al. (2009) ⁷	Horse Riding	Unilateral fasciotomy
Sofat et al. (1999) ³	Alcohol intoxication	Bilateral fasciotomies
Ballesteros et al. (2009) ¹⁴	Prolonged kneeling position during spinal surgery	Bilateral fasciotomies

renal failure. The patient was subsequently transferred to a tertiary centre for renal dialysis and split skin grafting (meshed autograft). Neurovascular supply to the limb was preserved.

3. Discussion

Previous case reports of acute bilateral compartment syndrome of the legs have highlighted various potential causes. These are summarised in Table 1. In comparison, our patient's history gives minimal clues regarding the aetiology. Similar clinical presentations of bilateral atraumatic compartment syndrome with rhabdomyolysis have been caused by alcohol intoxication³ and excessive exercise.⁴

There have been a number of case reports relating limb position to compartment syndrome. Prolonged surgical lithotomy position is a documented cause of acute bilateral lower limb compartment syndrome⁵ as well as bilateral gluteal compartment syndrome.⁶ Isolated bilateral peroneal compartment syndrome has been described after horse riding.⁷ It was suggested that flexion of the hip and knee while horse riding may obstruct the vascular supply to the lower limb. The authors reported that this can, in conjunction with ankle dorsiflexion, lead to a significant increase in peroneal compartment pressure. Furthermore, alteration in limb position has been demonstrated to cause variation in compartment pressure.⁸ However, the only other case in the literature of a patient presenting with leg compartment syndrome after prolonged decubitus position occurred unilaterally and involved a history of trauma.⁹ Therefore, we suggest that inadvertent positioning during sleep, causing pressure on the leg compartments is a potential cause of compartment syndrome in the present case.

In published reports of acute atraumatic bilateral compartment syndrome, the diagnosis has often been delayed, leading to negative sequelae. The presentation of our patient with normal vital signs, despite onset of rhabdomyolysis and acute renal failure was atypical. Blood test results in combination with a high index of suspicion for compartment syndrome, allowed early diagnosis. Furthermore, successful treatment of this patient's condition was possible due to a well-coordinated multi-disciplinary team approach. Early involvement of physicians and intensive care teams prevented further insult to the renal system. Finally, appropriate input from a tertiary centre with a specialist renal unit and reconstructive surgery department enabled good functional recovery with the patient managing well with activities of daily living and returning to her job as a shop assistant at final follow-up.

4. Conclusion

We report a unique case of spontaneous atraumatic bilateral compartment syndrome of legs complicated by rhabdomyolysis.

A high index of suspicion, early diagnosis and a multidisciplinary approach can avoid a potentially devastating outcome.

Conflict of interest

There are no conflicts of interest related to this paper or any of the authors.

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Ethical approval

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

Author contributions

T. Khan prepared the draft manuscript.

G.H. Lee collected details of the case and performed a detailed literature review.

A. Alvand critically analysed and edited the draft.

J. Mahaluxmivala as senior author revised the text and approved the final submission.

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