

# CASE REPORT

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## A RARE CASE REPORT OF EMPHYSEMATOUS CYSTITIS IN A DIABETIC PATIENT

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**ABSTRACT:** Emphysematous cystitis is a rare disease caused by gas fermenting bacterial and fungal pathogens. Clinical symptoms are non-specific and diagnostic clues often arise from the unanticipated imaging findings. The clinical manifestations can vary from an incidental diagnosis on abdominal imaging to severe sepsis. Gas-forming infections of the urinary tract (emphysematous nephritis, pyelitis and cystitis) are potentially life-threatening and require prompt evaluation and management. We describe a case of a male patient with diabetes and neurogenic bladder due to spinal cord injury diagnosed to have emphysematous cystitis that was managed successfully with strict glycemic control, intravenous antibiotics, bladder irrigation with antibiotics and cystoscopy removal of necrotic tissue.

**KEYWORDS:** pneumaturia, emphysematous cystitis, E. Coli, Pyelonephritis, Pyelitis.

**INTRODUCTION:** Emphysematous cystitis is an uncommon manifestation of infection of the urinary bladder produced by gas forming organisms. The presentation may be atypical and contrary to the degree of inflammation. Patients may present with few clinical findings. A high index of suspicion, especially in susceptible populations, is needed. We report a case of elderly-aged, poorly controlled, diabetic male who presented with dysuria, fever and pneumaturia and was found to have *Escherichia coli* emphysematous cystitis that resolved completely with antibiotic treatment.

**CASE REPORT:** A 52-year-old male with type 2 diabetes mellitus and traumatic paraplegia with neurogenic bladder (with indwelling catheter with periodical change) since 3 years, presented to our emergency department. He complained of fever with chills of one week duration, haematuria and acute urinary retention with vomiting, abdominal and loin pain. He also complained of paraesthesia of feet and weight loss. His past medical history is significant for coronary artery disease, hyperlipidemia, hypertension and insulin dependent diabetes mellitus (most recent HbA1c of 9.7 two weeks prior). He had 48-pack year history of smoking.

On physical examination, his temperature was 100.9°F, pulse 88, BP 123/71 mm/hg. Cardiac and pulmonary examinations were unremarkable. His abdomen was soft, tender with positive bowel sounds and no organomegaly detected. The rectal examination was normal with a non-tender, smooth prostate. On neurological exam, there was no renal angle tenderness. Sensory system examination was normal and patient had lower limb muscle wasting, absent DTRs and extensor plantar reflex.

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**INVESTIGATIONS:** Laboratory evaluation revealed leucocytosis (white cell count, 13,600 cells/mm<sup>3</sup>) with predominant neutrophils (90%), mild anemia (haemoglobin, 10 gm/dl) and plasma glucose of 310 mg/dl. Urine analysis revealed red, cloudy urine and red blood cells on microscopy. Serum creatinine was 2.0 mg/dl and HbA1c was 9%. Urine culture grew *Escherichia coli* with sensitivity to piperacillin/tazobactam.

Ultrasound abdomen showed collection of air within bladder wall and lumen (Fig. 1.) and X-ray KUB also showed a semicircular gas shadow in bladder region (Fig. 2.). CT abdomen revealed intraluminal and intramural gas in the bladder with thickening of bladder wall [Fig. 3.]. Kidneys were normal in size and shape and there was no perinephric stranding or gas in renal parenchyma or renal pelvis.

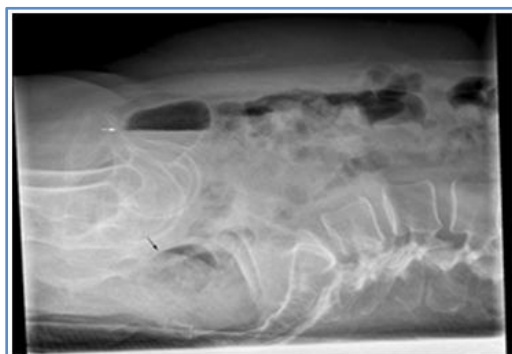
Later Cystoscopy was done; it revealed necrotic mucosal patches with slough covering bladder mucosa and vesicular lesions over lateral walls and dome of bladder. While taking biopsy, air bubbles coming out from bladder wall were observed.

Patient was managed with piperacillin/tazobactam for 2 weeks and oral levofloxacin and nitrofurantoin for another 4 weeks along with bladder catheterization. Good glycemic control was established with insulin therapy.

Patient improved clinically, became afebrile and repeat urine culture was sterile. Figure 1d shows disappearance of intra mural and intraluminal gas on follow up CT imaging after 8 weeks. Serum creatinine was decreased to 1.1 mg/dl.



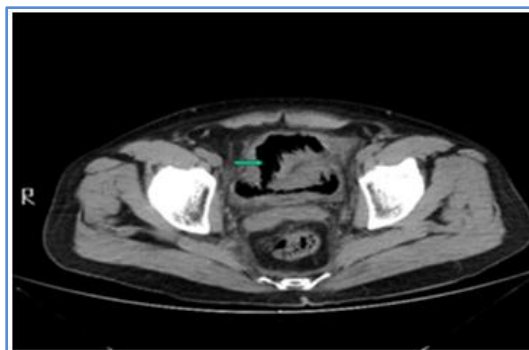
**Figure 1**



**Fig. 2: X-ray of pelvis showing gas in the urinary bladder wall (Arrow)**

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**Fig. 3: CT scan of the pelvis revealing gas in the bladder and the bladder wall**

**DISCUSSION:** Emphysematous cystitis is a rare condition, often occurring in diabetic and immunocompromised patients and the first case was reported by Hueper in 1926.<sup>[1]</sup> There has been a recent increase in reported cases due to a wider use of abdominal imaging and a greater awareness of this uncommon disease.<sup>[2]</sup> The organism most commonly responsible is *E. coli* (58%). Other organisms reported include *Klebsiella pneumoniae*, *Pseudomonas aeruginosa*, *Proteus mirabilis*, *Candida albicans*, and *Candida tropicalis*, *Aspergillus fumigatus*, *Staphylococcus aureus*, Group D *Streptococcus*, *Enterococcus faecalis*, *Enterobacter aerogenes*, and *Clostridium perfringens* and *C. welchii*.

The clinical presentation of emphysematous cystitis is varied; patients can be asymptomatic, describe hematuria, or irritative voiding symptoms, or present with an acute abdomen with severe sepsis.<sup>[3]</sup> Of reported patients with emphysematous cystitis, 7% were asymptomatic and were diagnosed incidentally on abdominal imaging for other concurrent illnesses. Emphysematous cystitis is a rare entity characterized by pockets of gas in and around the bladder wall produced by bacterial or fungal fermentation.<sup>[4,3]</sup> Patients may complain of irritative symptoms, abdominal discomfort or pneumaturia. A history of pneumaturia is highly suggestive.<sup>[5-8]</sup> The radiographic findings provided the first and only diagnostic clue.

The disease is often associated with female sex, immunocompromised state, diabetes mellitus, previous recurrent urinary tract infections, urinary stasis, neurogenic bladder and in transplant recipients.<sup>[2]</sup> Therefore, in susceptible patients, with the above risk factors along with signs and symptoms of urinary tract infection, the index of suspicion for this entity should be high. The most common organism is *E. coli*,<sup>[3]</sup> but other organisms reported to produce emphysematous cystitis include *Enterobacter aerogenes*, *Klebsiella pneumoniae*, *Proteus mirabilis*, *Staphylococcus aureus*, streptococci, *Clostridium perfringens*,<sup>[9]</sup> and *Candida albicans*.<sup>[10]</sup> The mechanism by which gas appears in the wall of the bladder may involve either transluminal dissection of gas or true infection of the bladder wall with pathogens.

Diagnostic entities associated with gas in the genitourinary tract include emphysematous pyelonephritis, emphysematous pyelitis, and gas-forming renal abscess. Patients with emphysematous cystitis are not as acutely ill as those with pyelonephritis or pyelitis. Abdomino-pelvic CT scan can further delineate the extent of disease. It is important to differentiate emphysematous cystitis from emphysematous pyelonephritis, in which gas involves the renal

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parenchyma, since the latter has an increased mortality and generally requires nephrectomy. In contrast surgical intervention is rarely needed in emphysematous cystitis except when an anatomical abnormality like an obstruction or stone is present.<sup>[11]</sup>

The source of this gas within the urinary tract is from infection, trauma, vesico-enteric fistulas from radiation therapy, rectal carcinoma, diverticular disease or Crohn's disease and iatrogenic causes, such as diagnostic or surgical instrumentation. History, physical exam and imaging are the best modalities to differentiate the above etiologic causes. Fistulous tracts, abscess, can be excluded on CT scan.

Emphysematous cystitis requires aggressive treatment with parenteral antibiotics and bladder drainage.<sup>[11]</sup> Delayed diagnosis may lead to unfavorable outcomes including overwhelming infection, extension to ureters and renal parenchyma, bladder rupture and death. Improved outcomes may be achieved by early recognition of the infection, by clinical and radiological assessment, and by appropriate antibiotic therapy.

**CONCLUSION:** Emphysematous cystitis most often is not diagnosed by routine or systematic approach. It is a rare entity, detected on imaging, and the physician should be cautious and the diagnostic approach to individual patients is based on clinical suspicion, available clinical and radiological data, and considers emphysematous cystitis in the differential diagnosis of pneumaturia in a patient with known risk factors.

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