CASE REPORT

Recurrent spontaneous compartment syndrome of the thigh

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SUMMARY

Acute compartment syndrome (ACS) is a surgical emergency, in which tissue pressure becomes greater than perfusion pressure leading to tissue ischaemia. It is typically a consequence of trauma. We present a case in which a patient suffers blunt trauma to the thigh, but develops ACS 2 years after this injury and consequently endures 10-episodes of ACS (no precipitating event or cause) in the same thigh over 10 years. On the 10th presentation the patient was found to have arteriovenous malformation on MR angiography which were embolised and the fasciotomy wound closed with a split-thickness skin graft. A thorough literature search deemed this case to be the first reported recurrent spontaneous ACS of the thigh. Prompt recognition and treatment of ACS is vital. Clinicians should thoroughly investigate such patients postoperatively and involve vascular/plastic surgeons and interventional radiologist to provide optimum care and prevent recurrence.

BACKGROUND

Acute compartment syndrome (ACS) is a surgical emergency. It is a condition where pressure within an osseofascial compartment rises, leading to a vicious cycle of bleeding, oedema and inflammation further increasing compartment pressure as the compartment is closed with progressive neurovascular compromise and eventual tissue death.¹ The aetiology of this condition can vary from open/closed fractures to burns to viral myositis.²

Though thigh compartment syndrome is less common than in the lower limbs, there have been many reported cases over the past 20 years but only one case has been identified within the literature where there was no precipitating factor such as trauma, illness or coagulopathy.³ A recent systematic review cites blunt trauma as the commonest cause of thigh compartment syndrome.⁴ However, the term 'spontaneous' compartment syndrome (in the absence of a clear precipitating event) has been used rather frequently in the literature to suggest a non-traumatic cause such as diabetes mellitus, hypothyroidism, nephrotic syndrome and secondary to medication.⁵⁻⁸

A thorough search of the literature did not reveal any cases of recurrent spontaneous compartment syndrome of the thigh. We present a case in which a patient indeed suffers blunt trauma to the thigh, but only develops compartment syndrome 2 years after this injury and consequently endures 10 episodes of spontaneous (no precipitating event or

cause) compartment syndrome in the same thigh over a 10-year period.

CASE PRESENTATION

A 31-year-old man presented with excruciating sudden onset, a traumatic right thigh pain and swelling preceded by a popping sensation.

The patient reported suffering a right femoral fracture during a game of rugby when he was 18 which was treated with internal fixation. He subsequently had removal of metal work and 9 months later had an episode of compartment syndrome which was treated conservatively. Since then he had a further 10 episodes of right thigh spontaneous compartment syndrome (including this episode) over a 10 year period with nine fasciotomies with subsequent delayed primary closure. While living in Bournemouth, UK the patient had been extensively investigated by vascular/orthopaedic teams, with no cause found.

The patient had no other medical/family history and did not take any regular medication. On examination, the patient objectively appeared to be in extreme pain, the right thigh swollen and tense with reduced movement at the knee, however, the limb was neurovascularly intact. The differential pressure between the compartment and diastolic blood pressure consistently measured <30 mm Hg.

Routine blood tests and xrays were unremarkable.

He then underwent his 10th fasciotomy (figure 1), decompression of the anterior compartment and removal of a large haematoma, with subsequent vac dressing application and then split-thickness skin grafting.

The patient then had MR angiography, revealing abnormal peripheral vessels in the right lateral thigh were identified, arising from both profunda and superficial femoral artery (SFA). Thus limited embolisation was carried out.

Figure 1 Fasciotomy wound. Change of vacuum dressing with surrounding signs of previous fasciotomy scars.

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OUTCOME AND FOLLOW-UP

The patient was followed up at 6 weeks and at 12 weeks with no further recurrence.

DISCUSSION

In the case presented above, our patient had a history of recurrent, spontaneous ACS to his right thigh. A thorough search of the literature revealed few individual cases of recurrent ACS of lower leg and upper limbs, but did not reveal any recurrent spontaneous ACS. These reported cases are often related to recurrent trauma or insult to the previous site of fasciotomy, for instance, post mid-shaft tibia fibula fracture, upper arm ACS after blunt trauma or cellulitis, forearm ACS association with chronic osteomyelitis, foot ACS following a calcaneal fracture repair and hand ACS.^{9–12} Some of these cases are summarised in table 1 below.

ACS occurs when the pressure within a closed fascial compartment rises to a point where tissue perfusion to the affected compartment is compromised.¹³ This can occur in areas of the body where there is limited or no capacity for tissue expansion.¹⁴

In contrast to the lower leg where muscle compartments are tightly enclosed by the muscle fascia, cases about compartment syndromes of the thigh are uncommonly reported in the literature.⁵ Khan *et al* summarised all ACS in the thigh reported in the literature. The mentioned aetiologies include blunt trauma, trauma to skeletal and muscle, penetrating trauma, postprocedure, tumour infiltration, drug-induced, exercised-induced, coagulopathy and snake bite.³ Among these factors, blunt trauma was the most common cause of ACS.⁴

Yet when compartment syndromes of the thigh do arise, early recognition and treatment is essential due to the potential for serious morbidity and mortality involved. Schwartz *et al*¹⁵ reviewed 21 cases of ACS of the thigh and reported a mortality rate of 47%, and significant long-term morbidities including sensory deficit and motor. Though the majority of ACS are decompressed on clinical findings, McQueen and Court-Brown demonstrated that ‘use of a differential pressure of 30 mm Hg as a threshold for fasciotomy led to no missed cases of acute compartment syndrome’.¹⁶

Although our patient had trauma to his right thigh when he was aged 18, his subsequent recurrent compartment syndromes first developed 2 years after his initial injury and each episode without a precipitating cause.

Indeed, spontaneous ACS are a rare occurrence and have only been reported in individual patient cases in the literature. Apart from one case reported from Khan *et al* of a single episode of idiopathic spontaneous thigh compartment syndrome, the

Figure 2 Embolisation with coils of superficial femoral artery branch and distal part profunda vessel.

others were related to non-traumatic causes. The associations of which included: simvastatin-induced myositis, long-term anticoagulation, post-thrombolytic therapy and muscle infarction in patients with type one diabetes mellitus.^{5–8 13}

Despite our patient having none of the above risk factors, our patient had multiple, spontaneous thigh ACS. After undergoing an urgent fasciotomy, vascular surgeons and interventional radiologists were involved postoperatively. A diagnostic MR angiogram was carried out due to the main intraoperative finding of haematoma within the anterior thigh compartment. Several abnormal vessels arising from the lateral profunda and SFA branches were identified, and embolised using coils with satisfactory occlusion and relief of discomfort (figure 2). We suspected that bleeding from an AV fistula may have been the cause for the haematoma found, giving rise to spontaneous ACS. Arteriovenous malformations (AVM) give rise to direct connections between the arterial and venous system without an intervening capillary system (figure 3).

Bail *et al*² described a patient with 3 months gradual onset of swelling and discomfort in the right thigh after sustaining a blunt trauma. Postoperative angiography demonstrated several

Table 1 Reported cases of recurrent acute compartment syndrome

Authors	Patient's initial injury and fasciotomy	Time to recurrent ACS	Injury/insult leading to the compartment syndrome	Compartments involved for the recurrent ACS
Gaskill <i>et al</i> ⁹	Exercise-induced compartment syndrome (EICS)—4-compartment fasciotomy of lower leg	Months	Mid-shaft tibia fracture and fibula fracture	4-compartment fasciotomy
Gaskill <i>et al</i> ⁹	Bilateral forearm fasciotomies secondary to a crush injury	1 year	Significant swelling following infected cat bite	Forearm 3-compartment fasciotomy
Hanypsiak <i>et al</i> ¹⁰	Midshaft synostosis fracture—4-compartment fasciotomy of lower leg	1 year	Blunt injury following a kick	Anterior, deep posterior and lateral compartment fasciotomy of lower leg
Goldie <i>et al</i> ¹¹	Mid-shaft fracture of the radius and ulna—forearm fasciotomy	12 years	Swelling and collection secondary to chronic osteomyelitis	Forearm fasciotomy

ACS, acute compartment syndrome.

Figure 3 MRI of arteriovenous malformations in left thigh (taken from radiopedia.com).

AV-fistulae in the distal internal iliac artery which the authors believe were the cause of the haematoma. Bail *et al*² highlighted the importance of being aware of possible previously, undiagnosed AV-fistulae and AV-malformations as a cause of post-trauma haematoma formation which can potentially lead to ACS requiring immediate surgical treatment.

Despite this, when patients acutely present with a haematoma in the thigh, bleeding from an AV-fistulae in the pelvis or thigh should not be considered as a common causation, but rather an atypical one.² Likewise Lowe *et al* discussed the importance of having a systematic approach for formulating differential diagnoses of a painful and swollen limb in the emergency setting. In addition to ACS, these include fractures, acute ischaemia, thrombosis, life-threatening infections such as necrotising fasciitis and gas gangrene.¹⁷ These are surgical emergencies as important as ACS that should not be overlooked.

While ultrasonography and measuring intracompartmental pressures may be helpful diagnostic adjuncts where a definitive diagnosis cannot be immediately reached, careful selection of relevant investigations along with good understanding of pathophysiology is required to avoid delaying potentially limb-saving surgical treatment. As irreversible muscle necrosis can occur as early as 3 h after onset of ischaemia.¹⁴ Mabvuure *et al*¹⁴ stressed the importance of emergent fasciotomy where clinical features are clear.

It is debatable whether abnormal vessels seen on the latest angiography postoperatively in our patient were as a result of new AV-fistula formation secondary to multiple previous surgical procedures in the same compartment or whether these may have been pre-existing malformations as described by Bail *et al*. Nonetheless these were not seen on past imaging.

Following surgery, the fasciotomy wound was managed using vacuum dressing (VAC) and the plastic surgery team were involved achieving closure of the wound by using a split thickness skin graft. Many different methods of managing fasciotomy wound have been described in the literature, and yet none of these are proven to be more superior than the others.^{18 19} Yang *et al*²⁰ reported that patients treated with vacuum dressing have shorter time between initial fasciotomy and definitive closure compared with gauze packing.¹⁸

Although there are still uncertainties to be explained with regards to the recurrent nature of our patient's thigh ACS, it

was apparent from our case that management of ACS does not only involve the orthopaedic team. In fact, it involves the joint efforts of the multidisciplinary team from the outset. By having prompt initial recognition and referral by the emergency physicians and seeking the expertise of orthopaedic surgeons, anaesthetists, vascular surgeons, interventional radiologists and plastic surgeons postoperatively all vastly contributed to a better patient-centred outcome.

Learning points

- ▶ Acute compartment syndrome (ACS) of the thigh is less common than other areas of the limbs, but clinicians should remain vigilant and admit all suspicious cases for bed rest, elevation and a period of observation.
- ▶ A thorough history, examination and routine blood tests should be performed as there are many non-traumatic causes of ACS.
- ▶ Compartment pressures should be taken early and periodically.
- ▶ Involve experts within the multidisciplinary team early.
- ▶ Urgent fasciotomy should be carried out if clinical suspicion is high and not be delayed by investigations.

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Competing interests None.

Patient consent Obtained.

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