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Case report

Emphysematous cystitis complicated by pelviperitonitis: A rare form of urinary tract infection. A case report

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ABSTRACT

Introduction: Emphysematous cystitis is a rare and serious urinary tract infection characterized by the presence of gas within the bladder wall. Factors that increase the risk of developing this condition include advanced age particularly in women, diabetes mellitus, neurogenic bladder, the use of urethral catheters, vesicorectal fistula, and bladder outlet obstruction.

Case presentation: We present the case of a 32-year-old woma, with a two-month history of retroviral infection, admitted for dysuria associated with hypogastric pain and fever, progressing over the past week. The urgent uro-CT scan led to the diagnosis of emphysematous cystitis, requiring a multidisciplinary approach for management. Discussion: Emphysematous cystitis (EC) arises from infections caused by gas-producing bacteria. Complications associated with EC may involve bladder rupture, peritonitis, and/or emphysematous pyelonephritis, hence the importance of early diagnosis and adequate management.

Conclusion: Our case enriches the literature on a rare pathology and underlines the importance of accurate and rapid diagnosis to avoid complications that can threaten renal and vital prognosis.

1. Introduction

Emphysematous cystitis (EC) arises from infections caused by gasproducing bacteria [1]. Risk factors for EC include female gender, age over 60 years, diabetes mellitus (DM), and urinary stasis. Complications associated with EC may involve bladder rupture, peritonitis, and/or emphysematous pyelonephritis.

Emphysematous cystitis most commonly caused by *Escherichia coli* and the computerized tomography (CT) scan is the gold standard in diagnosis emphysematous cystitis. However, X-ray finding with clinical presentation should provoke the suspicious of this diagnosis [2].

Timely diagnosis and proper treatment are essential for managing this distinctive and potentially life-threatening urinary tract condition [3]. Management varies based on severity, ranging from bladder drainage and antibiotic therapy to cystectomy or other surgical interventions.

We report our experience with a 32-year-old woman with a history of immunodeficiency, currently untreated, who presented with a urinary tract infection characterized by dysuria, hypogastric pain, and fever.

This work has been reported in line with the SCARE criteria and cite the following paper [4].

2. Case presentation

This concerns a 32-year-old Ivorian girl living in Southern Morocco known to have a retroviral infection who refused treatment for the past two months. She was admitted to the emergency department for dysuria associated with hypogastric pain in the context of fever and deterioration in her overall well-being.

Upon admission, the clinical examination identified a conscious patient presenting with hypotension at 90/50 mmHg, tachycardia at 115 beats/min, fever at 39,2 $^{\circ}\text{C}$ and hypogastric pain in abdominal palpation. Respiratory status remained stable, diuresis was preserved with pyuria, and there was evidence of deterioration in her overall well-being with asthenia, weight loss and anorxia.

Biologically, we noted a infectious syndrome characterized by leukocytosis with a white blood cell count of 21,000/mm³ [4000-10,000/mm³], a C-reactive protein (CRP) level of 230 mg/L [0-6 mg/L],

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and a markedly elevated procalcitonin level of 33.2 μ g/L [0-0.5 μ g/L], along with normochromic normocytic anemia with a hemoglobin level of 8.6 g/dL [12-16 g/dL]. The renal assessment revealed severe kidney injury with a serum creatinine level of 14.5 mg/dL [0,6–1,2 mg/dL], a plasma urea level of 2.4 g/L [0,15–0,45 g/L], with identification of *Escherichia coli* on urine cytobacteriological examination.

The abdominal ultrasound performed in the emergency department revealed a bladder with thickened wall and heterogeneous echogenic content, showing air bubbles, along with moderate bilateral ureterohydronephrosis.

The Contrast-injected axial (Fig. 1A) and sagittal (Fig. 1B) scans through the pelvis, showing a thickened, irregular bladder wall, with anterior parietal air bubbles, and turbid, suppurative endoluminal contents. In axial section at tubular nephrography time and coronal section at 10-minute late time, he revealed a defect in renal parenchymal enhancement, with eIntoduction vidence of ureterohydronephrosis in relation to the underlying infection, and absence of contrast medium elimination at late time, complicated by multiple pelvic suppurative collections with onset of pelviperitonitis (Fig. 2).

Three days after admission, the patient underwent surgery with a surgical scrubbing in the operating room and a bilateral nephrostomy tube placement, which yielded 1000 cc of cloudy urine with an important amount of gas. Intraoperative cultures revealed the presence of *Escherichia coli*, prompting the initiation of dual antibiotic germsensitive (Ceftriaxone, ciprofloxacin). Due to acute kidney injury, the patient required renal replacement therapy and underwent six hemodialysis sessions following the insertion of a simple femoral catheter.

The patient showed consciousness clinical improvement, and inflammatory parameters got normalized, although the CRP took longer to return to normal levels.

The patient took a 14 days treatment as recommended and was referred after to an infectiologist for a better following up of her retroviral infection.

3. Discussion

Urinary tract infections are linked to gas formation; emphysematous infections can involve the upper urinary tract, including the renal pelvis (pyelitis), the kidneys (pyelonephritis), and the urinary bladder (cystitis) [5].

Emphysematous cystitis is a relatively rare condition caused by gasproducing microorganisms, with only 135 cases reported in the Englishlanguage literature prior to 2006. A literature review, which searched PubMed and Ichushi-Web (The Japan Medical Abstracts Society) for case reports of emphysematous cystitis published in either English or Japanese between 2007 and 2013, identified a total of 102 reported cases [6].

Risk factors for EC include female gender, age over 60 years, diabetes mellitus (DM), and urinary stasis, immune-suppressed conditions such as retroviral infection as described in our case and in a case reported by Yoon Chin Yap from a 67-year-old gentleman with underlying diabetes mellitus, hypertension and retroviral disease on treatment presented with abdominal discomfort for a week, generalized colicky in nature with a pain score of two. He had not passed motion for past 4 days, however he was still able to pass flatus. This was associated with loss of appetite and loss of weight. In view of the co-morbidities, risk factor and radiological examinations carried out, he was treated as emphysematous cystitis and responded well with the standard treatment. High index of suspicious and prompt management is crucial in emphysematous cystitis to help prevent severe complications [2].

Emphysematous cystitis is characterized by the presence of gas in the bladder lumen and, more significantly, within the bladder wall. Regardless of whether classified as type 1 or type 2, poorly controlled diabetes mellitus remains the most common predisposing factor, found in over 70 % of cases, particularly in women. A systematic review of 135 cases of emphysematous cystitis revealed that 64 % of the patients were female, with 67 % of them having diabetes. *Escherichia coli (E. coli)* was the most frequently isolated pathogen in this analysis [7]. Our case is perfectly consistent with the data in the literature. Female gender, immunodepression and causative agent are the main points raised in our case and described in the various series reported in the literature.

The fermentation of concentrated glucose in the urine and surrounding urinary tissues results in the buildup of acids, leading to a decrease in local pH [8]. Gas-producing bacteria then convert these acids into carbon dioxide by secreting specific enzymes. In non-diabetic individuals, urinary lactate has been proposed as a potential substrate for gas production. In this instance, the patient has diabetes, and the urine culture revealed the presence of *Pseudomonas aeruginosa*. The most commonly reported pathogens in a retrospective analysis were *Escherichia coli* and *Klebsiella pneumoniae*, representing 65 % of the isolates.

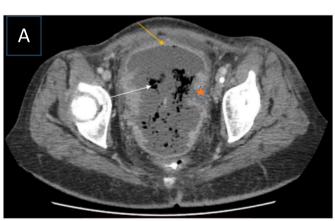
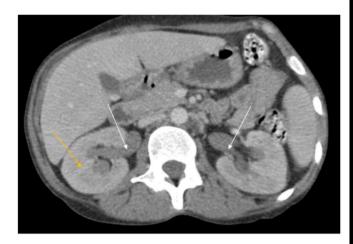




Fig. 1. Contrast-injected axial (A) and sagittal (B) scans through the pelvis, showing a thickened, irregular bladder wall (orange star), with anterior parietal air bubbles more clearly (orange arrow), and turbid, suppurative endoluminal contents (white arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)



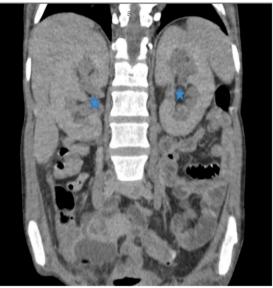


Fig. 2. Scannographic sections after injection of iodinated contrast, in axial section at tubular nephrography time and coronal section at 10-minute late time, showing a defect in renal parenchymal enhancement (orange arrow), evidence of ureterohydronephrosis in relation to the underlying infection (white arrows), and absence of contrast medium elimination at late time (blue stars). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Other organisms identified as potential causes include Proteus, Enterococcus, Pseudomonas, and Clostridium [9]. The microbiology results are essential to identify the germ and adapt antibiotic treatment. Our case describes an EC secondary to an *Escherichia coli* retained on bacteriological samples realised during surgery.

The clinical features are not specific to emphysematous cystitis. When symptomatic, emphysematous cystitis causes abdominal pain in 80 % of cases, it may be associated with purulent urine and systemic signs such as fever and altered general condition. Pneumaturia, whose exact mechanism is not entirely clear, is a very specific sign especially found during bladder catheterization, it is present in approximately 70 % of cases and may be, more or less, associated with lower urinary tract signs [10]. Our patient had a clinical presentation very suggestive of EC. She was admitted to the emergency department for dysuria associated with hypogastric pain in the context of fever.

Imaging examinations play a vital role in making an accurate diagnosis. Zouheir Ibrahim Bitar [11] reported the case of an 83-year-old man with insulin-dependent diabetes mellitus had a medical history significant for myocardial infarction complicated by severe cardiomy-opathy. He also had a prior episode of acute calculous cholecystitis, which was managed with percutaneous drainage and responded well to antibiotic therapy. He later presented with a new episode of fever, suprapubic tenderness, and urinary retention, which was identified using point-of-care ultrasound (POCUS). Additionally, echogenic gas was observed within the bladder wall, along with shadowing artifacts. Ultrasound revealed diffuse thickening of the bladder wall and increased echogenicity. A diagnosis of emphysematous cystitis was suspected, and a urinary catheter was inserted. Computed tomography (CT) of the kidneys, ureters, and bladder confirmed diffuse bladder wall thickening with intramural air.

Effective management of emphysematous cystitis relies primarily on prompt identification of the condition, the initiation of intravenous antibiotic therapy, and maintaining adequate glycemic control. Supportive measures such as bladder catheterization and irrigation especially in the presence of hematuria with clot formation may also be necessary. In approximately 10 % of cases, when medical treatment fails to halt the progression of the infection, surgical options such as partial or total cystectomy or bladder debridement may be required. In this particular case, the patient showed a favorable clinical response to

intravenous antibiotics and the placement of a Foley catheter, without the need for surgical intervention [12].

Prognosis may differ, with early detection generally resulting in better clinical outcomes [12]. Timely identification of emphysematous cystitis is essential. A delayed diagnosis can lead to severe complications such as septic shock, bladder necrosis and rupture, infection spreading to the ureters and renal tissue, and an increased risk of death. The reported mortality rate for patients with emphysematous cystitis is approximately 7 %. The delay in initiating appropriate care for our patient was primarily due to late presentation to the emergency department. This postponement contributed to the progression of the condition into severe sepsis secondary to pelviperitonitis, ultimately requiring urgent surgical intervention.

Our case highlights the importance of early diagnosis of this serious pathology to ensure adequate management.

4. Conclusion

Emphysematous Cystitis (EC) is a rare form of Urinary Tract Infection characterized by gas formation on the bladder walls. The presentation includes: asymptomatic forms, classic cystitis up to sepsis. EC is essential to diagnose and recognize, since it can quickly progress to lifethreatening conditions. This report that EC can present in a typical fashion, and that treatment should account for patient-specific factors, even if it deviates from the norm. Early diagnosis and aggressive medical and surgical management of gas-forming organisms are vital.

Author contribution

Bouchoual Mohammed: Writing - review & editing.

El Khand Ali: Investigation.

El Ouazzani Mouad: Visualization.

Anibar Sara: Visualization.

Jabrane Marouane: supervision. Arrayhani Mohamed: validation.

Patient consent statement

Written informed consent was obtained from the patient for

publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Parental consent: Was not necessary as our patient is an adult and not a minor.

Ethical approval

I'd like to inform you that in our establishment, we don't ask for ethical approval because we don't have an ethics committee and it's just a case report for which we've only asked for the patient's and family's agreement.

Institution: Research and Innovation in Nephrology and Autoimmune diseases Laboratory (RINAD). REGNE Research Laboratory. Faculty of Medicine and Pharmacy. Ibn-Zohr University. Agadir-Morocco.

Guarantor

Pr Bouchoual Mohammed.

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Conflict of interest statement

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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