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Emphysematous cystitis revealed by necrotizing fasciitis: An unusual presentation

Sami Fendri, Ahmed Bouzid*, Haitham Rejab, Kais Fourati, Ayman Trigui, Salah Boujelben

General Surgery Department, Habib Bourguiba Hospital, University of Sfax, 0,5 El Ain Street, Sfax, 3029, Tunisia

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ABSTRACT

Emphysematous cystitis (EC) is a severe infection of the bladder that usually affects older women with diabetes mellitus. In rare situations, EC can lead to serious complications such as necrotizing fasciitis.

We report the case of a 77-year-old woman who developed emphysematous cystitis complicated with rapidly progressive, gas-producing, necrotizing inflammation located on the right lower abdominal wall. The patient had aggressive drainage and debridement in order to control the extensive necrosis. To our knowledge, this is the third case report that describes a possible association between EC and NF requiring aggressive surgical intervention for both diseases.

1. Introduction

Emphysematous cystitis (EC) is a rare disease characterized by a urinary tract infection associated with intramural and/or intraluminal bladder gas. It typically presents with abdominal pain with severe sepsis (80% of patients), pneumaturia (70%), dysuria (50%), and urinary urgency (50%). $^{1-3}$ In rare situations, EC can lead to serious complications such as necrotizing fasciitis (NF). The combination of EC and NF is extremely rare and reported only in two cases reports. 4,5

2. Presentation

A 77-year-old obese woman was admitted to our emergency department with septic shock. Her medical history was notable for hypertension, type 2 diabetes and a previous scare of a right trochanteric fracture operated on one month before.

Upon admission, the patient's temperature was 39 °C, blood pressure was 92/48 mm Hg, heart rate was 125 beats/min, respiratory rate was 24 breaths/min, and oxygen saturation of 91% on room air body. Physical examination disclosed necrosis of the right anterior abdominal wall extended to the genital region and painful palpation in the right iliac fossa with no signs of peritoneal irritation (Fig. 1). There were no clinical signs of surgical wound infection.

Biological tests showed an elevated WBC at 41000/mm3 and positive C-reactive protein at 250 mg/dl. An acute renal failure was noticed in

creatinine level at 200 mmol/L. After initial resuscitation, an emergent abdominal and pelvic CT associated with Cystography was performed with the aim of excluding an intra-abdominal collection or surgical-site infection after hip fracture surgery.

Radiology showed an air-fluid level inside the urinary bladder and extensive gas within the bladder wall and lumen, consistent with emphysematous cystitis. We, as well, found subcutaneous edema extending from the right groin along the right lower abdominal wall with a large amount of subcutaneous gas (Fig. 2). Emphysematous cystitis complicated with necrotizing fasciitis was suspected which requires immediate surgical treatment.

The intraoperative findings reveal extensive necrosis of the right lower abdominal wall tracking into the pubis and right flank. All necrotic tissue was excised (Fig. 3). A permanent urinary catheter was placed for bladder drainage and decompression.

Urine and blood culture revealed Klebsiella pneumonia. The patient has received empiric intravenous antibiotic therapy. Additional surgical debridement had been performed in the following days. A refractory septic shock was installed and yet the patient died in the 5 PO day.

3. Discussion

Emphysematous cystitis (EC) is considered a severe infection of the bladder that usually affects older women with diabetes mellitus. It is defined as the presence of air at the level of the bladder wall and

Abbreviations: list: EC emphysematous cystitis, NF necrotizing fasciitis.

^{*} Corresponding author. General Surgery Department, Habib Bourguiba Hospital, University of Sfax, Route ElAin 0.5km, Sfax, 3021, Tunisia. E-mail address: drbouzidahmed89@gmail.com (A. Bouzid).



Fig. 1. Clinical pictures of the patient before surgery.

sometimes in the lumen. It is typically presented with abdominal pain with severe sepsis (80% of patients), pneumaturia (70%), dysuria (50%), and urinary urgency (50%). The urinary bladder wall perforation, resulting from EC, is an extremely rare and life-threatening condition. When the infection spreads in the Retzius and Bogros space, it can lead to necrotizing fasciitis of the lower abdominal wall.

However, Necrotizing Fasciitis (NF) are rapidly progressive lifethreatening bacterial infection In addition, NF is usually caused by a synergistic infection involving both aerobes and anaerobes and often produces gas gangrene.³

Although it was a rare case, our patient had significant risk factors for developing both EC and NF including a history of right trochanteric fracture, morbid obesity, and poorly controlled diabetes.

The clinical signs of both Emphysematous cystitis and Necrotizing Fasciitis are highly variable and nonspecific, ranging from asymptomatic to severe sepsis. ^{1,2} Early diagnosis based on clinical presentation and imaging modalities is essential in initiating medical and surgical treatment. In the absence of rapid control of the spread of the infection, necrotizing fasciitis progresses to disseminated vascular coagulation, septic shock, and then death.

This is the third case report, to our knowledge, that describes a possible association between EC and NF. 4,5

The overall mortality of necrotizing fasciitis and perforated emphysematous cystitis is 30%–40% and 20%.^{2,3} respectively. The association of these two entities can be devastating.

In our case, the initial presentation can have a misleading impression because of the fact that the skin above the involved area may exhibit a normal appearance and patients may show relatively mild local signs, despite extensive subcutaneous destruction and systemic dysfunction. In such situations, imaging exams are mandatory to establish the diagnosis.

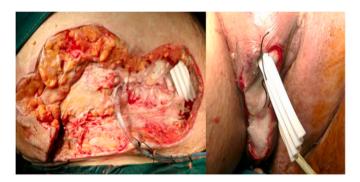


Fig. 3. Clinical pictures of the patient after surgical debridement.

Antibiotic administration and bladder drainage are the best medical therapy for uncomplicated EC. 1,2 Yet, when EC is associated with NF, early surgical resection and debridement of all necrotic tissue is necessary. 4,5

4. Conclusion

To conclude, it is imperative to maintain a high index of suspicion for perforated emphysematous cystitis in patients who present with necrotizing infections of the abdominal wall, flank, back, or groin.

Early diagz nosis and timely surgical treatment are mandatory for treating patients with perforated complicated emphysematous cystitis associated with necrotizing fasciitis.

Authors contributions

SF and AB: conception and design of the work, drafting of the work, AB and HR: acquisition of data, drafting of the work, KF and AT and SB: conception of the work, drafting of the work, All authors had access to the data and have approved the final article.

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Declaration of competing interest

There are no competing interests to disclose.

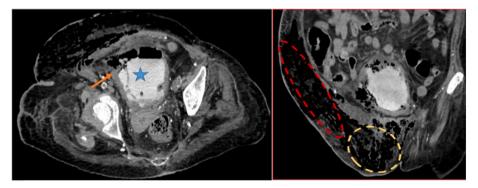


Fig. 2. CT with cystography findings: urinary bladder (blue star) associated with extensive air within its wall (orange arrow). The red dashed line delimited area shows the subcutaneous gas in the anterior abdominal and the yellow one shows the genital area. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

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