CASE REPORT

Spontaneous bilateral compartment syndrome in a HIV-positive patient

Donald James Davidson, Yasir Mehmood Shaukat, Reza Jenabzadeh, Chinmay M Gupte

Department of T&O, Imperial College Healthcare NHS Trust, London, UK

Correspondence to
Donald James Davidson,
d.j.davidson.02@cantab.net

SUMMARY

Spontaneous bilateral compartment syndrome is a very rare condition but one which requires swift diagnosis and urgent surgical decompression by fasciotomies in order to achieve the best outcome. We present the case of a 31-year-old HIV-positive man. The case highlights the perils of being sidetracked by an atypical clinical history instead of acting on the classical clinical examination findings. We will discuss the presentation and management of this patient, review the literature and highlight the key learning points. The most important learning point being that no matter how atypical the history, if a patient presents with limb pain out of proportion to the injury (with or without pain on passive stretch), sensory changes and a loss of motor power, then a diagnosis of acute compartment syndrome must be considered.

BACKGROUND

Acute compartment syndrome (ACS) is a surgical emergency that requires expedient diagnosis and immediate surgical decompression. Our case demonstrates the risk of overlooking the obvious clinical diagnosis by being sidetracked by an atypical clinical history. As far as we are aware this is the first case report in the literature of spontaneous bilateral compartment syndrome in a HIV-positive patient.

CASE PRESENTATION

A 31-year-old man presented to our accident and emergency (A&E) department via ambulance reporting of excruciating pain bilaterally in his legs. The patient had awoken that morning, 8 h earlier, with bilateral leg pain. The patient was unable to attend work, as a make-up artist, due to the pain which had progressed and was now unbearable. He described no previous similar episode and had been well the day before with no pre-existing malaise. On direct questioning, the patient denied any history of trauma and described no unusual or strenuous activity precipitating the pain; he did note, however, that he had undertaken slight exercise the evening before at his regular yoga class. The patient had a history of HIV (CD4 count $900 \times 10^6/L$, viral load $<20/L$) and hepatitis B infection. He had started Atripla (600 mg efavirenz, 200 mg emtricitabine and 300 mg tenofovir disoproxil fumarate) 3 months earlier and denied any other prescription, over the counter or recreational drug use.

The patient had first presented via ambulance to a neighbouring A&E department where the decision was made, due to the spontaneous bilateral nature of the symptoms and presumed vascular aetiology, to transfer the patient to our institution as it was suspected that he may require vascular surgical intervention.

On examination, the patient was in agony despite the administration of large doses of intravenous opiates. He was unable to move his feet or ankles due to pain which was exacerbated by passive stretching. He had tense posterior, anterior and lateral compartments with significantly decreased sensation in the dorsum and soles of his feet, bilaterally. Intact peripheral pulses were present with a capillary refill time of less than 2 s. A triphasic Doppler signal was evident bilaterally. The patient admitted having had blood taken but the results were not available on initial assessment.

Despite the atypical history (spontaneous, bilateral symptoms and no precipitating cause) it was evident given the examination findings that the patient had bilateral leg compartment syndrome which required immediate decompression. The patient was taken to theatre immediately. His compartment pressures were measured and his ΔP (the difference between diastolic blood pressure and compartment pressure) was significantly less than 30 mm Hg in each of his four leg compartments bilaterally. The patient had a bilateral two-incision four-compartment fasciotomy performed (figure 1). Evidence of muscle escape was apparent in each compartment confirming the diagnosis. The compartment musculature was bulging, pale and

Figure 1 Bilateral two-incision fasciotomy wounds at the time of the second look surgery. Note the oedematous musculature.

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oedematous, but did reperfuse, and there was no need for extensive early debridement of necrotic tissue.

Postoperatively, the results of the A&E department blood test confirmed rhabdomyolysis (creatinine kinase 200 332 IU/L (normal range 25–195 IU/L)) and this was coupled with a small amount of dark brown urine evident on passage of his urinary catheter. The patient developed an acute kidney injury (creatinine 460 µmol/L) and was admitted to intensive care where he received renal replacement therapy until his kidney function recovered.

During his admission no definite cause for his compartment syndrome was ever established despite comprehensive investigation. The histological samples taken at the time of the primary fasciotomies, the wound cultures and his myositis and metabolic myopathy screens were all unremarkable. The patient's care was coordinated with the input of numerous specialty teams including orthopaedics, plastic surgery, infectious diseases, rheumatology, nephrology and intensive care.

OUTCOME AND FOLLOW-UP

The patient spent 4 weeks in intensive treatment unit and was discharged 2 months after presentation. He had his bilateral fasciotomy wounds covered with split thickness skin grafts. His renal function improved (discharge creatinine 92 µmol/L). He was able to mobilise independently without any aids and he regained good movement in his feet and ankles. He did have some residual decreased sensation in the dorsum of his right foot, but the other neurological deficits fully recovered.

DISCUSSION

ACS is a limb-threatening, and occasionally life-threatening, condition characterised by increased interstitial pressure within an osteofascial compartment to a critical point resulting in reduced tissue perfusion leading to myoneural ischaemia.¹ The treatment for ACS is urgent decompression by fasciotomy. Delayed diagnosis has long been established as the main determinant of poor outcome including myonecrosis, contractures, functional impairment, amputation, renal failure and even death.^{1 2} Insufficient knowledge of the condition has long been associated with delayed diagnosis.²

ACS is most commonly caused by bony fracture, open or closed, which accounts for as much as 69% of cases.³ Other causes include crush injury, soft tissue injury, osteosynthesis, arterial injury, temporary vascular occlusion/revascularisation, burns, drug overdose and viral myositis.⁴ Principally, the diagnosis is made clinically in the presence of one or more signs: pain out of proportion to the injury (with or without pain on passive stretch), sensory changes and a loss of motor power.^{5 6} Compartment pressure can be measured directly or monitored in high-risk patients.² A differential pressure (compartment pressure subtracted from diastolic blood pressure (ΔP)) of less than 30 mm Hg is an indication for urgent compartment decompression.⁵

On review of the literature, it is apparent that spontaneous unicompartmental ACS is an uncommon presentation; known causes include viral myositis, hypothyroidism, leukaemic infiltration, nephritic syndrome, anticoagulation, diabetic muscle infarction and statin therapy.⁷ Bilateral ACS is rare and often associated with delayed diagnosis leading to negative sequelae.⁴ A handful of case reports exist describing bilateral ACS caused by prolonged operative positioning, exercise, alcohol intoxication, methanol poisoning, vibrio vulnificus infection and statin therapy.⁴ There is only one previous report of a spontaneous bilateral compartment syndrome and, although no definite cause

was identified, the authors suggested that inadvertent leg positioning while sleeping may have been the precipitant.⁴ HIV infection has been described in a small number of reports as a precipitant to ACS; in one case HIV-induced thrombocytopaenia precipitated acute forearm compartment syndrome after minimal trauma and in another ACS was triggered by HIV-related myositis.^{8 9} Myositis is a known side effect of anti-retroviral therapy, most commonly due to abacavir hypersensitivity, and antiretroviral-induced myositis has also been described in the presence of a coincident lactic acidosis.¹⁰

Our patient's presentation was very atypical: he had spontaneous compartment syndrome which was bilateral with no history of trauma or likely precipitant. It is known that ACS patients without a fracture have significantly greater time delays to fasciotomy and it is suspected that this is due to clinicians' lack of awareness of the risk.⁵ In this case, despite classical clinical examination findings, the patient was transferred from one institution to another delaying diagnosis and treatment; this highlights the perils of being sidetracked by an atypical story.

In conclusion, the presentation of spontaneous bilateral ACS in a HIV-positive patient is rare. However, no matter the underlying pathology or how atypical the presentation, clinical examination findings must dictate management in ACS with the establishment of a swift diagnosis, permitting early fasciotomy to achieve the best possible outcome.

Learning points

- ▶ No matter how atypical the history, if a patient presents with limb pain out of proportion to the injury (with or without pain on passive stretch), sensory changes and a loss of motor power, then a diagnosis of acute compartment syndrome (ACS) must be considered.
- ▶ Early recognition and swift intervention are necessary to achieve the best outcome in ACS.
- ▶ Rhabdomyolysis must be considered in patients presenting with ACS and again early recognition and intervention improve outcome.
- ▶ Where the presentation for ACS is atypical a multispecialty approach to patient care is advocated for expert input into the establishment of investigation and management plans.

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Competing interests None.

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