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Case Report

Emphysematous osteomyelitis: a case report and review of the literature

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SUMMARY

We report the case of a 15-year-old girl with pelvic and sacral emphysematous osteomyelitis caused by *Fusobacterium necrophorum*. This infection was cured following four surgical procedures and 4 weeks of intravenous then 4 weeks of oral antibiotics. We review our case alongside the 24 previously reported cases of emphysematous osteomyelitis in the literature. The 25 cases include 15 monomicrobial and 10 polymicrobial infections. The causative organism(s) in all but three cases included an anaerobe or a member of the *Enterobacteriaceae* family. A significant underlying comorbidity was reported in 18 cases. At least 15 cases required one or more surgical procedures. There was a significant associated mortality with eight (32%) patients dying in hospital at 7 to 56 days after the diagnosis of emphysematous osteomyelitis.

1. Introduction

Intraosseous gas was first described as a sign of osteomyelitis in 1981.[1] When seen in the extra-axial skeleton, intraosseous gas is virtually pathognomonic for emphysematous osteomyelitis; in rare cases the differential diagnosis includes trauma, post-surgical change, lymphangiomatosis of the bone, degenerative disease, osteonecrosis, and neoplasm.[2] Conversely, intraosseous gas seen in vertebral bodies is almost always due to a non-infectious cause. Indeed intravertebral gas was long considered to exclude infection, with almost all cases due to degenerative disease, or less commonly, osteonecrosis or a neoplastic process.[3,4] Osteomyelitis is likely when extensive intravertebral gas, bone edema, and/or adjacent collections are present.

We report the case of a 15-year-old girl with pelvic and sacral emphysematous osteomyelitis caused by *Fusobacterium necrophorum*. We review the 24 cases of emphysematous osteomyelitis reported in the literature.

2. Case report

A 15-year-old, previously healthy girl was admitted to the emergency department following a low-speed road traffic accident. She suffered only minor injuries and was discharged the same day with mild right hip pain. Four days later she presented to her general practitioner with low back pain, fever, and a sore throat. She was diagnosed with tonsillitis and treated with oral amoxicillin–clavulanate and ibuprofen.

She presented again to the emergency department the following day with ongoing back pain and was noted to have a temperature of 37.9 °C. Lumbar spine X-rays were normal and she was discharged the same day with provisional diagnoses of myalgia and tonsillitis. She returned to the emergency department on day 13 post-road traffic accident with ongoing back pain radiating to her right knee. She was afebrile but had an antalgic gait. Her neurological examination was normal. She was discharged with analgesia.

Later that day she became unwell with nausea, vomiting, and abdominal pain radiating through to her back. She was incontinent of urine overnight with subsequent urinary retention, and the following day complained of numbness in both legs and required assistance to walk. She returned to the emergency department by ambulance that evening.

On arrival she was febrile, tachycardic, and hypotensive. She was significantly distressed and uncooperative, hampering any neurological examination. It was noted however that she had little movement of her lower limbs. Her white blood count was 3.9×10^9 cells/l with neutrophils showing severe toxic changes. Other laboratory results included creatinine 145 μmol/l, bilirubin 39 μmol/l, γ-glutamyl transferase 117 U/l, alkaline phosphatase 551 U/l, aspartate transaminase 76 U/l, and alanine transaminase 49 U/l. She was treated for presumed biliary sepsis with intravenous amoxicillin, gentamicin, and metronidazole before being transferred to the intensive care unit where she was sedated and intubated.

An abdominal ultrasound revealed a distended gallbladder with associated sludge, however a subsequent computed tomography

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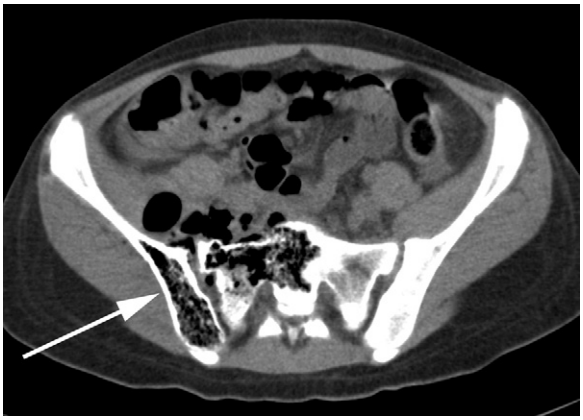

Figure 1. Computed tomography of the pelvis showing extensive emphysematous osteomyelitis of the right ilium (arrow) and S1.

(CT) scan of her abdomen and pelvis revealed a large gas-containing abscess in the right iliacus muscle extending into the right psoas muscle, and extensive intraosseous gas in the adjacent S1 vertebra and ilium consistent with emphysematous osteomyelitis (Figure 1). Her antibiotic treatment was changed to amoxicillin, flucloxacillin, aztreonam, and metronidazole and she was transferred to a tertiary intensive care unit. A magnetic resonance imaging (MRI) scan confirmed the CT findings and also revealed a gas-containing epidural abscess from T12–S1 with associated cord compression (Figure 2).

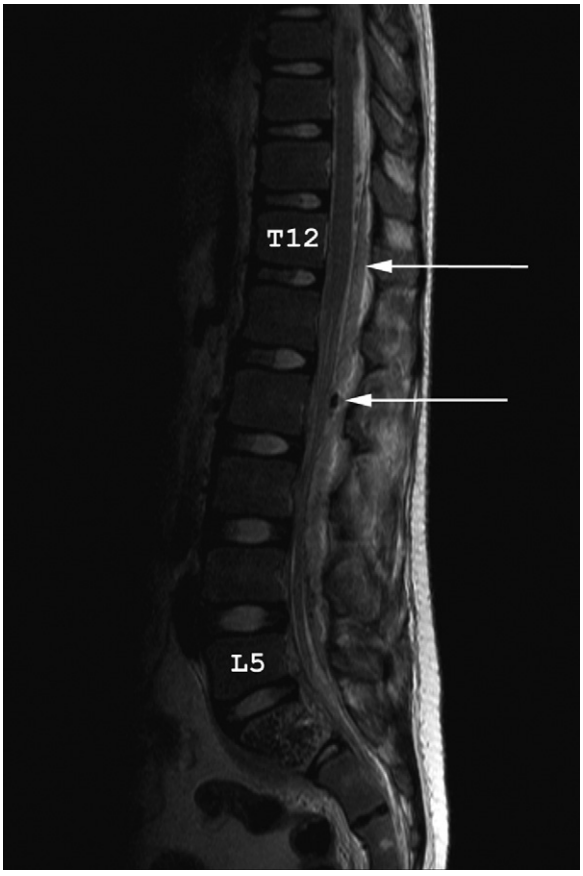

Figure 2. T2-weighted magnetic resonance image of the lumbar and sacral spine showing a posterior epidural abscess (top arrow) at the T12–S1 vertebral levels and epidural gas (bottom arrow).

Urgent surgical decompression released foul smelling gray pus from the epidural and pelvic abscesses. All surgical specimens and blood cultures revealed a monomicrobial growth of *F. necrophorum* sensitive to penicillin, clindamycin, and metronidazole. Her antibiotic treatment was changed to intravenous metronidazole 500 mg every 8 h and clindamycin 300 mg every 6 h.

A CT scan of her head and neck did not reveal an abscess or any evidence of thrombophlebitis. Further abdominal surgery was required on days 3, 5, and 7, however minimal pus was found and all subsequent surgical samples remained sterile.

Her clinical improvement was slow and she remained febrile for 2 weeks. Due to ongoing nausea and vomiting, her antibiotics were changed after 2 weeks to intravenous benzylpenicillin monotherapy, 1.2 g every 6 h. This was continued for a further 2 weeks before completing a further 4 weeks of oral phenoxymethylpenicillin 500 mg four times daily; a total of 8 weeks of antibiotic treatment.

She was eventually discharged from the rehabilitation unit 15 weeks after admission. Unfortunately she had ongoing bilateral leg weakness requiring crutches to mobilize and required ongoing self-catheterization and manual bowel evacuation. At 18 months follow-up she remained free of relapse.

3. Discussion

There are 25 cases of emphysematous osteomyelitis (including our case) reported in the literature[1,2,4–19] (see Table 1). There is no gender association, with 13 of the 25 cases occurring in females. The median age at presentation was 51 (range 14–78) years.

The 25 cases include 15 monomicrobial and 10 polymicrobial infections. The mechanism of infection in 14 of the 15 monomicrobial cases was felt to be via hematogenous spread, whereas the mechanism of infection in seven of the 10 polymicrobial cases was felt to be due to contiguous spread from an intra-abdominal source of infection ($n = 3$), contiguous spread from a skin/soft tissue source of infection ($n = 1$), or following orthopedic or intra-abdominal surgery ($n = 3$).

Significant underlying comorbidity was reported in 18 of the 25 cases, most commonly diabetes ($n = 6$) and malignancy ($n = 5$). There were no recorded comorbidities in seven cases. In all three monomicrobial cases with no reported underlying comorbidity the causative organism was *F. necrophorum* (cases 3, 4, and 6). This is consistent with the epidemiology of disseminated necrobacillosis, which is well known to occur in young healthy patients.[20] Only three polymicrobial cases (cases 16, 21, and 24) were not associated with prior surgery or a contiguous focus of infection; of these, two had a significant underlying comorbidity.

The causative organism in the monomicrobial cases was either an anaerobe (including four cases of *F. necrophorum*) or a member of the *Enterobacteriaceae* family, except for one case caused by *Mycobacterium tuberculosis*. The causative organisms in all but two of the polymicrobial cases also included an anaerobe or a member of the *Enterobacteriaceae* family. In these two cases of post-surgical infection (cases 24 and 25) the cultured organisms were *Staphylococcus aureus*, a non-hemolytic Streptococcus, an *Enterococcus spp*, and a *Pseudomonas spp* in one, and *Streptococcus intermedius* and an *Enterococcus spp* in the other. It is possible that prior antibiotic treatment and/or culture methods may have hindered the anaerobic culture in these two cases.The monomicrobial causes of emphysematous osteomyelitis are similar to reported monomicrobial causes of other gas-forming infections (gas-forming soft tissue infection, gas-forming brain abscess, gas-forming liver abscess, and emphysematous urinary tract infection), which include *Escherichia coli*, *Klebsiella pneumoniae*, *Enterobacter aerogenes*, and *Clostridium spp*.[21–24]