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## Case report

# Severe sepsis secondary to emphysematous cystitis



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### KEYWORDS

Emphysematous cystitis;  
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### Abstract

Emphysematous cystitis (EC) is a rare, rapidly progressive infection characterized by gas formation within the bladder wall. Gas production is mainly due to fermentation of glucose or albumin by the pathogenic organisms. We present the case of a patient with EC who presented with severe sepsis of unknown origin. He was also found to have two vesical calculi and a horseshoe kidney. EC was diagnosed on ultrasonography which showed circumferential, acoustic shadowing around the bladder wall. Computed tomography (CT) confirmed the diagnosis. Rapid healing could be achieved with appropriate antibiotics and bladder drainage. Endoscopic disintegration of the stones was performed three weeks later, and follow-up was uneventful.

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## Introduction

Emphysematous infections of the urinary system are rare infections characterized by gas producing pathogens in the urinary system. Emphysematous pyelonephritis is well known to be a life-threatening infection, while EC is less aggressive and has a good prognosis with rapid healing when appropriately treated [1]. The presentation of EC varies widely, ranging from asymptomatic features to septic shock. Herein, we present a new case of EC in a patient who presented with severe sepsis in the absence of known risk factors. The patient was also found to have two vesical calculi and a horseshoe kidney.

## Case report

A 46-year-old man presented with bilateral flank pain, predominantly on the left side, minimal dysuria and fever for the previous 7 days. His past medical history revealed that he had had open surgery for appendicular peritonitis four years before.

Physical examination revealed a temperature of 39 °C, normal blood pressure and a pulse rate of 103 bpm. The patient's face had a grayish color with mucosal jaundice. On palpation, the abdomen was soft with minimal tenderness in the left flank. The patient's urine was dark and cloudy.

Laboratory work-up showed a high creatinine level of 3.8 mg/dl and a normal glucose level. Blood count showed 37.000 white blood cells/mm<sup>3</sup>, thrombocytopenia with a platelet count of 30.000/mm<sup>3</sup>

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**Figure 1** Ultrasonography showing circumferential, acoustic shadowing around the bladder wall.

and anemia with a hemoglobin level of 9.5 g/dl. The C-reactive protein (CRP) was 332 mg/dl. The hepatic panel revealed hyperbilirubinemia with a bilirubin of 40  $\mu\text{mol/L}$  and a direct bilirubin of 34  $\mu\text{mol/L}$ . The transaminase enzyme levels were normal.

Plain abdominal radiography performed in the emergency unit was normal.

On ultrasonography, moderate bilateral dilatation of the urinary tract, a horseshoe kidney and circumferential acoustic shadowing around the bladder wall were seen (Fig. 1). Non-contrast-enhanced abdominal CT revealed moderate bilateral dilatation of the urinary tract, predominantly on the left side, a horseshoe kidney, and the presence of two small vesical stones and diffuse gas throughout the bladder wall.

The diagnosis of EC with bladder calculi was made. The patient was hospitalized, and after taking urine and blood samples for urinalysis and blood culture, intravenous empiric antibiotic therapy was started. An 18 Fr urinary catheter was inserted which drained cloudy urine with gas bubbles, indicating the diagnosis of pneumaturia. Since the urine culture revealed *Klebsiella pneumoniae* resistant to ampicillin, ofloxacin and cephalosporins, the empiric antibiotics were changed to Imipenem. Blood cultures performed on admission had been negative.

The clinical and biological conditions of the patient improved after one week. A repeat abdominal CT, performed 10 days later, showed complete resolution of the gas in the bladder wall. Three weeks later, endoscopic transurethral disintegration of the calculi was carried out. The urinary catheter was removed on the following day. The antibiotics were administered for a total of 6 weeks. On follow-up, the patient was asymptomatic with normal findings on ultrasonography, normal chemistry tests and negative findings on urinalysis.

## Discussion

EC is a rare disease with about 200 cases published on Medline. It is a rapidly progressive infection characterized by gas formation within the bladder wall with or without intra-luminal air. It should be differentiated from pneumaturia which is the presence of air in the bladder lumen, mostly due to vesico-colic fistulae which may occur when procedures other than endoscopy are used [2].

The pathogenesis of EC is not completely understood, but gas production is mainly due to fermentation of glucose or albumin by the pathogenic organisms. This gas consists of nitrogen, hydrogen, oxygen and carbon dioxide [3].

The presence of gas-producing organisms, a high tissue glucose concentration and impaired tissue perfusion are all in favor of the development of emphysematous infections of the urinary tract [3].

A high glucose concentration within the tissues promotes the production of carbon dioxide through natural fermentation processes [4]. In fact, diabetic patients represent more than 60% of all reported cases of EC according to the literature [5,6]. However, in non-diabetic patients fermentation of urinary albumin may be the cause of gas production [7]. Another suggested factor promoting gas production is an impaired host response, involving vascular compromise and impaired catabolism within the tissues [5].

Persons at risk of the disease are the elderly, women, diabetics and patients with an immunosuppressive co-morbidity and/or urinary tract outlet obstruction, such as a neurogenic bladder [5,8]. Our patient had none of these factors; however, his condition was life-threatening. It is not known whether the calculi found in the bladder had formed in situ or had migrated from the upper system. While the presence of moderate dilatation in the upper urinary system would be indicative of stone migration, the macroscopic shape of the calculi would suggest in situ composition.

The clinical presentation of EC varies widely and ranges from asymptomatic features or minimal lower urinary tract symptoms to severe sepsis. According to a review of the literature, the most frequent symptoms and findings on physical examination are abdominal pain, pneumaturia after catheterisation and urinary retention, noted in 80%, 70% and 60% of the patients, respectively [6]. Our patient had jaundice which was due to severe sepsis and resolved after one week following the administration of antibiotics. This sign was also described in another report [8].

Severe sepsis is defined by sepsis and organ dysfunction [9]. This is due to hypoperfusion, which may be present in normally fit adults like our patient who had a normal blood pressure, but with a high creatinine level secondary to severe sepsis. Laboratory test findings were remarkable for thrombocytopenia and hyperbilirubinemia reflecting the severity of urosepsis.

Positive blood cultures can be noted in 50% of the cases [8] with gram-negative bacteria being the causative pathogens in most cases. *Escherichia coli* and *K. pneumoniae* are the organisms most often isolated [5]. Other organisms found are *Pseudomonas aeruginosa*, *Proteus mirabilis*, *Candida albicans* and *Aspergillus fumigatus*.

The diagnosis of EC is established by radiological findings. Plain abdominal radiography may show a radiolucent curvilinear area drawing the bladder wall [8], but this is rather uncommon. Ultrasonography usually shows bladder wall thickening with marked echogenicity, but the sensitivity of this sign is low (46%) [6,8]. A pathognomonic sign of circumferential, acoustic shadowing around the bladder wall is rarely noted. Diagnosis is usually established by CT showing diffuse gas throughout the bladder wall. The sensitivity of CT is high, even with non-contrast-enhanced CT. Contrast-enhanced CT is useful when a vesico-colic fistula is suspected. Some authors perform cystoscopy for EC, but this

examination is not essential for diagnosis. When it was done in one of the cases reported in the literature, it showed diffuse submucosal emphysema [10].

Treatment of EC consists of the administration of antibiotics which should be adjusted to the results of urine culture, bladder drainage and the treatment of co-morbid disorders. In our case, parenchymal infection did not worsen the prognosis, but it was the reason for extending the duration of treatment to 6 weeks. In other published cases, the duration of treatment varied from 1 to 3 weeks [6,10]. The prognosis of EC is generally good with an improvement of the patient's condition within the first week. Complications are noted in 10–20% and the mortality rate is low (7%) [1,5,6].

#### Conflict of interest

The authors have no conflicts of interest to declare.

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