

Severe hemorrhage in patient carrier of diabetes mellitus type 2 with emphysematous cystitis: a case report and literature review

Hemorragia grave em doente portador de diabetes mellitus tipo 2 com cistite enfisematosa: um relatório de caso e revisão de literatura

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ABSTRACT

Background: Emphysematous cystitis is a disease of the bladder with retention of gas in the bladder wall or lumen. The risk factors include urinary tract infection by gas producing anaerobic bacteria and fungi, diabetes mellitus and compromised immunity. Primary risk factor being diabetes mellitus, emphysematous cystitis can occur with unusual presentations like hypervolemic shock. Imaging technologies are widely used to diagnose and detect emphysematous cystitis. However, better diagnosis can be achieved by computed tomography (CT) of the abdomen and pelvis. Early and accurate diagnosis of emphysematous cystitis in diabetic patients can result in better prognosis of the disease.

Case presentation: Here, we report for the first time a case of emphysematous cystitis with hypovolemic shock with type 2 diabetes mellitus, and present a review of literature on the topic. The diabetic patients with emphysematous cystitis should be cautiously monitored for hypervolemic shock along with the routine management of the disease.

Conclusions: Such patients should be kept on constant monitoring for hypervolemic shock along with conventional diagnosis and management of the disease.

Keywords: emphysematous cystitis, diabetes, hypervolemic shock.

RESUMO

Antecedentes: A cistite enfisematosa é uma doença da bexiga com retenção de gás na parede da bexiga ou no lúmen. Os factores de risco incluem infecção do tracto urinário por bactérias e fungos anaeróbios produtores de gás, diabetes mellitus e imunidade comprometida. Sendo a diabetes mellitus o principal factor de risco, a cistite enfisematosa pode ocorrer com apresentações pouco sensuais como o choque hiper-volémico. As tecnologias de imagem são amplamente utilizadas para diagnosticar e detectar a cistite enfisematosa. No entanto, um melhor diagnóstico pode ser conseguido por tomografia computadorizada (TC) do abdómen e da pélvis. Um diagnóstico precoce e preciso da cistite enfisematosa em doentes diabéticos pode resultar num melhor prognóstico da doença.

Apresentação do caso: Aqui, relatamos pela primeira vez um caso de cistite enfisematosa com choque hipovolémico com diabetes mellitus tipo 2, e apresentamos uma revisão da literatura sobre o tema. Os doentes diabéticos com cistite enfisematosa devem ser cautelosamente monitorizados para o choque hipovolémico juntamente com a gestão de rotina da doença. **Conclusões:** Tais pacientes devem ser mantidos em constante monitorização para o choque hiper-volémico juntamente com o diagnóstico convencional e a gestão da doença.

Palavras-chave: cistite enfisematosa, diabetes, choque hiper-volémico.

1 BACKGROUND

Emphysematous cystitis is a complicated urinary tract disease characterized by the presence of air in the wall and bladder lumen [1]. Emphysematous cystitis is most commonly observed in middle-aged diabetic women (50 years). The diagnosis is made on the basis of the radiological imaging of the abdomen. The patients present a varied range of symptoms ranging from asymptomatic urinary tract infection to urosepsis and septic shock [2]. Here, we present a case of emphysematous cystitis with hypovolemic shock with type 2 diabetes mellitus.

2 CASE PRESENTATION

A 56 year old, white, female, was admitted to the Emergency Department of the General Hospital of Roraima (HGR) with complaints of suprapubic pain, fever, and open hematuria. She presented with type 2 diabetes mellitus and was on treatment with metformin and reported recurrent urinary tract infection in the last 2 months.

3 INVESTIGATIONS

On physical examination the patient was found to be in regular general condition, dehydrated +++ / 4+, bleached +++ / 4+, blood pressure-90x60 mmHg, heart rate-110 bpm, respiratory rate-22 sibling , temperature-38°C, glasgow coma scale-13 (AO: 3, RV: 4, RM: 6), suprapubic palpation with presence of bexigoma and blood on urethral ostium external. She was admitted and recieved a semi-intensive therapy regimen for immediate clinical support. Laboratory tests revealed: Hb-5.5, Ht-16%, Leuc-17,700, B-10%, Ins-76%, U-113, C-4.3. Arterial blood gas analysis showed metabolic acidosis and Urine I test revealed hematuria. ultrasonography examination showed intravesical clot and mild hydronephrosis bilaterally.

Computed tomography (CT) of the abdomen and pelvis was used for a better evaluation of the clinical case. CT demonstrated presence of gas in the bladder wall, in the proximal ureter and upper pole of the left kidney (Fig. 1, Fig. 2 and Fig. 3) features consistent with pyelo-cystitis. The patient was admitted for endoscopic intravesical clot evacuation and cystoscopy under anesthetic block. Universal enantema was found upon bladder examination.

4 TREATMENT

The patient was maintained in a semi-intensive therapy regimen with hydration support, glycemic control (intravenous insulin), blood pressure control, antibiotic therapy (Rocefin 2g/day) and open bladder drainage. On the seventh day of treatment, laboratory tests (Hb-10, Ht-34%, Leuc-5,300, S-69%, Eos-4%, Linf-20 %, C-1.5, U-29, Glic-98, and K: 4.3) as well as clinical manifestations of sepsis showed significant improvements. CT did not show gas formation in the excretory and bladder routes and the patient was discharged on the 12th day of hospital admission.

5 OUTCOME AND FOLLOW UP

The patient was followed up on an outpatient basis. After a urodynamic study, the patient showed to have a neurogenic bladder with a clean intermittent catheterization and has no recurrence of urinary tract infection.

6 DISCUSSION AND CONCLUSION

Emphysematous cystitis is a rare infectious disease of the bladder wall caused by the infections of gas forming bacteria and fungi [1]. This disease is typically defined and

diagnosed radiologically and by CT [3]. The presence of air in the urinary tract was for the first time reported in 1671, in a patient with pneumaturia. Bailey reported 19 cases of emphysematous cystitis with intraluminal and interstitial collections of gas [4]. Clinical presentation of emphysematous cystitis is varied, patients may be asymptomatic or may present dysuria pneumaturia or sepsis.

The bacterial infectious agents most prevalent in this type of infection is *E. Coli*. Other infectious microbes that cause emphysematous cystitis are *Klebsiella pneumoniae*, *Pseudomonas aeruginosa*, *Proteus mirabilis*, *Candida albicans*, and *Candida tropicalis*, *Aspergillus fumigatus*, *Staphylococcus aureus*, Group D *Streptococcus*, *Enterococcus faecalis*, *Enterobacter aerogenes*, and *Clostridium perfringens* and *Cl. welchii*. This disease mostly affects the diabetic and immunocompromised patients [5]. Several studies have reported emphysematous cystitis in aged patients with uncontrolled diabetes mellitus [6-10]. A literature review reported diabetes in >50% of emphysematous urinary tract infections. Females at advanced ages are more frequently affected [11]. The ratio of affected females to males is reported to be 2:1 [12]. Recently in a report this ratio of affected females to male was reported to be 2.2:1 [13]. There are reports about the occurrence of emphysematous cystitis without associated diabetes mellitus in males [14].

High tissue concentration of glucose in the diabetic patients and poor tissue perfusion favor the development of emphysematous infection by the anaerobic microbes in the urinary tract in diabetics. This multifactorial theory is the most accepted theory to explain the pathophysiology of this entity in the diabetic patients but this theory does not explain the cause of emphysematous cystitis in a significant number of non diabetic patients. In such cases the urinary anaerobic microbes utilize the urinary albumin as substrates and produce gas. Another theory put forth to explain the cause of emphysematous infection states that the compromised immunity of the host and improper or inefficient intracellular metabolism results in gas production within these tissues [15].

Others factors like neurogenic bladder, anatomical barrier in the urinary outlet obstruction, urinary tract infections, catheters, and immune-deficiency also predispose the patients to emphysematous cystitis [16].

The management of emphysematous cystitis depends on the severity of the disease. Use of broad-spectrum antibiotics usually shows good prognosis [16,17] where the cause is microbial infection. In a review comprising of 135 published cases by Thomas et al [17], it was reported that 10% of patients required combined medical and surgical intervention with an overall mortality of 7%.

Besides the use of antibiotics, bladder drainage, glycemic control and surgical intervention are the most used management strategies to treat emphysematous cystitis. In the diabetic emphysematous cystitis the best way of disease management is glycemic control.

Preventive measures for emphysematous cystitis has not been clearly explained till date. However, spreading awareness and educating the patients and the care givers about the consequences and potential to cause threat to life can improve the outcomes [18].

The use of CT defines the severity and extent of the disease as well as allows the technical management for future intervention. A delay in diagnosis may cause bladder rupture, septicemia, peritonitis and death [3].

Emphysematous cystitis should be suspected in diabetic patients who do not respond to conventional therapy of complicated urinary tract infection. In patients with few comorbidities and with favorable response, conservative treatment is encouraging initially. However, necrotizing infection requires more aggressive treatment that includes surgery. In the present study the patient was managed with hydration support, glycemic control, blood pressure control, antibiotic therapy and open bladder drainage. To the best of our knowledge, the present case of emphysematous cystitis with hypovolemic shock with type 2 diabetes mellitus is the first reported case ever. Since the diabetic patients with emphysematous cystitis can present with hypervolemic shock, such patients should be kept on close monitoring for hypervolemic shock along with the conventional diagnosis and management of the disease.

DECLARATIONS

Ethics approval and consent to participate: The consent was obtained directly from the patients.

Consent for publication

Not applicable

Availability of data and material

Not applicable

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

MML Jr was the major contributor in writing the manuscript, and performed the surgery. TQAS analyzed and interpreted the patient data. All authors read and approved the final manuscript.

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FIGURE LEGEND

Fig 1. CT showing air (gas) in the anterior bladder wall

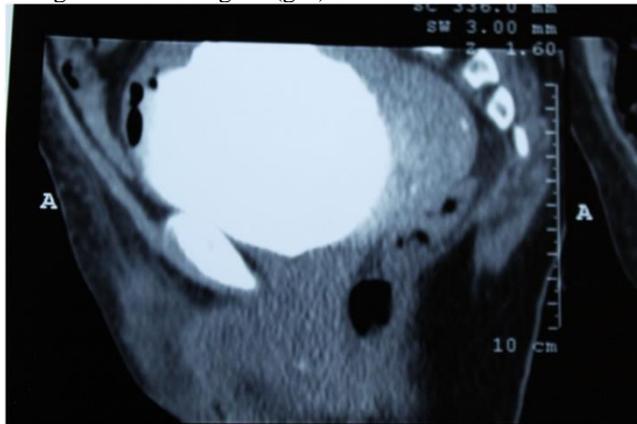


Fig 2. CT showing air (gas) in the left ureter



Fig 3. CT showing air (gas) in the left excretory pathway

