

# Emphysematous cystitis: A case report and literature review of 113 cases

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## Abstract

Emphysematous cystitis (EC) is a rare complicated urinary tract infection characterized by gas formation within the wall of the urinary bladder. Although EC has multifactorial etiology, commonly seen in elderly, diabetic and female sex; most of the cases of EC present along with emphysematous pyelonephritis (EPN) and full-blown urosepsis. We present a case of EC with EPN presented with features of sepsis managed conservatively. Furthermore, we reviewed the literature of published cases reports of EC with or without EPN from 1999 to 2019 (20 Years). From eligible 113 case reports, data of clinical presentation, demographic profile, risk factors, diagnostic methods, treatment, and prognosis were analyzed.

**Keywords:** Emphysematous cystitis, emphysematous pyelonephritis, literature review, urosepsis

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## INTRODUCTION

Emphysematous cystitis (EC) is a rare life-threatening infection of bladder wall by gas forming microorganisms, characterized by misleading nonspecific clinical features. Bailey in 1961 defined EC as “cystitis emphysematosa” and linked it with pneumaturia.<sup>[1,2]</sup> Clinical presentation of EC is variable ranging from incidental detection on imaging to septic shock.<sup>[3]</sup> The detection of EC is increasing because of frequent use and advancement in cross-sectional imaging leading to better understanding of this rare diseases. The aim of the present study is to revisit this urological emergency with review of existing literature. All articles published in English literature in PubMed® from 1999 to 2019 identified using keywords “Emphysematous” and “cystitis.” Total 267 publications were found and after exclusion of animal studies, paper in languages other than English, review articles and case

series, 113 case reports were eligible for analysis and described here in discussion.

## CASE REPORT

A 62-year-old male known case of type II diabetes mellitus for the past 6 months on oral hypoglycemic agents presented to emergency with left flank pain, intermittent fever with chills, suprapubic pain, generalized weakness for 7 days and burning micturition associated with foul smelling cloudy urine for 3 days. His vitals at presentation were temperature of 101°F, pulse 102/min, BP108/78 mm of Hg, and respiratory rate of 22/min. Abdomen was soft with mild tenderness in suprapubic region and renal angle. In laboratory parameters, his blood sugar was 370 mg/dl, HbA1C 11.5%, total leukocyte count 17070/mm<sup>3</sup> (neutrophils

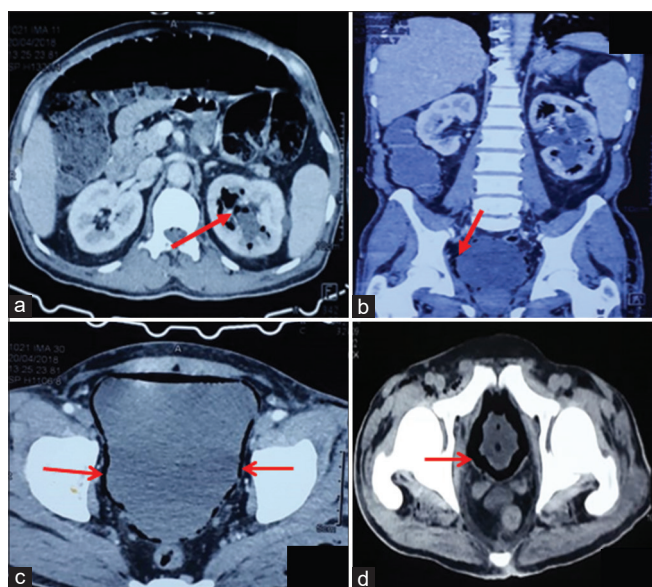
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90%), hemoglobin 7.9 mg/dl, blood urea 80 mg/dl, serum creatinine 1.03 gm/dl, and serum bilirubin was 1.35 mg/dl. Provisional diagnosis of urosepsis was made and urine was sent for routine microscopy and culture. The patient was started on piperacillin–tazobactam and intravenous crystalloid. For strict sugar control regular insulin on sliding scale with blood sugar charting was started. Ultrasonography showed left pyelonephritis and floating echoes in bladder. Foley catheterization was done in emergency and 400 ml of turbid urine was drained instantly. Computed tomography (CT) revealed left hydronephrosis, tiny calculi, multiple air foci in left renal pelvis and parenchyma suggestive of grade II emphysematous pyelonephritis according to classification by Huang *et al.*<sup>[4]</sup> [Figure 1]. There was classical finding of “beaded necklace” pattern of gas in the wall of urinary bladder which confirmed the diagnosis of EC [Figure 1]. After primary resuscitation, the patient underwent cystoscopy and left double J stenting which showed multiple air filled submucosal vesicles in the bladder, further strengthening the diagnosis of EC [Figure 2]. Urine culture grew *Escherichia coli* ( $>10^5$  colonies/ml) which was sensitive to previous antibiotics. The same injectable antibiotic continued until leukocyte count comes to normal. The patient became afebrile after 48 h and was discharged on 7<sup>th</sup> day advising culture directed oral antibiotic (levofloxacin 750 mg once a day for next 2 weeks).



**Figure 1:** Computed tomography scan (a) axial section showing left emphysematous pyelonephritis with air foci in the left renal parenchyma and pelvicalyceal system (b) coronal section showing air in bladder wall (red arrow) (c) full bladder and (d) empty bladder axial section of computed tomography scan showing classical “beaded necklace” appearance of air in the wall of urinary bladder

## DISCUSSION

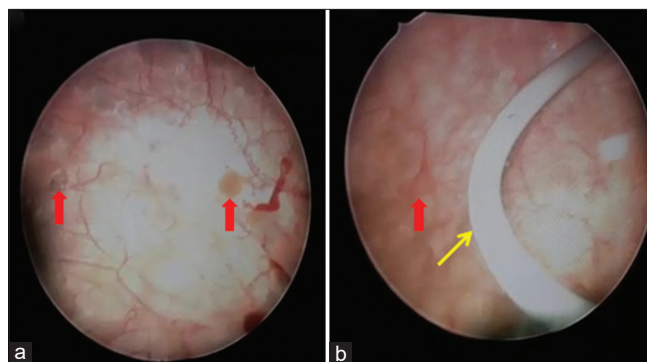
In 113 identified cases of EC in literature, mean age of presentation was 67.94 (11–96) years. Most cases were seen in female (65.49%) and age older than 60 years (75.22%). Apart from age and sex, the most common predisposing factor was diabetes mellitus (60.18%) and comorbidities ( $>2$  comorbid conditions in 49.55%). Other risk factors were neurogenic bladder (9.73%), malignancy on chemotherapy (8.85%), immunosuppressed state (transplant recipient), and postsurgery [Table 1].

Most patients presented with features of sepsis (32.74%) and uncomplicated urinary tract infection (16.81%). Sepsis is defined as systemic inflammatory response syndrome with documented infection.<sup>[5]</sup> Other modes of presentation were pain abdomen (25.66%), hematuria (6.19%), and fever (3.54%). Nine (7.96%) patients were diagnosed incidentally on imaging for other reason.

Five (4.42%) patients had spontaneous bladder perforation associated with EC. Out of these, open repair of intraperitoneal perforation was done in two cases. Another patient with extra peritoneal perforation was managed conservatively and two patients required emergency cystectomy.<sup>[6,7]</sup>

Confirmatory diagnosis was made on non-contrast computed tomogram of abdomen in 108 (95.57%), on X-ray of abdomen in 2 (1.78%), on exploratory laparotomy, on transurethral biopsy done for suspicion of bladder mass and on fluorodeoxyglucose positron-emission tomography/ CT done for follow-up of nodular sclerosis in one each.

Details of urine culture were available in 93 patients. *E. coli* was the most common pathogen 61/93 (65.59%), followed by *Klebsiella* (22.58%), *Enterococcus* (3.22%),



**Figure 2:** Cystoscopy (a) multiple submucosal air filled vesicles (red arrows). (b) Vesicles (red arrow) with coil of the left DJ stent (yellow arrow)

**Table 1: Demographic profile, risk factors, diagnostic method, and the most common isolated urinary pathogen of the reviewed population**

Variable	Value (n=113)
Mean age	67.94 (11-96)
Age >60 years, n (%)	85 (75.22)
Predisposing/risk factors, n (%)	
Female	74 (65.49)
Diabetes mellitus	68 (60.18)
Diabetic female	45 (39.82)
Age >60 years, diabetic	51 (45.13)
Malignancy	10 (8.85)
Neurogenic bladder	11 (9.73)
Confirmatory diagnosis, n (%)	
CT scan	108 (95.57)
X-ray	2 (1.78)
Exploratory laparotomy	1 (0.88)
Biopsy	1 (0.88)
PET/CT (incidental)	1 (0.885)
Chief presentation (113), n (%)	
Sepsis	37 (32.74)
Pain abdomen	29 (25.66)
UTI	19 (16.81)
Incidental	9 (7.96)
Hematuria	7 (6.19)
Others	12 (10.61)
Urine culture (93), n (%)	
<i>Escherichia coli</i>	61 (65.59)
<i>Klebsiella</i>	21 (22.58)
<i>Enterococcus</i>	3 (3.25)
<i>Candida</i>	3 (3.25)
<i>Citrobacter</i>	2 (2.15)
Sterile	3 (3.25)

CT: Computed tomography, PET: Positron emission tomography, UTI: Urinary tract infection

*Candida* (3.22%), and *Citrobacter* (2.15%). The most common antibiotic used (available in 49 cases) as empirical treatment was piperacillin–tazobactam (20.40%) and meropenem (20.40%) followed by ceftriaxone (18.37%) and injection ciprofloxacin (12.24%).

Only 2 patients died; one 71 years old renal transplant recipient male patient with recent history of transurethral resection of prostate and another 81 years old female with septic shock and uncontrolled diabetes mellitus.<sup>[8,9]</sup>

A review of 135 cases of EC by Thomas *et al.* reported that median age of patients was 66 years, 64% were female and 67% had diabetes mellitus, which is comparable to present analysis but overall mortality rate was 7% which is higher than present study (1.78%).<sup>[3]</sup> Low mortality in the present review may be attributed to early diagnosis and prompt treatment because of better understanding of disease in the current era of advanced imaging.

Clinical presentation of EC is highly variable; patient can be asymptomatic or may present with sepsis and septic shock. According to Grupper *et al.*, the most common symptom in their series was abdominal pain and classic

symptoms of UTI were seen in only approximately 50% of the cases.<sup>[10]</sup>

Plain conventional radiography of abdomen was commonly used in past to clinch the diagnosis of EC but due to high sensitivity of CT scan for detection of air foci and to effectively rule out other possible differential etiologies; currently CT scan is the imaging of choice.<sup>[11]</sup> Three stages of EC have been described in literature based on imaging.<sup>[12]</sup> In Stage 1, a clear 1-mm zone may be seen around the contrast medium and no free gas is seen in the bladder lumen. In Stage 2, the bladder wall is irregular and thickened because of increased intramural gas, and still there is no free intraluminal gas. In Stage 3, free gas in the bladder lumen is evident radiographically.

A study by Amano *et al.* of 72 cases outside Japan and 30 inside Japan, showed that *Escherichia coli* (56% and 64%) and *Klebsiella pneumoniae* (41% and 25%) were the two major organisms isolated in urine cultures.<sup>[13]</sup>

Bjurlin *et al.* reported that 90% of cases of EC can be managed conservatively with medical management and only 5%–10% of cases need some surgical intervention.<sup>[3,14]</sup> Medical management consists of culture directed antibiotics, intravenous fluid, bladder drainage, strict control of blood sugars, and treatment of predisposing conditions. The total duration of antibiotic course varied between 2 and 3 weeks.<sup>[2]</sup>

## CONCLUSIONS

EC is a rare urological emergency and is potentially life-threatening which needs early diagnosis and prompt resuscitation. Bladder drainage, broad spectrum antibiotics, and treatment of predisposing factors are the main stay of treatment.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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