

Case Report

## Emphysematous Cystitis in a Patient with Type 2 Diabetes Mellitus

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A 62-year-old woman with a history of poorly controlled type 2 diabetes mellitus was admitted to our hospital with a 3-week history of mild fever, vomiting, and anorexia. Abdominal computed tomography (CT) showed bilateral hydronephrosis and gas accumulation in the urinary bladder wall and left ureter. Laboratory tests showed leukocytosis and elevated C-reactive protein level. Urine culture showed heavy growth of *Escherichia coli*. The final diagnosis was emphysematous cystitis. The patient was treated with systemic antibiotics and drainage using a urethral catheter. The clinical and radiographic findings resolved rapidly, and she was discharged from the hospital on day 28. Emphysematous cystitis is a relatively rare urinary tract infection associated with gas formation, and has the potential for a serious outcome if untreated. Early detection by imaging studies such as CT is important in providing prompt treatment and favorable clinical outcome.

**Key words:** computed tomography, diabetes mellitus, emphysematous cystitis

**E**mphysematous cystitis is a relatively rare infection characterized by accumulation of air within the urinary bladder wall and the bladder lumen [1]. Diabetes mellitus, neurogenic bladder, and dysuria are the major risk factors of emphysematous cystitis [2]. The clinical presentation varies from asymptomatic to fatal; one case presented with fulminant septic shock, and another case developed bladder rupture [1]. Lack of improvement following conservative medical management necessitates bladder extraction in some cases. Here we report a case of emphysematous cystitis with early diagnosis and successful treatment.

### Case Report

A 62-year-old woman was admitted to our hospital with 3-week history of mild fever, vomiting, and anorexia. She had received regular follow-up at the local clinic for 10 years to manage her type 2 diabetes mellitus, hypertension, neurogenic bladder, and chronic hepatitis C. In spite of intensive insulin therapy (8 units of insulin lispro before each meal and 14 units of insulin glargine at bedtime), her hyperglycemia was difficult to control due to poor compliance and HbA<sub>1C</sub> was approximately 12%. She did not have diabetic nephropathy, but had retinopathy and autonomic neuropathy including orthostatic hypotension and neurogenic bladder, and she suffered frequent urinary tract infection. She had first consulted her local physician, and her symptoms had improved, but only temporarily; thus, she was referred to our hos-

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pital for further management.

The first impression of the patient at physical examination was that she was thin. The radial pulse was regular at 87 beats/min. Body temperature was 36.5°C, blood pressure was 165/95 mmHg, and arterial oxygen saturation at rest measured by pulse oximetry was 97%. Consciousness was clear. The lungs were clear on auscultation, and the heart sounds were normal with no murmurs. The lower abdomen was distended up to the umbilicus with no tenderness. There were no peritoneal signs, and the bowel sounds were normal. She was not taking any medication that induces neurogenic bladder, and did not report any urination trouble on admission.

The laboratory evaluation on admission is summarized in Table 1. The white blood cell count of 10,500/ $\mu$ l and C-reactive protein (CRP) level of 7.13 mg/dl were indicative of an inflammatory process. Urinalysis showed macroscopic hematuria, proteinuria, and pyuria, suggestive of urinary tract infection. Urine culture revealed growth of *Escherichia coli* of  $1 \times 10^7$  CFU/ml. Fasting plasma glucose was 281 mg/dl, and HbA1c was 10.9%. A plain radiograph of the abdomen showed air in the lumen of the urinary bladder, with the formation of an air-fluid level (Fig. 1). Abdominal computed tomography (CT) showed left hydronephrosis and an accumulation of air in the wall of the urinary bladder and left ureter (Fig. 2A-C). There were no findings of ureteric obstruction, emphysematous pyelonephritis, or pelvic organ prolapse

including cystocele and uterine prolapse. Contrast-enhanced CT at day 11 of hospitalization revealed no findings of obstructive pyelonephritis or emphysematous pyelonephritis (Fig. 3).



Fig. 1 X-ray of the abdomen taken on admission. Air-fluid level was detected within the pelvis.

Table 1 Results of laboratory tests

Urinalysis		CBC		$\gamma$ -GTP	61 IU/L
Color	Dark red	Leukocytes	10,500/ $\mu$ l	LDH	40 IU/L
pH	6.0	Red blood cells	$3.72 \times 10^6$ / $\mu$ l	Na	139.1 mEq/L
Protein	2+	Hemoglobin	12.1 g/dl	K	3.55 mEq/L
Occult blood	3+	Hematocrit	35.4%	Cl	90 mEq/L
Leukocytes	3+	Platelets	$20.1 \times 10^4$ / $\mu$ l	Ca	9.2 mg/dl
Sugar	3+	Serological tests		P	2.9 mg/dl
Ketone bodies	–	CRP	7.13 mg/dl	T-Chol	156 mg/dl
Urinary sediment		Blood Chemistry		BUN	14.3 mg/dl
red blood cells	>100/HPF	Total Protein	7.2 g/dl	Cr	0.8 mg/dl
leukocytes	>100/HPF	Albumin	3.6 g/dl	UA	3.3 mg/dl
bacteria	2+	AST	75 IU/L	BS	281 mg/dl
		ALT	11 IU/L	HbA1c	10.9%
		ALP	311 IU/L		

CBC, complete blood count; CRP, C-reactive protein; AST, aspartate aminotransferase; ALT, alanine aminotransferase; ALP, alkaline phosphatase;  $\gamma$ -GTP, gamma glutamyl transpeptidase; LDH, lactate dehydrogenase; T-Chol, total cholesterol; BUN, blood urea nitrogen; Cr, serum creatinine; UA, uric acid; BS, blood sugar; HbA1c, glycosylated hemoglobin.

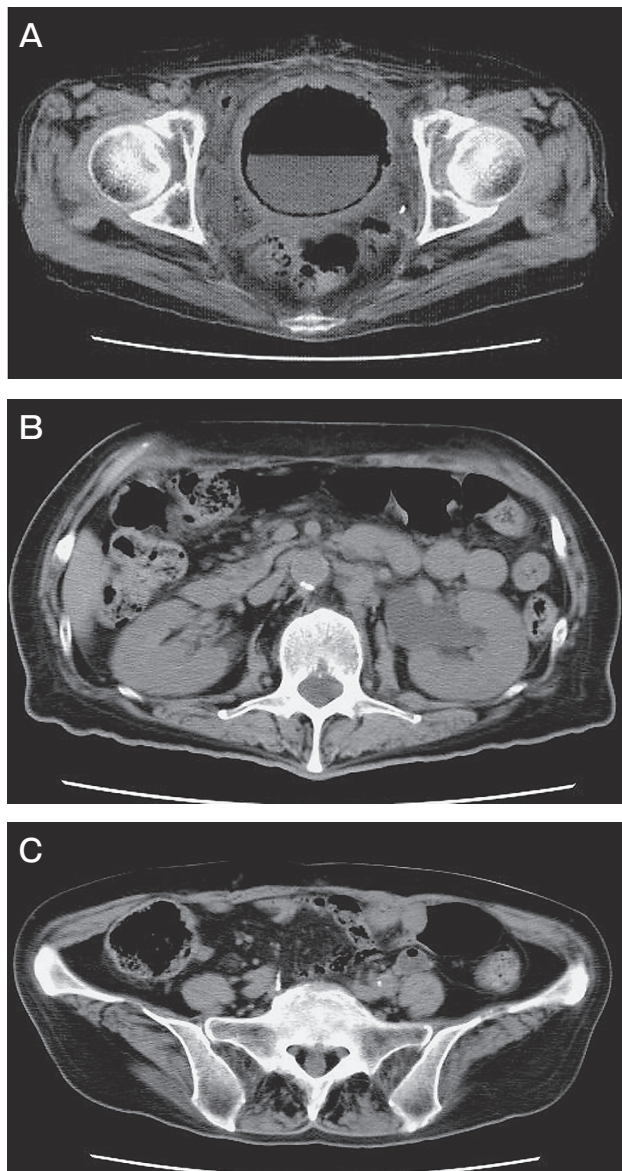


Fig. 2 Computed tomography taken on admission showing (A) gas accumulation in the wall and lumen of the urinary bladder, (B) left hydronephrosis, and (C) gas in the left ureter.

Following drainage through an indwelling urethral catheter, we started antibiotic treatment (cefotiam and levofloxacin). Based on the appetite loss of the patient and the persistent hyperglycemia ( $\sim 400$  mg/dl) on admission, she was placed on an intravenous infusion of fluid and insulin. Her appetite recovered gradually, along with improvement of urinary findings and decreases of CRP and blood glucose levels, so we restarted intensive insulin therapy (Fig. 4). Abdominal



Fig. 3 Contrast-enhanced computed tomography taken at day 11 of hospitalization. Left hydronephrosis is improved compared with the image on admission (Fig. 2A). There were no findings of obstructive pyelonephritis or emphysematous pyelonephritis.

CT at day 11 of hospitalization showed persistent bladder wall emphysema but collapse of the bladder (Fig. 5A), and improvement of the left hydronephrosis (Fig. 5B). To prevent urinary tract infection, distigmine bromide was administered for neurogenic bladder due to diabetic neuropathy as well as strict glycemic control and hydration. She was discharged from the hospital on the 28th hospital day with no residual urine after removal of the urethral catheter.

## Discussion

Among the infections associated with diabetes mellitus, urinary tract infection is the most frequent, and it encompasses difficult entities such as emphysematous pyelonephritis and emphysematous cystitis [3, 4]. Emphysematous cystitis is a relatively rare infection, characterized by gas accumulation within the bladder wall and the bladder lumen [5, 6]. 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 [7]. To clarify the etiology and characteristics of emphysematous cystitis, we have summarized the previous case reports and reviews published in English in Table 2.

We identified 153 cases of emphysematous cystitis over the last 50 years. The median age of the cases was 69 years old, and most cases were in women (63.4% vs. 36.6%, ratio 1.7 : 1). Diabetes mellitus is present in two-thirds of patients with emphysematous

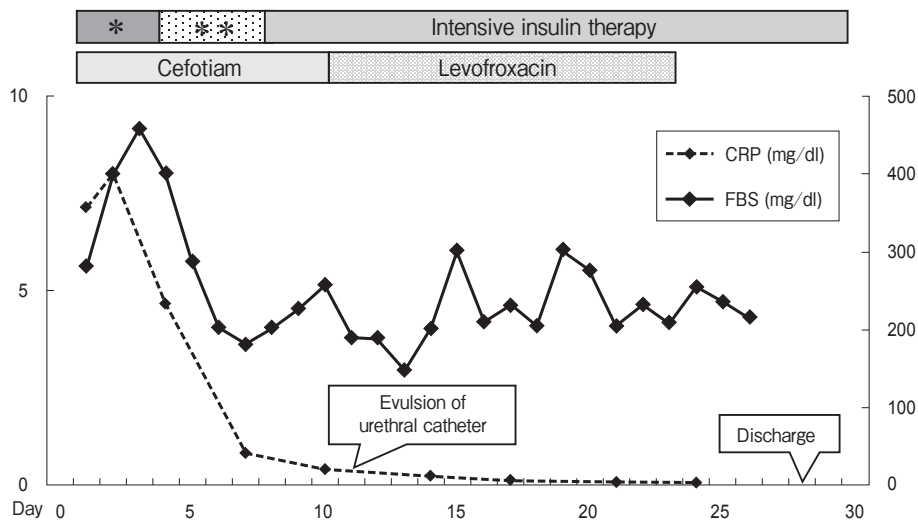


Fig. 4 Clinical course. \*Co-infusion of fluid and subcutaneous injection of insulin based on blood glucose level. \*\*Continuous intravenous infusion of insulin.

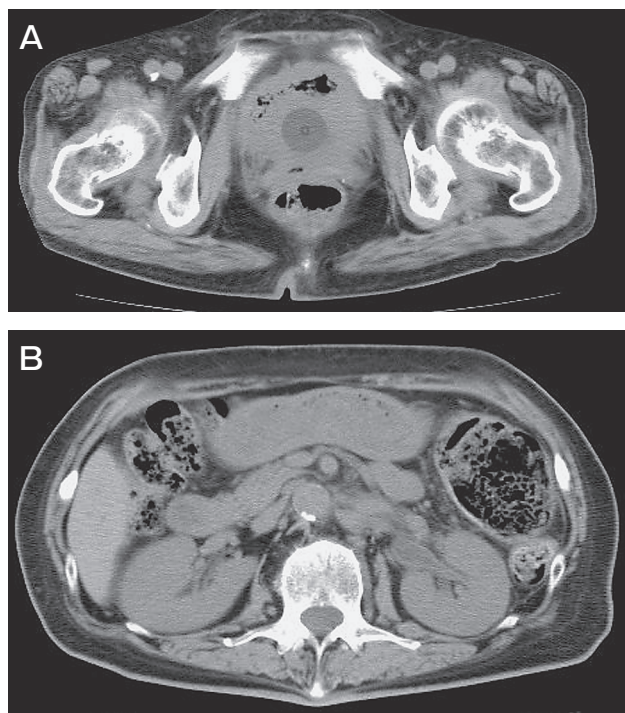


Fig. 5 Computed tomography taken on day 11 showing (A) persistence of emphysema in the bladder wall but collapse of the bladder, and (B) improvement of left hydronephrosis.

cystitis, and is more common in women than in men (70.1% vs. 60.7%). The proportion of type 1 diabetes mellitus is high (42.4%) in cases of emphysema-

tous cystitis compared with the general population (approximately 10%). Glycemic control is usually difficult in type 1 diabetes mellitus due to absolute insulin insufficiency, and it is thought that high glucose concentration within the tissues acts as a favorable substrate for organisms. The most common pathogens in this form of cystitis are *Escherichia coli*, followed by *Klebsiella* species and *Enterococci* [2, 7]. Compared with common urinary tract infections, the frequency of *E. coli* is low [4].

The accumulation of gas produced by bacteria through sugar fermentation in the bladder submucosa and lumen is the direct cause of emphysematous cystitis, but its pathogenesis is poorly understood [1]. Multiple risk factors are considered in the pathogenesis of emphysematous cystitis [8]: 1) persistent hyperglycemia, which provides excess glucose to bacteria, 2) fragility of the infection defense mechanism due to hyperglycemia, 3) dysuria due to diabetic nephropathy, and 4) lower urinary tract obstruction, such as refractory and relapsed urinary tract infection, neurogenic bladder, and benign prostatic hyperplasia.

The overall death rate in the reviewed cases was 7% (Table 2). Though the prognosis of emphysematous cystitis is comparatively good, some cases develop septic shock and bladder rupture [1]. Accordingly, early drainage and treatment with an appropriate antibiotic are important. Prompt diagno-

sis by radiological studies such as CT is important because early treatment leads to a favorable clinical outcome. CT can identify clearly the level and distri-

**Table 2** Demographics and outcomes of the reviewed population, and the comonnest isolated urinary pathogens

Variable	
Mean age, years:	62.7
n/N (%):	
Men	56/153 (36.6)
Women	97/153 (63.4)
DM	102/153 (66.7)
Type 1 DM	25/59 (42.4)
Type 2 DM	34/59 (57.6)
Women with DM	68/97 (70.1)
Women without DM	29/97 (29.9)
Men with DM	34/56 (60.7)
Men without DM	22/56 (39.3)
Overall death rate	10/151 (6.6)
Pathogens (133 cases), n (%):	
<i>Escherichia Coli</i>	76 (57.1)
<i>Klebsiella pneumoniae</i>	29 (21.8)
<i>Enterococcus aerogenes</i>	9 (6.8)
<i>Clostridium perfringens</i>	7 (5.3)
<i>Candida albicans</i>	5 (3.8)
<i>Eberococcus faecalis</i>	3 (2.3)
Group D <i>Streptococcus</i>	3 (2.3)
<i>Pseudomonas aeruginosa</i>	3 (2.3)
<i>Proteus mirabilis</i>	3 (2.3)
<i>Aspergillus fumigatus</i>	1 (0.8)
<i>Candida tropicalis</i>	1 (0.8)
<i>Clostridium welchii</i>	1 (0.8)
<i>Enterobacter cloacae</i>	1 (0.8)
<i>Staphyrococcus aureus</i>	1 (0.8)

DM, diabetes mellitus.

bution of the gas, so it is useful for a definite diagnosis. Bladder extraction might become necessary when the clinical findings do not improve after conservative medical management, so cooperation with urologists is important. Moreover, glycemic control is important because hyperglycemia itself contributes directly to the gas production.

In conclusion, we reported a case of emphysematous cystitis associated with poorly controlled diabetes complicated with neuropathic bladder. Early diagnosis of emphysematous cystitis using a CT scan is necessary to provide prompt treatment by appropriate antibiotics and urinary drainage.

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