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Simultaneous Diagnosis of Emphysematous Osteomyelitis and Emphysematous Pyelonephritis in a Diabetic Patient

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Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
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Conflict of interest: None declared

Patient: Female, 57-year-old
Final Diagnosis: Emphysematous osteomyelitis and emphysematous pyelonephritis
Symptoms: Diarrhea • nausea • vomiting • weakness
Medication: —
Clinical Procedure: Conservative therapy
Specialty: Infectious Diseases

Objective: Rare disease
Background: Emphysematous osteomyelitis of the spine is characterized by intravertebral or intraosseous air. Emphysematous pyelonephritis (EP) is the infection of the renal parenchyma and perirenal tissues caused by gas forming microorganisms and thus is characterized by gas formation. Prompt diagnosis and initiation of necessary treatment is crucial, as both entities are associated with high mortality rates.

Case Report: A 57-year-old female with uncontrolled hyperglycemia presented to the emergency department with history of sudden onset of weakness, nausea, vomiting and diarrhea for 3 days and with a fall on the same level the previous day. Laboratory examinations revealed leukocytosis, lymphopenia, thrombocytopenia, deteriorated renal function, and hyperglycemic hyperosmolar non-ketotic state. She was placed on aggressive intravenous hydration and insulin infusion pump. Due to the deterioration of her medical condition, she underwent abdominal and pelvic CT scanning that revealed emphysematous osteomyelitis of the spine and emphysematous pyelonephritis. Despite vigorous fluid resuscitation and systemic broad-spectrum antibiotic therapy, the patient's condition deteriorated further and eventually led to death within 48 h.

Conclusions: This case of fatal emphysematous osteomyelitis of the spine and EP serves as a significant reminder of those rare life-threatening entities, which affect patients with comorbidities, such as diabetes mellitus and other etiologies causing immunosuppression. The aim of the present case report is to highlight the importance and contribution of computed tomography in diagnosing these conditions and to emphasize the rare coexistence of these 2 emphysematous entities.

MeSH Keywords: Diabetes Complications • Osteomyelitis • Pyelonephritis

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Background

Emphysematous osteomyelitis is defined as a bone infection caused by a variety of gas-producing bacteria [1]. This entity is uncommon, with only 26 cases reported in a recent review and only 10 cases involving the spine [2]. The reported mortality rate is 32%. Early diagnosis is essential for initiating therapy and decreasing lethal outcomes. Ram et al. (1981) described the existence of gas as the pathognomonic radiological finding of subsequent infection in the appendicular skeleton [1,3].

EP is a fulminant infection of the renal parenchyma and perirenal tissues, characterized by gas formation [4]. Monomicrobial cases concern 86–95% of patients and the causative organisms are the *Enterobacteriaceae* species (80–95%), *Aerobacter aerogenes* (10%), and rarely *Pseudomonas* and *Candida* [5]. The majority of patients present with fever, lumbar pain, and urinary symptoms. Although this entity is rarely encountered in clinical practice, EP has a predilection for diabetic patients and is associated with high mortality [4]. Early diagnosis with urgent medical and surgical intervention can decrease the mortality of this potentially lethal disease.

In this case report, we present one such fatal case diagnosed in an internal medicine department. We herein describe features that could be helpful in differentiating emphysematous osteomyelitis of the spine from other gas-producing entities, while we also present the main features of emphysematous pyelonephritis.

Case Report

A 57-year-old female visited the trauma and emergency department with history of falling on the same level the previous day, with sudden onset of weakness, nausea, vomiting, and diarrhea for 3 days. She was a known smoker, obese, without regular medical attention, and she also had uncontrolled hypothyroidism. Physical and laboratory examinations revealed leukocytosis, lymphopenia, thrombocytopenia, deteriorated renal function, and hyperglycemic hyperosmolar non-ketotic state. The blood investigations revealed leukocytosis and mild thrombocytopenia. Her blood urea and serum creatinine were 78 (normal range 17–52 mg/dl) and 3.1 (normal range 0.7–1.3 mg/dl) mg/dl, respectively. Urinalysis showed glycosuria (>1000 mg/dl) and pyuria, but negative results for ketones. Her blood glucose concentration was 1680 mg/dl, with a glycated hemoglobin of 16.3%. She had no past medical history of diabetes mellitus.

She was diagnosed with hyperglycemic hyperosmolar non-ketotic state. Arterial blood gases revealed normal pH. Serum osmolarity was calculated at 335 mosm/l. The inflammatory markers

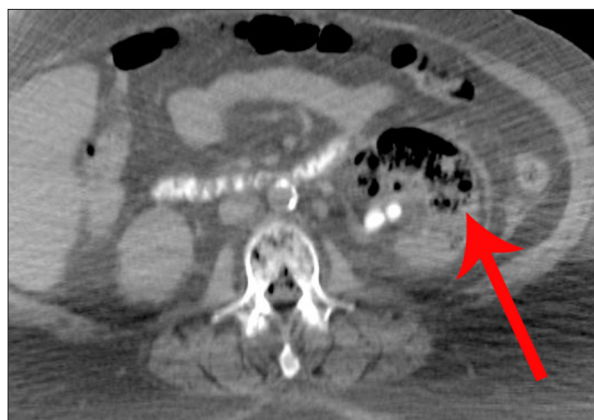


Figure 1. CT of the abdomen, axial slice: gas within the left kidney and infiltration of septa in the perirenal space (red arrow).

were as follows: C reactive protein (CRP) was 43.2 mg/dl (normal range 0–0.5mg/dl) and procalcitonin (PCT) was 56.34 ng/ml (normal range <0.5 ng/ml). On admission, she was afebrile and her vital signs were stable. She was placed on vigorous intravenous hydration and insulin infusion pump.

After 12 h, she was sedated and intubated in the intensive care unit (ICU) due to the deterioration of her medical condition. She was started empirically on injection ceftriaxone of 2 g qDay intravenously (IV) and clindamycin 1.8 g/day IV divided q8hr.

Abdominal and pelvic CT scan showed gas within the left kidney and revealed infiltration of septa in the perirenal space (Figures 1, 2). Computed tomography also revealed moderate intraosseous gas within the L3 and L4 lumbar vertebral bodies and emphysema in the epidural space (Figures 2, 3). By correlating the clinical, biochemical, and radiological findings, she was diagnosed with emphysematous pyelonephritis and emphysematous osteomyelitis of the spine. In addition, *Klebsiella pneumoniae* was identified in urine culture. The antibiotic therapy was changed to meropenem 1 g iv q8hr owing to the complicated sepsis and the diagnosis of emphysematous osteomyelitis and pyelonephritis.

Despite full supportive measures, including vigorous fluid resuscitation and systemic antibiotic therapy, the patient's condition progressed to septic shock and multiple organ dysfunction. As a result, she died due to her medical condition and devastating consequences of the underlying disease within 48 h after admission.

Discussion

A review of case reports by Luey et al. in 2012 found 25 cases of emphysematous osteomyelitis. The spine was the most

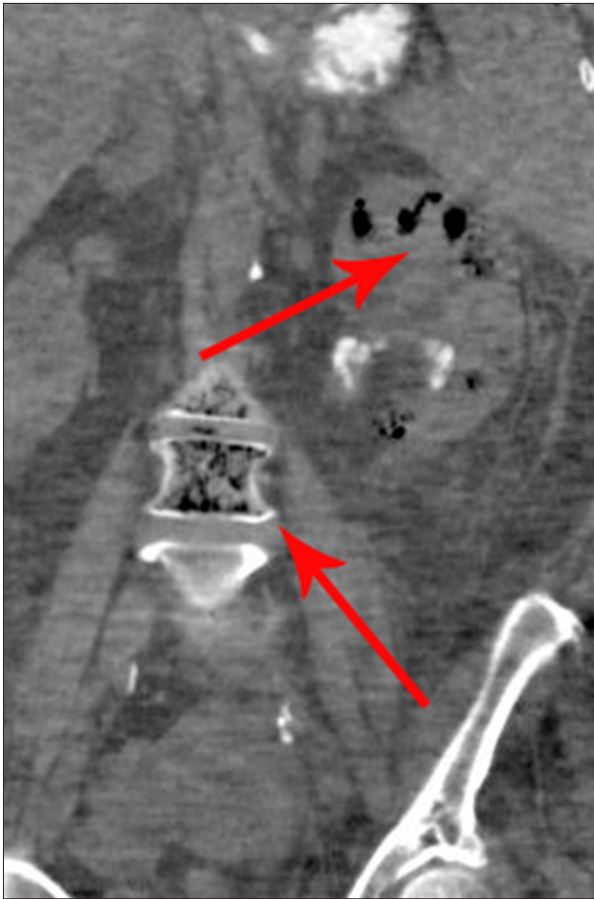


Figure 2. CT of the abdomen, coronal slice: emphysematous osteomyelitis of spine and emphysematous pyelonephritis (red arrows).

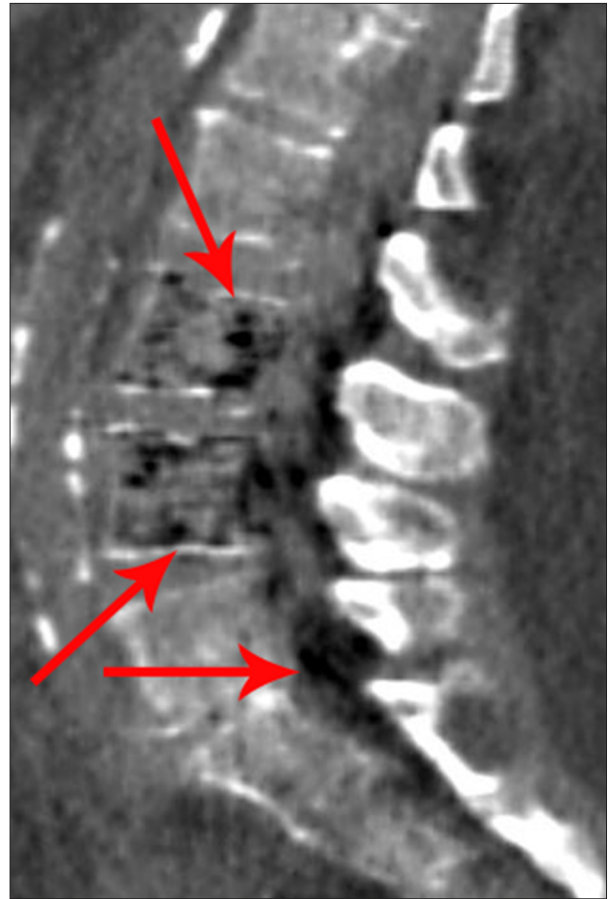


Figure 3. CT of the lumbar spine, sagittal slice: intraosseous gas within the L3 and L4 lumbar vertebral bodies and emphysema in the epidural space (red arrows).

common infected area [2]. This review revealed that the median age was 51 years, most patients were elderly and were suffering from 1 or more comorbidities, like diabetes mellitus or malignancy. There was no difference between males and females. *Fusobacterium* and *Enterobacteriaceae* were the most frequently found microorganisms. The reported mortality was estimated to be 32% [2].

The causative organisms are generally anaerobes or members of the *Enterobacteriaceae* family; however, the infection can be mono- or poly-microbial [2]. The monomicrobial causes of emphysematous osteomyelitis are similar to the causes of other emphysematous infections, which include *Escherichia coli*, *Klebsiella pneumoniae*, *Enterobacter aerogenes*, and *Clostridium* [2, 5].

When a clinical suspicion of emphysematous osteomyelitis is raised, antibiotics with activity against the common causative bacteria should be administered [1,2,6]. Details regarding effective antibiotic duration for the treatment of emphysematous osteomyelitis are not available. Emphysematous

osteomyelitis is a rare infection that should be considered whenever intraosseous gas is seen, particularly in the extra-axial skeleton [2,7]. Aggressive antimicrobial and surgical intervention is also required, as emphysematous osteomyelitis is a highly fatal entity [2].

Emphysematous pyelonephritis is an uncommon necrotizing infection with a predilection for diabetic patients. EP disproportionately affects females, with a female: male ratio of 3: 1 [8]. This increased occurrence in women could be explained by their increased susceptibility to urinary tract infection (UTIs). *Escherichia coli*, *Klebsiella pneumoniae*, *Proteus mirabilis*, and *Pseudomonas aeruginosa* are the most frequent pathogens [9].

The diagnosis is confirmed based on the radiological findings. Abdominal CT is the most useful and sensitive tool [11,12]. The treatment should be intensive with vigorous fluid resuscitation, systemic administration of antibiotics, strict glycemic control, and careful control of electrolytes. Before the development of interventional radiology, early surgery and nephrectomy was a significant part of the therapeutic treatment plan [10].

Percutaneous drainage was implemented in some cases and this procedure had a successful outcome in carefully selected patients [17–19]. Conservative treatment is associated with a mortality rate of 70%, while surgical treatment is characterized by 30% mortality [13]. Generally, surgical treatment is considered the criterion standard in patients with risk factors.

To the best of our knowledge, coexistence of emphysematous pyelonephritis and emphysematous osteomyelitis of spine has not been previously reported in the relevant literature. In our case report, we describe a patient without a medical history of diabetes mellitus diagnosed with complicated urinary tract infection (UTI), as *Klebsiella pneumoniae* was identified in urine culture. Abdominal and pelvic CT scan revealed emphysematous pyelonephritis and L3–L4 emphysematous osteomyelitis

of the spine. The infection possibly extended through transient bacteremia from urinary tract to the L3 and L4 lumbar vertebral bodies and epidural space.

Conclusions

We report a rare case of both emphysematous osteomyelitis and emphysematous pyelonephritis in a patient with uncontrolled hyperglycemia. This case emphasizes the importance of a high index of suspicion for these rare infections in patients with type 2 diabetes mellitus. It also underlines the important role and contribution of computed tomography in the diagnosis of these entities and timely initiation of aggressive treatment.

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