Acute respiratory distress syndrome associated with femoral osteomyelitis

Introduction

Emphysematous osteomyelitis, an infection of bone caused by gas-forming microorganisms, is an extremely rare but serious condition. It has a high mortality (32%), especially in immunosuppressive conditions such as diabetes mellitus and malignancy. Emphysematous osteomyelitis has been reported in vertebrae and the extra-axial skeleton, including the pelvis, sacrum, femur, tibia, fibula and midfoot.

This article reports a case of emphysematous osteomyelitis caused by *Klebsiella pneumoniae* in a newly diagnosed diabetic 58-year-old man who presented with fever and shortness of breath for 3 days. He was referred to the authors' hospital from a rural hospital because he had pulmonary septic emboli with acute respiratory distress syndrome. Emphysematous femoral osteomyelitis of *K. pneumoniae* was confirmed through computed tomography, culture of abscess and operative findings. He was discharged on the 57th day after debridement and full-course intravenous antibiotics.

Discussion

Emphysematous osteomyelitis is an infection of bone caused by gas-forming

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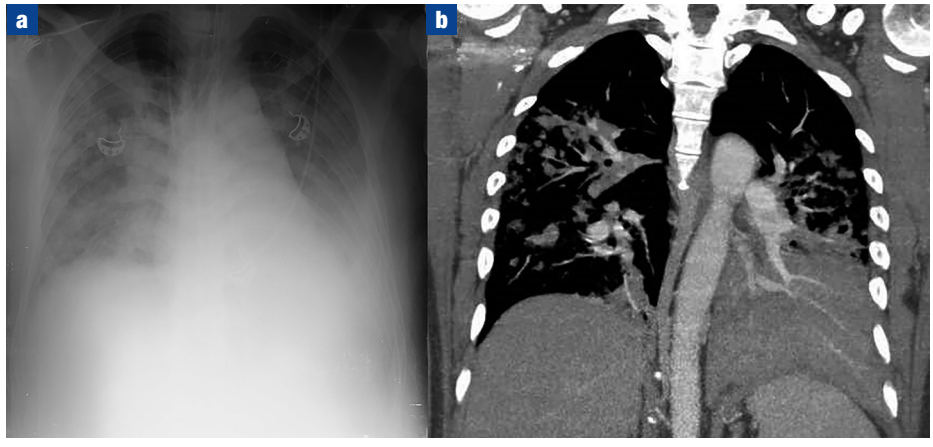
Figure 1. **a.** Chest X-ray showed bilateral diffuse infiltration with consolidations. **b.** Multidetector computed tomography showed consolidations of both lungs and numerous small nodules in the bilateral lung fields.



CASE REPORT

A 58-year-old healthy Taiwanese man had received traditional manoeuvres for muscle strain of the right leg 2 weeks previously. He suffered from fever and shortness of breath 3 days before admission. Abnormal chest X-ray and plain film of the right femoral shaft were noted at a rural hospital.

He was referred to the authors' hospital after endotracheal intubation for acute respiratory distress syndrome caused by emphysematous osteomyelitis of the right femur complicated with septic shock. On arrival, his vital signs were respiratory rate 34 breaths/min, heart rate 114 beats/min, blood pressure 56 mmHg and temperature 38.5°C. Physical examination showed crackles over both lungs and local swelling of the right thigh.

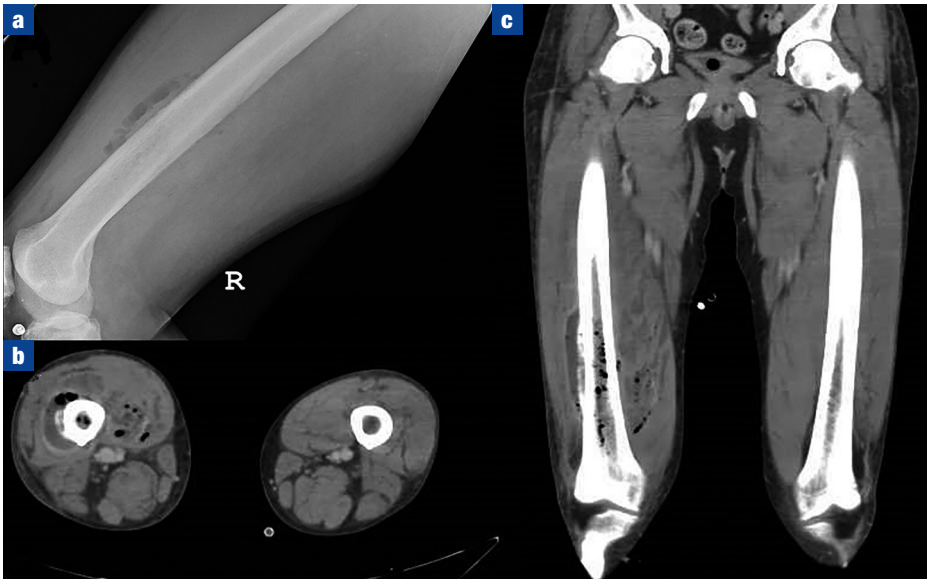
Laboratory investigations were white blood cell counts 22 100/mm^3 with segmented neutrophils of 84%, haemoglobin 10.6 g/dl, platelet counts 425x10^3/mm^3, blood urea nitrogen 28 mg/dl, creatinine 1.4 mg/dl, calcium 8.6 mg/dl, lactate 17.5 mg/dl (reference range 8–12 mg/dl), alkaline phosphatase 259 U/litre (reference range <190 U/litre), albumin 2.7 g/dl, glutamic-oxaloacetic transaminase 28 U/litre, glutamic pyruvic transaminase 42 U/litre, lactate dehydrogenase 313 U/litre (reference range < 240 U/litre), C-reactive protein 37.13 mg/dl (reference range <0.4 mg/dl), blood glucose 130 mg/dl and glycated haemoglobin 8.5%. Arterial blood gas was pH 7.469, arterial partial pressure of oxygen 63 mmHg, arterial partial pressure of carbon dioxide 34.2 mmHg, oxygen saturation 93% and bicarbonate 24.3 mmol/litre.

Multidetector computed tomography showed consolidation of both lungs, numerous small nodules in the bilateral lung fields (*Figure 1*), and air bubbles within the femur and quadriceps muscle (*Figure 2*).

Early goal-directed therapy with empiric antibiotics, fluid resuscitation and vasopressor for septic shock and the lung protective strategy with prone position for acute respiratory distress syndrome were applied. Debridement was carried out for emphysematous femoral osteomyelitis and the pathological report showed acute suppurative osteomyelitis with new bone formation. Culture of the abscess grew *Klebsiella pneumoniae* and cefazolin 1000 mg was given every 6 hours for 2 weeks. Anaerobic bacterial, fungal culture, acid-fast stain and culture for tuberculosis from the abscess and a biopsy from the emphysematous osteomyelitis were negative.

He was discharged on the 57th hospital day after debridement, wound care and a full course of intravenous antibiotics.

Figure 2. **a.** Plain film of the right femoral shaft showed air bubbles within the femur and periosteal soft tissue. **b** and **c.** Multidetector computed tomography showed air bubbles within the femur with bony destruction and swollen quadriceps muscles.



LEARNING POINTS

- Emphysematous osteomyelitis is a life-threatening infection caused by gas-forming bacteria.
- Intraosseous gas is an alarming sign of emphysematous osteomyelitis.
- Computed tomography is excellent in detecting early signs of emphysematous osteomyelitis.
- *Klebsiella pneumonia* is one of the most common microorganisms in emphysematous osteomyelitis, especially in diabetic patients.
- Early diagnosis, broad spectrum antibiotics and aggressive surgical intervention are recommended for emphysematous osteomyelitis.

microorganisms (Shanklesha et al, 2017). It is an extremely rare but serious condition with a mortality of 32%, especially in patients who are immunosuppressed. Diabetes mellitus and malignancy are common predisposing factors (Luey et al, 2012; Aiyappan et al, 2014).

Intraosseous gas in the appendicular skeleton is pathognomonic of emphysematous osteomyelitis with the uneven and bubbly distribution of gas and extension into adjacent soft tissue (Feng et al, 2011). A lucent lesion on X-ray suggestive of intraosseous gas is the first sign of emphysematous osteomyelitis, but plain radiographs may not pick up small amounts of intraosseous gas, which is confirmed on computed tomography (Feng et al, 2011; Mahesh et al, 2016). Emphysematous osteomyelitis has been reported in the vertebrae and extra-axial skeleton, including the pelvis, sacrum, femur, tibia, fibula and midfoot (Luey et al, 2012; Aiyappan et al, 2014; Lee et al, 2017).

Common microorganisms include Staphylococcus, Streptococcus, Enterobacteriaceae family (*Escherichia coli* and *K. pneumoniae*), anaerobic bacteria, and sometimes *Mycobacterium tuberculosis*, with mono- or poly-microbial infections. The monomicrobial causes of emphysematous osteomyelitis are similar to other gas-forming infections through haematogenous spread, and polymicrobial infections from the local infection of adjacent tissue (Putcharoen and Suankratay, 2007; Chen and Huang, 2016).

K. pneumoniae infection will cause the development of gas-forming pyogenic liver abscesses in patients with diabetes mellitus. In these patients, high blood glucose levels may provide a more favourable environment for gas formation via mixed acid fermentation of glucose. Emphysematous osteomyelitis caused by *K. pneumoniae* through forming gas via mixed acid fermentation of glucose was similar to other gas-forming infections in diabetic patients in Taiwan (Lee et al, 2004).

Polymicrobial infections of emphysematous osteomyelitis accounts for 40% (10/25) and 30% (3/10) of these cases were not associated with prior surgery or a contiguous focus of infection in a literature review (Luey et al, 2012). Polymicrobial infections were excluded through aerobic, anaerobic bacterial, and even fungal cultures of pus and/or surgical specimens. Early diagnosis through radiological study to confirm intraosseous gas and involved soft tissue, including plain films and computed tomography, is suggested in high suspicious cases.

Computed tomography is excellent in detection of early signs of emphysematous osteomyelitis such as intraosseous gas (Aiyappan et al, 2014). Magnetic resonance imaging has the benefit of detecting signal abnormalities of bone marrow and changes of soft tissue (Shanklesha et al, 2017). The presence of intraosseous gas is an alarming sign which must be recognized as soon as possible. Although effective antibiotic durations for treatment of emphysematous osteomyelitis are not confirmed, broad spectrum antibiotics for common microorganisms and aggressive surgical intervention are recommended because emphysematous osteomyelitis is associated with significant morbidity and mortality (Lee et al, 2017; Shanklesha et al, 2017). **BJHM**

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