Spontaneous lower limb subcutaneous emphysema: a diagnostic dilemma

Introduction

Subcutaneous emphysema is defined as the presence of air or gas in the soft tissues of the body (Chu and Glare, 2000). It is a rare and often self-limiting entity where patients can commonly remain asymptomatic (Chu and Glare, 2000). The pathogenesis is multi-factorial with an infective aetiology most commonly implicated as a result of conditions such as gas gangrene (clostridial myonecrosis), necrotizing fasciitis or crepitant cellulitis.

A pathological process that predisposes to a communication between a gas-filled viscus and the subcutaneous tissues such as penetrating trauma, pneumothorax, ruptured oesophagus, bowel perforation, barotrauma or air injection injuries may also result in subcutaneous emphysema (Doweiko and Alter, 1992).

This article describes a diagnostic dilemma in a patient with multiple medical comorbidities where clinical assessment elicited signs of spontaneous subcutaneous emphysema. The combination of a sudden increase in pain, rapid progression of crepitus and persistently elevated inflammatory markers suggested a diagnosis of necrotizing fasciitis. However, contrary to the normal clinical presentation of necrotizing fasciitis, there was no rapid progression to systemic toxicity and there were minimal skin changes with no evidence of

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Case Report

A 62-year-old man presented with a 1-day history of a painful swollen right calf and thigh. He had a previous deep venous thrombosis in the same calf 4 months earlier and had been commenced on warfarin. There was no history of trauma to the leg. He had no fever, sweating or rigors. He had a history of advanced metastatic prostatic carcinoma with obstruction of his lower urinary tract requiring bilateral ureteric stents and a permanent suprapubic catheter. A recent bone scan had also revealed widespread bony metastases. He had previously received radiotherapy to his cervical and lumbar regions and had been commenced on dexamethasone. His past medical history included type 2 diabetes mellitus and hypertension. On examination he was apyrexic and haemodynamically stable. He was well nourished but dehydrated. He had a tender erythematous swollen leg from the mid-calf extending to the groin and bilateral pitting oedema. The feet were well perfused. The rest of the examination was unremarkable.

1. What is the differential diagnosis and what investigations would you request?

Haematological analyses showed a white cell count of 18.2 x 10^9/litre, C-reactive protein of 150 mg/litre, urea of 23.9 mmol/litre, creatinine of 143 μmol/litre, D-dimer 15.19 μg/ml (0–0.5 μg/ml normal range), and a sub-therapeutic international normalized ratio of 1.4. A Doppler ultrasound scan of his right lower limb revealed thrombus within the right femoral vein with no proximal extension. He was rehydrated and commenced on therapeutic enoxaparin until his warfarin levels were optimized to a therapeutic level.

The patient described an increase in right lower limb pain on day 7 post-admission. On clinical assessment, the right leg remained swollen and erythematous with maximal tenderness elicited in the right calf. However, crepitus was present and extended proximally from the calf to the thigh. There was no evidence of any wounds, skin necrosis or vascular compromise. Haematological analysis confirmed a persistent inflammatory response: white cell count of 16.5 x 10^9/litre and C-reactive protein of 226 mg/litre. A plain X-ray of the right knee showed extensive gas within the soft tissues (*Figure 1*).

2. What are the main clinical concerns and how would you manage this patient now?

A provisional diagnosis of necrotizing fascitis was made. As the patient remained afebrile and haemodynamically stable, on the background of multiple medical comorbidities, he was treated conservatively with intravenous benzylpenicillin, clindamycin and ciprofloxacin and was monitored regularly overnight for evidence of necrosis or systemic sepsis. Blood cultures yielded coagulase-negative staphylococci. The pain reduced over the following 48 hours and his general condition gradually improved. A magnetic resonance imaging scan demonstrated multiple large abscess cavities and gas collections within the right thigh extending through the sciatic notch into the pelvis with involvement of all muscle compartments in the right thigh (*Figure 2*).

3. What management options are available at this stage?

Although the patient had made some clinical improvement, operative exploration was deemed appropriate based on superficial skin deterioration and ascending radiological sepsis. Several separate incisions were made in the right thigh and buttock over the areas of maximal crepitus. There was significant oedema combined with the presence of gas within the subcutaneous tissues and around the mid-superficial femoral artery. There was no obvious necrosis or collection identified (*Figure 3*). Cultures of wound, tissue fluid and muscle biopsies demonstrated staphylococcal and enterococcal isolates. The patient continued on intravenous antibiotics for the following 6 days. Unfortunately his condition deteriorated on day 18 post-admission and he died of multiorgan failure secondary to respiratory tract and lower limb sepsis.

Figure 1. Plain lateral view X-ray of right knee demonstrating subcutaneous emphysema (SE).

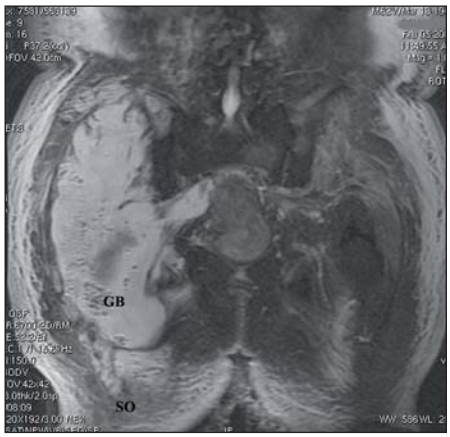

Figure 2. Coronal short tau inversion recovery (STIR) image of the pelvis showing a large right gluteal fluid collection containing gas bubbles (GB) and communicating with the pelvis via the sciatic notch. There is extensive subcutaneous oedema (SO).

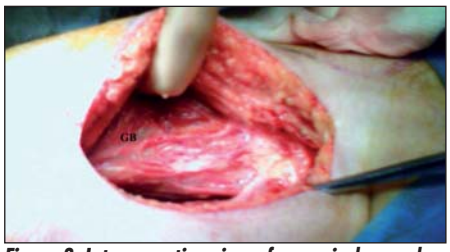

Figure 3. Intraoperative view of a surgical wound in the right thigh demonstrating gas bubbles (GB) within the tissues but no evidence of abscess formation or tissue necrosis.

subcutaneous fluid collections. This article presents a teaching case report and discusses the diagnostic difficulties associated with such cases.

Discussion

Cellulitis can usually be treated successfully with antibiotics alone. However, early and decisive surgical exploration and debridement for necrotizing fasciitis is paramount and is associated with improved clinical outcomes (Schmid et al, 1998).

Crepitant cellulitis differs from more serious soft tissue infections through an abundance of soft tissue gas, lack of marked systemic toxicity, minimal skin changes, a more gradual onset, less severe pain and absence of foul-smelling discharge (*Table 1*) (Mader and Calhoun, 1996). The Laboratory Risk Indicator for Necrotising Fasciitis (LRINEC) score was developed to distinguish between necrotizing fasciitis and other soft tissue infections using routine laboratory tests (*Table 2*) (Wong et al, 2004). Wong et al (2004) showed that a LRINEC score of 6 or above was associated with a positive predictive value of 92% and a negative predictive value of 96% for necrotizing fasciitis.

Plain film radiography will often demonstrate the presence of gas within the soft tissues in necrotizing soft tissue infections (Headley, 2003). However, absence of soft tissue gas does not exclude such infections (Headley, 2003). In the detection of tissue inflammation and necrosis, magnetic resonance imaging (MRI) is superior to ultrasonography or plain film radiography (Headley, 2003). Schmid et al (1998) reported that necrotizing fasciitis can be excluded when no deep fascial involvement is revealed. They also discussed that MRI tends to overestimate the extent of deep fascial involvement which was an important factor in this patient's case.

Table 1. Common clinical features of necrotizing soft tissue infections

Clinical feature	Crepitant cellulitis	Necrotizing fasciitis	Clostridial myonecrosis (gas gangrene)
Onset	Gradual	Acute	Acute
Sepsis	Mild	Moderate to severe	Severe
Pain	None or mild	Moderate to severe	Moderate to severe
Skin change	None or mild cellulitis	Pale red cellulitis	Dusky, swollen
Exudate	None or slight	Serosanguinous or foul-smelling	Serosanguinous
Gas	Abundant	None or minimal	None or minimal
Muscle necrosis	Minimal	Significant	Severe
Mortality	Uncommon	Common	Common

Table 2. Laboratory Risk Indicator for Necrotising Fasciitis (LRINEC) score

Variable (units)		Score
C-reactive protein (mg/litre)	<150	0
	>150	4
Total white cell count (per mm^3)	<15	0
	15–25	1
	>25	2
Haemoglobin (g/dl)	>13.5	0
	11–13.5	1
	<11	2
Sodium (mmol/litre)	>135	0
	<135	2
Creatinine (μmol/litre)	<141	0
	>141	2
Glucose (mmol/litre)	<10	0
	>10	1

A score >6 should raise suspicion of necrotizing fasciitis, and a score >8 is strongly predictive of necrotizing fasciitis.
From Wong et al (2004)

Wang and Hung (2004) reported the use of transcutaneous soft tissue oximetry as a novel method to assist in the rapid diagnosing of necrotizing fasciitis by differentiating it from cellulitis. Frozen section biopsy has been used successfully in providing a diagnosis before surgery (Stamenkovic and Lew, 1984). However, this invasive method relies on immediate histological evaluation (Stamenkovic and Lew, 1984). Therefore, the authors suggest mandatory surgical exploration and tissue biopsy to definitively distinguish myonecrosis or necrotizing fasciitis from cellulitis. In cellulitis, the muscle will be macroscopically healthy with minimal tissue necrosis in the superficial tissues.

Despite a provisional diagnosis of necrotizing fasciitis associated with a LRINEC score of 7, consideration of both the patient's existing life-limiting illness and the absence of necrotic changes or septic shock resulted in initial conservative management. When the MRI scans supported the clinical signs of progressive proximal infection combined with the LRINEC score of 7, surgical exploration was then deemed mandatory regardless of clinical progress. Amputation is sometimes necessary in necrotizing soft tissue infections but in this case it was not appropriate because of the patient's intercurrent disease and the proximal extension of infection into the pelvis.

Conclusions

This case highlights the difficulties in differentiating between immediately life-threatening conditions such as necrotizing fasciitis and other soft tissue infections such as cellulitis. It is not practical to submit every such patient to frozen section biopsy or MRI and the clinician should make a judgement to proceed to these investigations based on clinical and diagnostic information at the bedside. Management of suspected necrotizing soft tissue infections requires prompt and aggressive surgical debridement combined with appropriate antibiotic therapy.

Although crepitant cellulitis may have a more salient course, surgical debridement may still be required, as antibiotics will have diminished activity in any necrotic areas. Surgery may also prevent compartment syndromes caused by pressure from gas production or facilitate the removal of a foreign body. Despite such aggressive measures, some patients will not respond and deaths occur (Al-Wakeel et al, 1995). **BJHM**

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