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Case Report

Emphysematous cystitis with rare findings of subcutaneous emphysema and extraperitoneal pelvic gas: a case report

Milan C. Gunawardene*, Madura C. Ambegoda, Munipriya A. Willaraarachchi, Anuruddha M. Abeygunasekera

Department of Urology, Colombo South Teaching Hospital, Kalubowila, Sri Lanka

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***Correspondence:**
Dr. Milan C. Gunawardene,
E-mail: milangunawardena@yahoo.com

ABSTRACT

Emphysematous cystitis is a rare form of lower urinary tract infection with pathognomonic intramural and intraluminal gas. It commonly occurs in elderly females with uncontrolled diabetes mellitus. Subcutaneous emphysema and extraperitoneal pelvic gas are reported as rare presentations of emphysematous cystitis. Here we report emphysematous cystitis occurring in an elderly male with multiple co-morbidities who presented with fulminant sepsis and rare findings of subcutaneous emphysema and extraperitoneal pelvic gas.

Keywords: Emphysematous cystitis, Subcutaneous emphysema, Extraperitoneal pelvic gas, Case report

INTRODUCTION

Emphysematous cystitis (EC) is a rare, potentially life threatening, severe necrotizing infection of the urothelium and underlying bladder musculature.[1-3] But, it is less severe urinary tract infection (UTI) compared to other gas forming infections of the urinary tract.[2] Intramural and intraluminal gas from anaerobic fermentation is pathognomonic of EC.[4] This case report describes EC with subcutaneous emphysema and extraperitoneal pelvic gas in a patient with high pressure chronic retention (HPCR) and several other comorbidities.

CASE REPORT

A 77 years old man with diabetes mellitus, hypertension, dyslipidemia, stage-2 chronic kidney disease (CKD) and ischemic heart disease was admitted to emergency treatment unit with high fever, lower abdominal pain, reduced urine out-put and impaired consciousness of one-day duration. He was on an indwelling urethral catheter for HPCR. He was tachycardic (118 bpm), hypotensive (86/54 mmHg) and dyspneic (respiratory rate 22/min, saturation 92%) with a high random blood sugar level (424 mg/dl). His abdomen was distended and severely tender. There was subcutaneous emphysema in the lower anterior abdomen. Catheter revealed purulent urine with pneumaturia. After taking blood and urine for cultures and baseline investigations he was started on intravenous meropenem 500 mg eight hourly and resuscitated according to the surviving sepsis campaign protocol. Investigations revealed neutrophil leukocytosis (WBC-18,000, N%-91%), high C-reactive protein (CRP) level (70 mg/l), moderately field full pus cells and red cells in urinalysis, severe metabolic acidosis (pH-6.99, lactate >15 mmol/l, base excess-17.2 mmol/l) and high serum creatinine (355.6 µmol/l). There was EC with extraperitoneal pelvic gas and subcutaneous emphysema in the non-contrast computed tomography (CT) abdomen and pelvis (Figures 1-3).

His blood and urine cultures were positive for *Escherichia coli* (*E. coli*). Unfortunately, patient died within few hours and post mortem examination revealed severely necrotic urothelium without perforation of bladder wall.

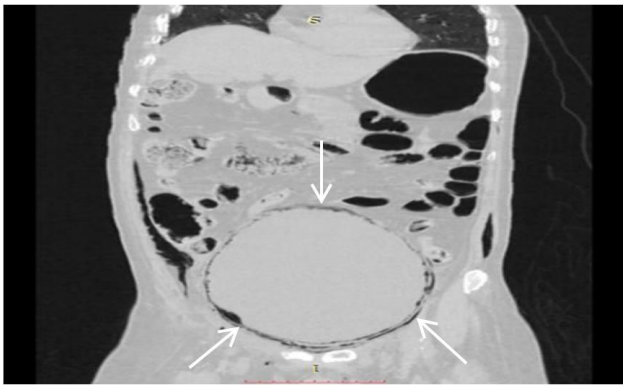

Figure 1: Coronal CT image (lung window) demonstrating air outlining the bladder wall (white arrows).

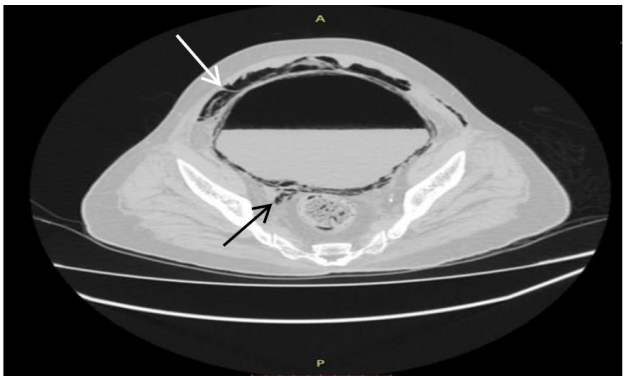

Figure 2: Axial CT image (lung window) demonstrating extraperitoneal pelvic gas in perivesical space (black arrow) and space of Retzius (white arrow).

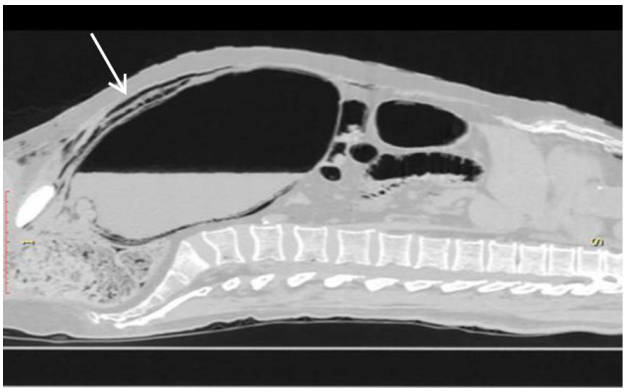

Figure 3: Sagittal CT image (lung window) demonstrating subcutaneous emphysema (white arrow) in lower anterior abdominal wall.

DISCUSSION

EC is an uncommon UTI characterized by presence of intramural and intraluminal gas due to severe necrotizing infection of the urinary bladder. Although most cases are under-diagnosed and under-reported, there is an increased incidence in recent years due to wider use of imaging studies.[4] It is a potentially life- threatening condition with 3-12% overall mortality. But, when concurrently occurs with emphysematous pyelonephritis, its mortality rate increases to 14-20%.[2] Therefore, early diagnosis and treatment is essential. Delayed diagnosis results in bladder perforation, overwhelming infection and death.[4,5]

EC is twice more common in elderly (60-70 years) females than men.[2,3,5,6] Uncontrolled diabetes mellitus (DM) is the major risk factor affecting 70% of patients.[2] Other risk factors include obstructive uropathy, diabetic cystopathy, neurogenic bladder, chronic UTI, long-term indwelling urethral catheters and immunosuppression.[1,2,5,6] Interestingly, there are reported cases in non-diabetic and well controlled diabetic patients as well.[5] Older age, uncontrolled DM, obstructive uropathy and indwelling catheter were risk factors for EC in this patient.

Gas forming uropathogens colonizing the urinary bladder are implicated in the pathogenesis. As in this patient, *E. coli* is the most common organism isolated in 60-70% of urine cultures.[3] *Klebsiella pneumoniae* is the second most common pathogen (10-20%).[2,3,5,6] *Enterobacter aerogenes*, *Proteus mirabilis* and *Streptococcus* species are isolated in 5-8% of cases. Rarely, *Pseudomonas aeruginosa*, *Clostridia*, *Enterococci*, *Staphylococcus aureus*, *Candida* species and aspergillus have also been reported.[2] Polymicrobial infections have been reported in very few cases.[7]

Pathophysiology of intramural and intraluminal gas production is multifactorial. High tissue glucose concentration and impaired tissue perfusion make a hostile environment for emphysematous infections. Gas forming uropathogens ferment glucose and lactate to produce nitrogen, hydrogen, oxygen and carbon dioxide. In non-diabetics, urinary albumin act as the substrate for gas production.[5]

EC patients present with a spectrum of symptoms ranging from asymptomatic to life-threatening fulminant sepsis.[1,4,8] Up to 7% of patients may be asymptomatic and incidentally found on abdominal imaging for other medical conditions.[2] Abdominal pain is the most common symptom reported in 80% of patients. Other common symptoms are pneumaturia (70%), gross hematuria (60%), features of acute cystitis (50%), fever (47.6%), severe sepsis (30-50%) and ischuria (10%).[2,6] Very rarely, as in this patient there may be subcutaneous emphysema which is considered as a poor prognostic factor.[6]

Hematological and biochemical investigations are non-diagnostic. CRP and WBC/DC level indicate the severity of infection and inflammation. Urinalysis cannot differentiate simple cystitis from EC.[8] 50% of patients have bacteremia. Urine gram staining, urine and blood cultures are essential to isolate the pathogen and start appropriate antibiotic therapy.[2] Blood and urine cultures

of this patient had *E. coli* which was sensitive to meropenem.

Imaging is essential to make the definitive diagnosis. Conventional plain abdominal radiography is extremely sensitive (97.4%) and diagnostic in 80% of cases.[4] In abdominal X-ray, curvilinear areas of increased radiolucency delineating the bladder wall represent intramural gas with characteristic cobblestone or beaded neck lace appearance. Intraluminal gas appears as an air-fluid level within the bladder.[2,5] Bladder wall is abnormally thickened in ultrasound scan.[4] Intramural and intraluminal gas appears as highly echogenic regions with dirty acoustic shadowing.[8] These foci of gas can be seen to move dependently with the patient position and referred to as "champagne" or "effervescent" sign.[7] But, none of these investigations were considered in this patient as we were looking for a sinister intra-abdominal pathology.

CT is the gold standard tool for diagnosis of EC.[1,2] It can detect cases which are not apparent on plain radiography. It determines the extent and severity of the condition while differentiating it from other causes for pelvic air.[2,5] It also confirms concomitant emphysematous pyelonephritis.[5] In addition to these, CT of this patient showed extraperitoneal pelvic gas in pre-vesical space of Retzius and peri-vesical spaces including subcutaneous emphysema in anterior abdomen without evidence of bladder perforation (Figure 2 and 3). This is a very rare finding described only in few case reports.[7]

Early medical treatment in 90% of patients reduces mortality and prevent the need for surgical intervention.[2] It includes broad spectrum parenteral antibiotics, immediate bladder drainage, tight glycemic control and treatment of other comorbidities.[3-5] Carbapenems (meropenem, imipenem and doripenem) are indicated as first line therapy. Third generation cephalosporins, aminoglycosides and fluoroquinolones are second line antibiotics.[2] Antibiotic combinations may be considered in patients with hemodynamic instability.[3] More specific antibiotics are indicated when sensitivity pattern is known. Generally, parenteral antibiotics are continued until the patient is apyretic and clinically stable.[3] Median duration of antibiotic therapy is usually ten days.[9] About 10% of patients who do not respond to medical treatment require surgical intervention. Bladder debridement and partial or total cystectomy are the preferred surgical options.[10]

CONCLUSION

EC is a rare clinical entity known to affect primarily patients with uncontrolled DM. Even though uncommon, non-diabetic patients can also be affected. Prevalence of the condition is underestimated as radiological assessment is not routinely done in all patients with UTI. CT is the gold standard investigation to confirm the diagnosis. Early medical therapy improves survival and prevents unnecessary surgical interventions. Delayed diagnosis can result in overwhelming infection, bladder perforation and death.

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