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CASE REPORT

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Pneumomediastinum as a rare complication in an immunosuppressed patient with emphysematous cystitis

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Abstract

The extravesical spread of gas into the extraperitoneal space is an unusual complication of emphysematous cystitis and rarely, can reach remote areas of the body. Herein, we present the case of an immunosuppressed woman with emphysematous cystitis and extensive spread of extraperitoneal free gas up to the mediastinum.

KEYWORDS

bone marrow transplantation, emphysematous cystitis, immunosuppression: case report, pneumomediastinum

1 | INTRODUCTION

Emphysematous cystitis is a rare subtype of bladder infection caused by gas-forming organisms and characterized by the presence of air bubbles inside the bladder wall. The incidence of the disease is higher in the female population. Risk factors include uncontrolled diabetes, immunosuppression, and chronic bladder obstruction.[1] *Escherichia coli* appears to be the most common pathogen isolated in the urine of these patients, followed by *Klebsiella pneumoniae*.[2] Emphysematous cystitis is a medical emergency, which may negatively impact patients' health without immediate intervention. Disruption of the bladder wall and gas spread into intraperitoneal or extraperitoneal space is an infrequent complication of emphysematous cystitis with little evidence in the literature. Diagnosis and management of patients presenting with gas spread to the abdomen are often challenging for physicians, especially in severely immunosuppressed patients. To the best of our knowledge, there are no reports of the presence of free gas within the mediastinum as a result of this rare lower urinary tract infection. We present a case of emphysematous cystitis in a severely immunosuppressed patient with an extensive spread of extraperitoneal free gas within the mediastinum and who was treated conservatively with bladder catheterization and intravenous antibiotics.

2 | CASE REPORT

A 55-year-old woman with a history of bone marrow transplantation due to the blast phase of chronic myelogenous

leukemia (CML) 20 months earlier, presented to the hematology department with complaints of abdominal pain and dysuria. She was hemodynamically stable with no fever. Physical examination revealed a mildly painful suprapubic abdominal mass.

The patient was severely immunosuppressed and received long-term corticosteroid treatment for chronic graft-versus-host disease. Complete blood count and chemistry panel revealed a total white cell count of 1.150 $10^3/\mu l$ with neutropenia, red cell 3.14 × 10 /μl, hemoglobin 9.0 g/dl, platelet 38 × $10^3/\mu l$, serum creatinine 0.21 mg/dl, and serum glucose 92 mg/dl. The microscopic examination of urine sediment revealed 10–12/hpf WBC and 2–4/hpf RBC with several microorganisms. A plain abdominal X-ray (Figure 1A) showed a circular line of increased radiolucency in the bladder area (a typical image of emphysematous cystitis).

An indwelling urinary catheter was placed, and 900 ml of urine was drained with air bubbles in the urine bag. Finally, abdominal computed tomography (CT) scan examination revealed the emphysematous appearance of bladder wall, along with the presence of extraperitoneal free gas around the bladder, in the perineum, retroperitoneal in the subdiaphragmatic space, and the mediastinum

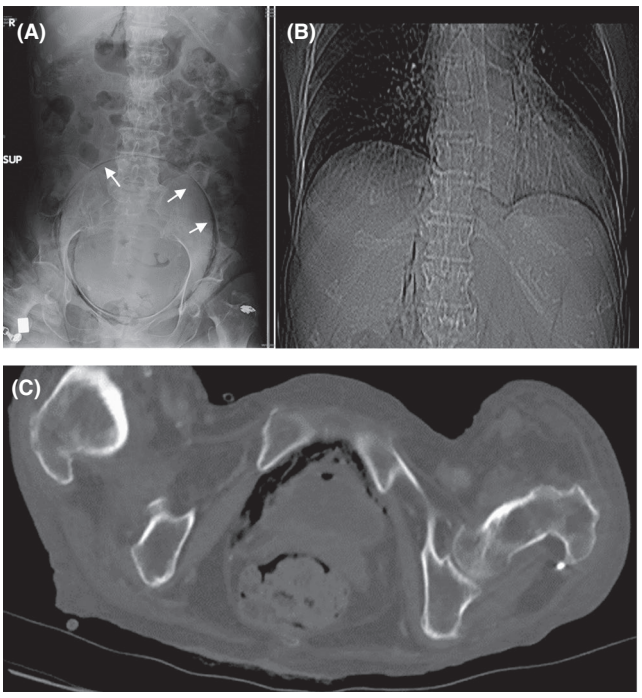

FIGURE 1 (A) Plain abdominal X-ray which depicts a circular line of increased radiolucency (white arrows) in the suprapubic area. (B) Image from CT topogram scan showing the subdiaphragmatic linear presence of gas in the possible route of inferior vena cava reaching to the mediastinum. (C) Coronal CT imaging of the pelvis demonstrating the presence of intramural and intraluminal gas bubbles in the bladder

(Figures 1B,C and 2A–D). Urine leakage into the perivesical space was not observed. Following the diagnosis of emphysematous cystitis, an empirical intravenous antimicrobial therapy was introduced including, colistin, ceftazidime/avibactam, and metronidazole, according to patient's previous positive cultures from infectious episodes. A multidisciplinary team consisting of consultant urologists, and treating physicians, decided that it was the best to proceed with a conservative management for the patient, because of the stable good clinical condition and the high risk of any invasive intervention. Urine culture was positive for Klebsiella pneumoniae, >100,000 colony-forming units (CFU), sensitive to colistin. Antibiotic treatment continued for 14 days, and a subsequent abdominal CT scan showed improvement, with absorption of the largest amount of free gas. The patient was discharged from the hospital after obtaining two consecutive negative urine cultures.

3 | DISCUSSION

Emphysematous cystitis constitutes a rare inflammation of the bladder caused by gas-forming organisms and characterized by the presence of intramural gas in the bladder wall. As in emphysematous infections of the upper urinary tract, the incidence of the disease is higher in the female population and in diabetic patients at a rate of 64% and 67%, respectively. Other risk factors include immunosuppression, recurrent urinary infections, and chronic bladder obstruction.[2]

In clinical examination, abdominal pain, urinary retention, and pneumaturia are the most frequent symptoms, although not specific for the disease.[3] Regarding imaging methods, abdominal CT scan seems to be the most sensitive examination that determines the diagnosis of emphysematous cystitis with the presence of intraluminal and intramural gas.[2,3]

Disruption of the bladder wall and leakage of free gas into the intraperitoneal or extraperitoneal space is a rare complication of emphysematous cystitis. The disruption of bladder serosa without involvement of mucosal layer results in the leakage of free air bubbles to the perivesical space that can extend to different parts of the body.[4] Previous cases of emphysematous cystitis have been reported with the presence of extraperitoneal free gas in the pelvis, Retzius space, and the femoral canal.[4–6]

There are no reports in the literature of gas presence in the retroperitoneal subdiaphragmatic space or in the mediastinum as a complication of emphysematous cystitis. Extraperitoneal free abdominal gas (originating from the abdomen) can spread anywhere in the abdomen and pelvis through the interconnected subperitoneal space. In our case, the potential route of free gas from the

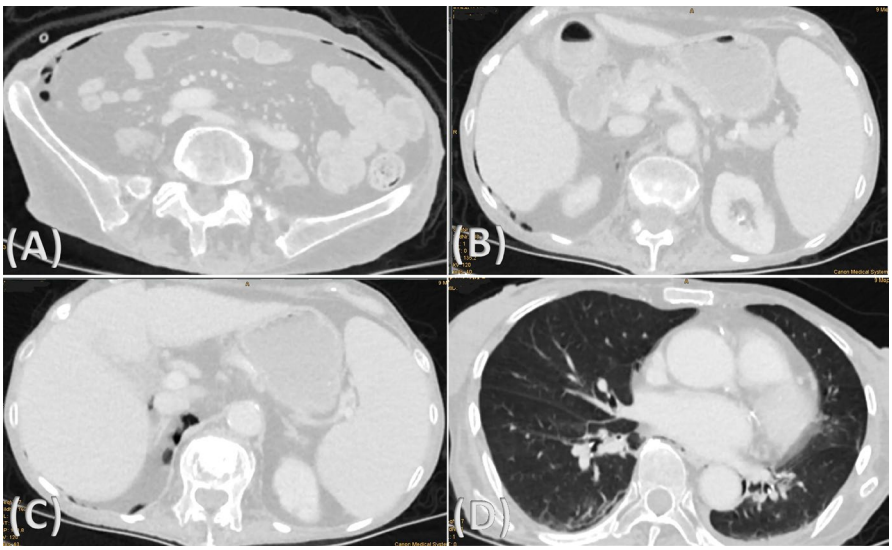

FIGURE 2 CT imaging of the abdomen (lung window) demonstrating the presence of retroperitoneal gas bubbles: (A) prevesical space and right iliac fossa, (B) posterior pararenal space. (C) around inferior vena cava and (D) mediastinum

perivesical space to the mediastinum could be the one described by Pannu and Oliphant.[7] Extraperitoneal free air from perivesical space can spread to prevesical space and then via infrarenal space to the posterior or anterior pararenal space. From the posterior pararenal space, extraperitoneal air can spread superiorly to the diaphragm and mediastinum.

Emphysematous cystitis is considered a severe urinary tract infection with potentially rapid progression to pyelitis or necrosis of the bladder wall, pneumoperitoneum, and urosepsis, especially in immunosuppressed patients.[8] Sadek et al.[5] reported poor outcomes in a patient with emphysematous cystitis and extensive emphysematous changes. Although the treatment is mainly conservative, with urinary drainage and administration of antibiotics, 10%–15% of patients with serious complications require surgical intervention. The overall fatality of emphysematous cystitis ranges from 7% to 9%, but this seems to increase vertically in case of extensive spread of gas to the body and principally to the upper urinary tract.

4 | CONCLUSION

Emphysematous cystitis is rarely complicated by the extended spread of gas to remote parts of the body. In the case of our patient, gas bubbles reached up to the mediastinum. Early diagnosis with immediate urinary drainage and antibiotics administration seem to have played a decisive role in the outcome of this severely immunosuppressed patient.

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CONFLICT OF INTEREST
The authors declare no conflict of interest.

AUTHOR CONTRIBUTIONS
GT performed the study design, collected the data, data analysis, and writing of the manuscript. ZB involved in study design, collected the data, and data analysis. SDB and NK collected the data. AP involved in data analysis. SG performed the review and editing of the manuscript. IS and CK involved in supervision.

ETHICAL APPROVAL
Written informed consent was obtained from the patient to publish this case report and any accompanying images.

CONSENT
Written informed consent was obtained from the patient.

DATA AVAILABILITY STATEMENT
Data sharing is not applicable to this article as no new data were created or analyzed in this study.

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