Urosepsis and Emphysematous Osteomyelitis: A Case Report

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Background

Emphysematous osteomyelitis (EO) is a rare, aggressive and potentially fatal condition[1]. First described in 1981, this form of osteomyelitis is caused by gas-forming organisms, and is characterised by the presence of intraosseous gas within the involved bone[1].
Common causative pathogens include *Escherichia coli, Klebsiella pneumoniae*, and anaerobic organisms. Polymicrobial infections have also been reported. The condition has a reported mortality rate of up to 37%[1].

Case Report

A 78-year-old man presented to ED with a one-week history of flu-like symptoms, and a four-day history of lower back and gluteal pain, with associated lower limb weakness.

Medical history was significant for type 2 diabetes mellitus, and prostate cancer with radical prostatectomy 10 years previously. He was not on insulin, but was prescribed a sodium-glucose co-transporter-2 (SGLT2) inhibitor.

The patient was febrile and confused on examination. He was unable to weight-bear and passive flexion at both hips caused severe pain.

He was found to be in a state of diabetic ketoacidosis (DKA), with raised inflammatory markers, thrombocytopenia and an acute kidney injury. He was initiated on broad-spectrum antimicrobial therapy with co-amoxiclav, and was commenced on protocol-based treatment for diabetic ketoacidosis.

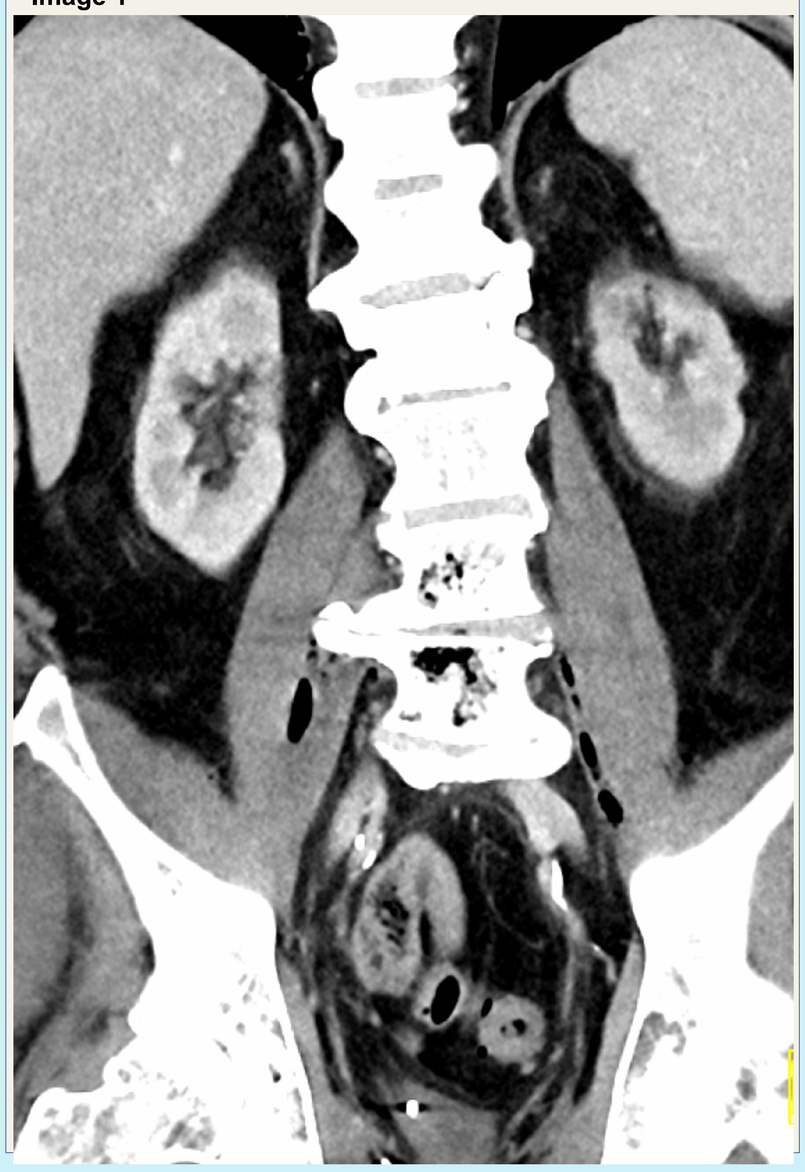
Laboratory test results on admission were: CRP 400mg/L, WCC 15.3x10^9/L, Creatinine 208umol/L, Urea 22.2mmol/L, PLT 96x10^9/L.

As lower back pain was the patient's only localizing symptom, cross-sectional imaging was performed to investigate for a source of infection. CT showed extensive pockets of air centred on the L4 and L5 vertebral bodies, with extension into the adjacent iliopsoas muscles bilaterally as well as the anterior epidural space from L3-S1 (image 1, 2, 4). *Klebsiella pneumoniae* was subsequently isolated in blood and urine cultures that had been sent at the time of admission.

Subsequent magnetic resonance imaging(MRI) showed an epidural abscess along the posterior body of L5 (image 3), with disk bulge causing severe canal and bilateral foraminal stenosis. Findings were consistent with emphysematous osteomyelitis and bilateral iliopsoas abscesses. Urosepsis with lymphovascular drainage to the lumbar region was considered the likely route of transmission.

The patient was switched to IV cefotaxime on the basis of sensitivities. He initially responded well to antibiotic treatment, with resolution of fever and improvement in his pain, coupled with down-trending inflammatory markers. Repeat blood cultures were sterile. His case was discussed with neurosurgical colleagues, who advised that there was no role for surgical intervention at this point. Our colleagues in interventional radiology (IR) were initially unable to recommend IR-guided drainage of the abscesses, due to their small size and challenging anatomical location.

Image 1



We therefore proceeded with medical management, including a prolonged course of IV antibiotics, optimization of blood sugars, correction of anaemia and electrolyte disturbance, and multidisciplinary team input.

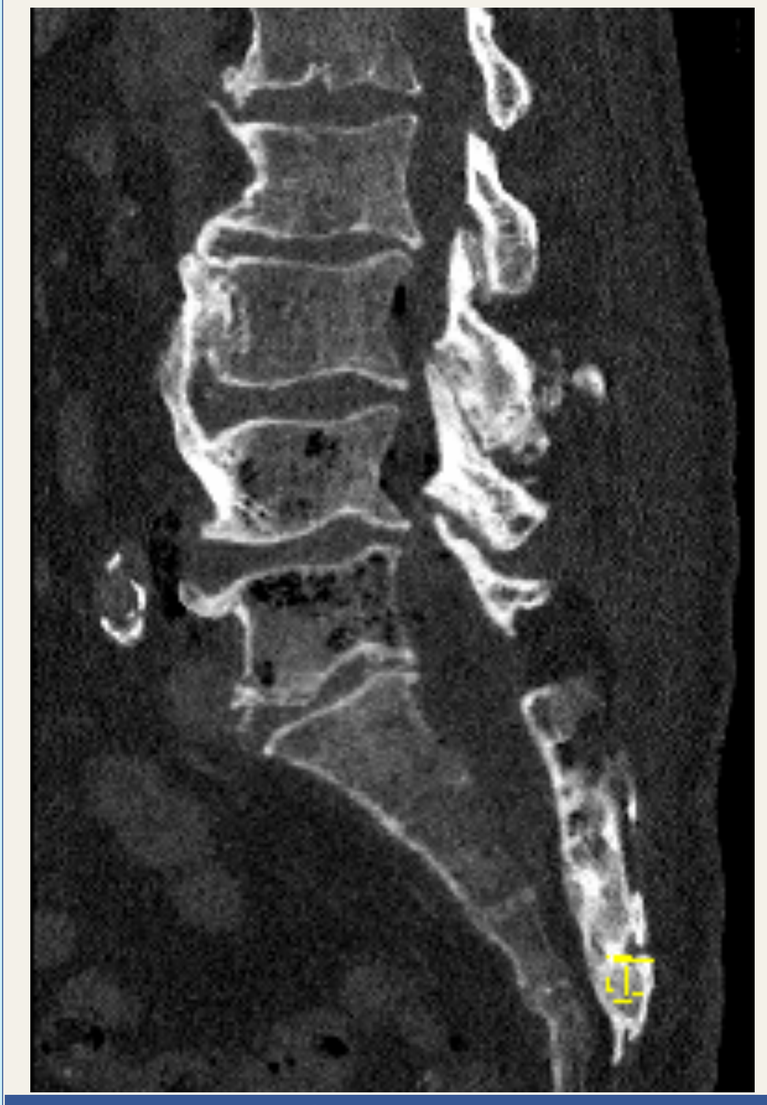
An interval scan on day 32 of IV antibiotics showed an increase in the size of the iliopsoas abscesses. At this point, the right iliopsoas abscess was deemed suitable for radiological drainage.
The right iliopsoas abscess was drained on day 39 of treatment. This led to significant clinical and biochemical improvement, and repeat imaging after a two-week interval showed resolution of the right sided iliopsoas abscess, with interval reduction in the size of the left sided abscess.

The patient continues to be managed medically. After 7 weeks of intravenous therapy he was switched to oral cotrimoxazole, with a planned total duration of 12 weeks antibiotics, and repeat imaging upon completion of this course. His mobility remains markedly impaired, and his cognitive status has declined over the course of his two month admission.

Timeline:

Day 1	**Admission**
Day 2	**Diagnosis of EO confirmed**
Day 32	**Increase in abscess size on imaging**
Day 39	**IR drainage**
Day 42	**PO switch**
Day 84	**Planned antibiotic stop date & interval scan**

Image 2



Discussion

Though one of the most common gram-negative species causing invasive infection, *K Pneumoniae* is not commonly associated with osteomyelitis. Typical associations with urinary tract, respiratory, intra-abdominal and intracranial infections are well documented[2]. However, a literature review of emphysematous osteomyelitis identified *K Pneumoniae* as the causative agent in 20% of all reported cases in the English literature[1], suggesting that it is a relatively common cause of this rare condition. Diabetes was a predisposing factor in 34% of cases, and nearly half of all cases involved the vertebrae[1].

In recent years, hypervirulent strains of *K pneumonia* have been identified, causing liver abscesses and metastatic complications including meningitis, necrotizing fasciitis, and ocular infections. Diabetes is also reported to predispose to this distinct invasive syndrome[3]. It has been suggested that emphysematous osteomyelitis caused by *K pneumoniae* should be included within the definition of this invasive syndrome[1]. In this case, the isolate was not tested for hypervirulent features.

Medical management of EO should include broad-spectrum antibiotics, before targeting therapy towards the causal microbe (where identified)[4]. The optimal duration of therapy has not been concluded due to the small number of cases. One literature review reported a high level of surgical intervention in EO of the spine, with 38% of cases requiring surgical intervention in the form of debridement, decompression, or decompression with instrumentation for fusion[4]. In our case, despite a prolonged course of IV antimicrobial therapy, significant clinical improvement was not seen until drainage of the iliopsoas abscess on his 39th day of antibiotic treatment.

This case highlights a rare but severe complication of invasive *K Pneumoniae* infection and emphasizes the importance of cross-sectional imaging in patients with new onset back pain in the setting of *Klebsiella* bloodstream infection. The underlying diagnosis of diabetes, coupled with identification of *K pneumonia* as a causative organism, makes this a relatively typical presentation of this a rare condition, This is, to our knowledge, the first description of emphysematous osteomyelitis presenting as DKA, and indeed the presence of DKA may have delayed diagnostic imaging to identify a definitive source of infection, as the initial priority was reversal of life-threatening acidosis. The patient's clinical course, with increase in abscess size despite medical management, supports the literature suggesting that surgical management (or in this case IR-guided drainage) is often necessary to achieve source control and clinical improvement.

With data limited to case reports and case series, there are no clinical practice guidelines to inform decision-making around surgical and medical management of this rare condition.

Image 3

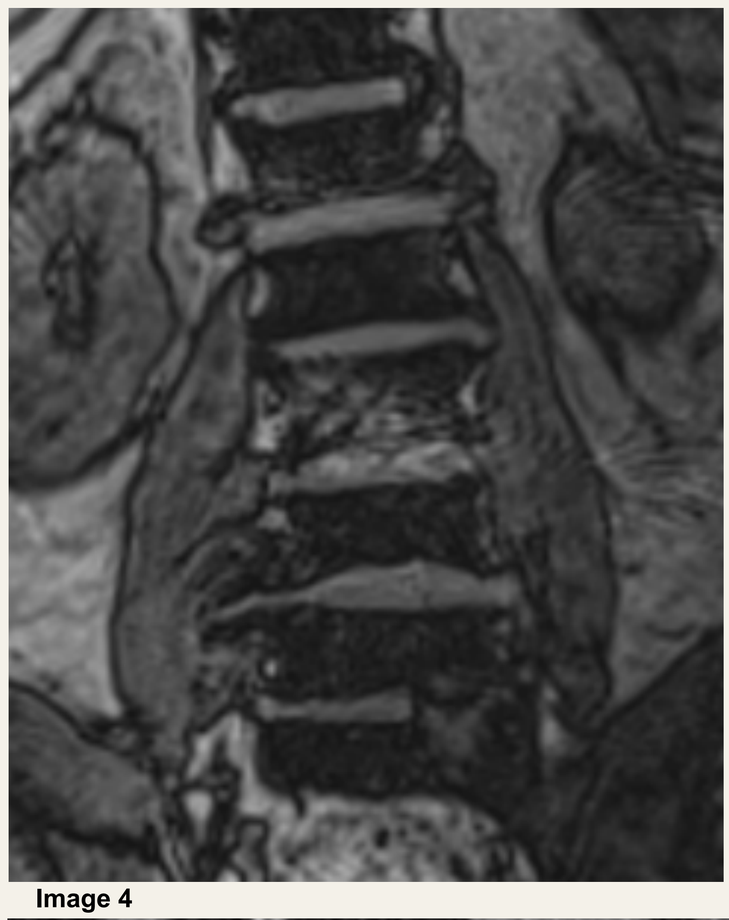
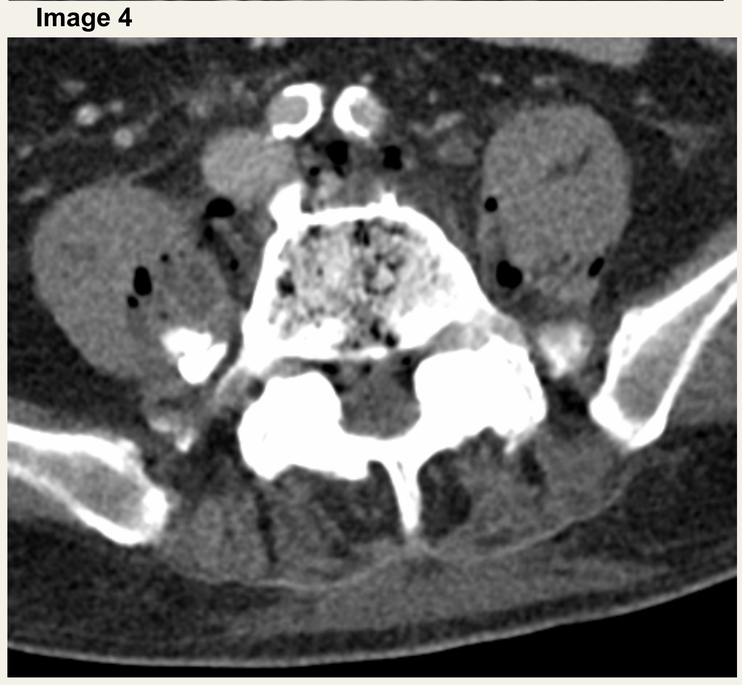


Image 4



References

1. Ono R, Uehara K, Kitagawa I. Emphysematous Osteomyelitis of the Spine: A Case Report and Literature Review. *Intern Med*. 2018;57(14):2081-2087. doi:10.2169/internalmedicine.0219-17
2. Vading M, Nauclér P, Kalin M, Giske CG. Invasive infection caused by Klebsiella pneumoniae is a disease affecting patients with high comorbidity and associated with high long-term mortality. PLoS One. 2018 Apr 6;13(4):e0195258. doi: 10.1371/journal.pone.0195258. PMID: 29624618; PMCID: PMC5889183.
3. Siu LK, Yeh KM, Lin JC, Fung CP, Chang FY. Klebsiella pneumoniae liver abscess: a new invasive syndrome. Lancet Infect Dis. 2012 Nov;12(11):881-7. doi: 10.1016/S1473-3099(12)70205-0. PMID: 23099082.
4. Park SS, Lee SE, Min CK. Emphysematous osteomyelitis due to *Escherichia coli* in multiple myeloma. *Blood Res*. 2016;51(4):224. doi:10.5045/br.2016.51.4.224