Reminder of important clinical lesson

Medical cause of compartment syndrome: a fatal case of *Clostridium septicum*Oleksa Rewa,¹ Christopher A Smith²¹Department of Critical Care, Sunnybrook Health Sciences Center, University of Toronto, Toronto, Ontario, Canada²Department of Medicine, Etherington Hall Queen's University, Kingston, Ontario, Canada

Correspondence to Dr Christopher A Smith, cas12@queensu.ca

Summary

A 57-year-old woman with non-Hodgkin's lymphoma presented to the emergency department with febrile neutropaenia. She complained of fever and malaise and had a normal physical examination. Ten hours later, she developed worsening right leg pain that required escalating doses of morphine. Her physical examination remained unremarkable. Owing to the increasing pain in her leg, an x-ray was performed which revealed subcutaneous emphysema. She subsequently developed numbness over her right leg and, then, 21 h after admission the leg became cold and ischaemic. Haemorrhagic bullae and bruising began to form and crepitus was palpated. She was taken to the operating room where debridement was attempted, but the surgeons found deep and extensive tissue necrosis. She was transferred to the intensive care unit for a trial of medical therapy and eventual palliation. The pathology revealed severe myonecrosis secondary to *Clostridium septicum*. She expired 36 h after presentation.

BACKGROUND

Myonecrosis and spontaneous gas gangrene secondary to *Clostridium septicum* is an uncommon cause of skin and soft-tissue infection with high morbidity and mortality.¹ Early recognition and prompt treatment with appropriate antibiotics and surgical debridement are essential for favourable patient outcomes.² In the case of this patient, even though antimicrobial therapy was initiated early in the course of her hospital stay, failure to recognise the classic findings of a compartment syndrome (unrelenting pain, then numbness and weakness followed by a cold pulseless leg, crepitus and skin necrosis) resulted in a delayed surgical consultation. Physicians should be aware that serious deep tissue injuries such as myonecrosis, pyomyositis, necrotising fasciitis or gas gangrene can present in this way. Delayed recognition is common as the disease spreads from the deep tissue to the surface and so clinical signs are only obvious once the disease process is advanced. A high degree of clinical suspicion is necessary to prompt early surgical exploration, which is the only chance for recovery.

CASE PRESENTATION

A 57-year-old woman presented to the emergency department with 5 days of malaise and fever. She had a history of follicular and large B cell lymphoma for which she was receiving R-CHOP chemotherapy. Her fourth cycle of treatment was 10 days prior to admission. She had no other significant history apart from an uneventful 3-day hospitalisation for neutropaenic fever 2 months ago.

INVESTIGATIONS

On arrival, her vitals were as follows: heart rate 122 bpm, blood pressure 118/68 mm Hg, temperature 38.6°C, respiratory rate 20 and oxygen saturation 99% on room air. Her examination was otherwise unremarkable. Most notably,

her musculoskeletal examination including all limbs was normal. Her initial laboratory findings revealed haemoglobin 99 g/l, white blood cell $1.4 \times 10^9/l$, absolute neutrophil count $0.87 \times 10^9/l$. Chemistry and chest x-ray were normal. The creatine kinase and myoglobin levels were never sent. Blood cultures were drawn.

DIFFERENTIAL DIAGNOSIS

At the time of presentation the patient was treated as undifferentiated febrile neutropaenia as per the institutional guidelines.

TREATMENT

She was isolated with neutropaenic precautions, had blood cultures drawn, received a bolus of 2 litres intravenous normal saline and was empirically started on Ceftazidime 1 g intravenous q8h.

OUTCOME AND FOLLOW-UP

Seven hours after her admission, she complained of a 'funny feeling' in her right 'hip' that progressed over the next 3 h to a nebulous pain in her right hip and leg. Examination revealed no neurological or skin abnormalities, and normal range of motion. The right hip x-ray was normal. Over the next 6 h the pain progressed to severe right knee pain requiring escalating doses of morphine. However, she was still ambulating and her clinical examination, apart from persistent tachycardia, remained unremarkable. Blood cultures drawn earlier returned positive for anaerobic Gram-positive bacilli. An x-ray of her right knee was performed, which showed subcutaneous emphysema (figure 1). Unfortunately, this finding was not initially recognised by the admitting team and was officially reported by radiology the following day.

She then deteriorated rapidly. She developed a 'numbness' of the toes that progressed to bilateral leg weakness

Figure 1 Subcutaneous emphysema clearly visible in both the anteroposterior and lateral views of the patient's right leg, signifying an underlying gas gangrene, in this case due to infection from *Clostridium septicum*.

and then a cold pulseless right leg accompanied by sudden profound hypotension. Skin changes followed, which included mottling and discolouration of the lower leg and thigh that was associated with crepitus (figure 2).

Gas gangrene was suspected and the orthopaedic surgery service was consulted. She was taken to the operating room but unfortunately fasciotomy revealed extensive deep myonecrosis of both legs and the pelvis (figure 3). A decision was made to change goals of care to palliation and she died shortly afterwards. The blood cultures grew *C septicum*.

DISCUSSION

C septicum myonecrosis is an uncommon condition but is associated with a high risk of significant morbidity and mortality. There are approximately 1000 reported cases of clostridial myonecrosis each year in the USA,³ but less than 10% of these occur spontaneously in the absence of trauma. For these reasons a high index of suspicion is essential to recognise this critical condition.

C septicum is an anaerobic, Gram-positive, spore-forming bacilli commonly found in the soil and gastrointestinal tract. It is often associated with malignancies, most commonly colorectal or, as in the case of our patient, haematological as well as in patients with diabetes mellitus.⁴ It

is an opportunistic pathogen which is thought to gain access to the bloodstream through breaches in the gastrointestinal mucosa, which then lead to systemic manifestations.²

Clinical presentations of *C septicum* infection often manifest as rapidly progressive skin and soft-tissue infection with fever, tachycardia, hypotension, delirium and leucocytosis.⁵ Other findings that may predict spontaneous myonecrosis include lack of trauma, pain out of proportion to physical signs, purpura, crepitus and haemorrhagic bullae.⁵

When recognised, the cornerstones of treatment involve the initiation of appropriate antibiotic therapy and early surgical consultation for excision and debridement of the affected tissues. Penicillin is the usual antibiotic of choice with a recommended dose of 10–12 million units daily, but other alternatives include cephalosporins, clindamycin, metronidazole, vancomycin and tetracycline.⁴ There are also reported uses of hyperbaric oxygen therapy as adjunctive therapy for treatment, and it has been recommended as an adjunct to medical and surgical therapy.⁶

Despite all these potential therapies, the mortality associated with *C septicum* myonecrosis approaches 100%,² especially if it progresses to cause a compartment syndrome. Our patient demonstrated a natural history of compartment syndrome secondary to *C septicum* myonecrosis. She developed severe limb pain out of proportion to the physical findings; then, she showed numbness of her limbs followed by weakness and an impaired vascular supply due to the rapidly progressing soft-tissue necrosis. Serious deep tissue infections require early recognition, appropriate management, as well as timely referral to appropriate surgical services to prevent poor patient outcomes. Increased vigilance and a higher clinical suspicion are necessary to prevent the recurrence of this unfortunate situation.

Figure 2 Gross superficial examination of the leg approximately 22 h after initial patient presentation. Areas of necrotic bullae as well as erythema secondary to ischaemia from compartment syndrome due to the underlying myonecrosis from *Clostridium septicum* are visible in the figure.

Learning points

- Recognise the early signs of compartment syndrome and that this condition can be caused by an infectious aetiology.
- Call for surgical consultation early when deep soft-tissue infections are suspected.
- Be aware of conditions that can lead to rapid deterioration and death in neutropaenic patients.

Figure 3 (Left) Gross intraoperative findings. As is visible, there are areas of deep necrotic tissue secondary to ischaemia from elevated pressures due to compartment syndrome. (Right) Histological findings from operative pathology. Gram stain-positive *Clostridium septicum* bacteria are visible in the surrounding muscle tissue and in necrotic tissue on the left side of the slide.

Competing interests None.

Patient consent Obtained.

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