CASE REPORT

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Bilateral emphysematous pyelonephritis and emphysematous cystitis: a rare case report

Obostrani emfizematozni pijelonefritis i emfizematozni cistitis

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Abstract

Introduction. Emphysematous pyelonephritis (EPN) is a rare type of severe kidney infection characterized by gas formation in the renal parenchyma, collecting system, or surrounding perinephric tissues. Emphysematous cystitis (EC) is a rare, potentially fatal condition characterized by the presence of gas in the bladder wall and lumen. We present a rare case of EPN with EC in a patient with diabetes mellitus (DM). **Case report**. A 38-year-old female patient with poorly regulated DM was referred to the Department of Urology at the General Hospital in Novi Pazar, Serbia, with malaise, fever, and flank and suprapubic pain. After physical examination, laboratory analyses, and radiological diagnostics, the diagnosis of bilateral EPN with EC was established. Throughout the hospitalization, the patient remained hemodynamically stable with preserved diuresis. The patient's urine and blood cultures were negative. The patient was treated with intravenous fluids and antibiotics (metronidazole, vancomycin, and meropenem). No other invasive form of treatment was needed due to successful conservative treatment. **Conclusion.** A timely diagnosis and rapid inclusion of specific antibiotic therapies enables the avoidance of the need for invasive forms of treatment in patients with EPN and EC. The conservative management of the presented patient was successful.

Key words:
cystitis; diabetes mellitus, tip 1; emphysema; pyelonephritis; treatment outcome.

Apstrakt

Uvod. Emfizematozni pijelonefritis (EP) je retka i teška infekcija bubrega koja dovodi do stvaranja gasova u bubrežnom parenhimu, sabirnom sistemu ili perirenalnom tkivu. Emfizematozni cistitis (EC) je retko i potencijalno fatalno stanje, koje karakteriše nakupljanje gasa u zidu i lumenu mokraćne bešike. Prikazan je slučaj EP sa EC kod osobe obolele od dijabetesa melitusa (DM). **Prikaz bolesnika**. Bolesnica stara 38 godina, sa loše regulisanim DM, hospitalizovana je na Odeljenju urologije u Opštoj bolnici Novi Pazar, Srbija, sa tegobama u vidu malaksalosti, povišene telesne temperature, bola u slabinskim regijama i donjem delu stomaka. Nakon kliničkog pregleda, laboratorijskih analiza i radiološke dijagnostike postavljena je dijagnoza obostranog EP sa EC. Sve vreme tokom hospitalizacije bolesnica je bila hemodinamski stabilna, sa očuvanom diurezom. Urinokultura i hemokultura bile su negativne. Bolesnica je lečena intravenskim rastvorima i antibioticima (metronidazol, vankomicin i meropenem). Nije bilo potrebe za drugim invazivnim oblikom lečenja, jer je konzervativna terapija bila uspešna. **Zaključak.** Blagovremenim uspostavljanjem dijagnoze i brzim uključivanjem antibiotske terapije omogućuje se izbegavanje invazivnog oblika lečenja kod bolesnika sa EP i EC. Primenjena konzervativna terapija imala je povoljan ishod kod prikazane bolesnice.

Ključne reči:
cistitis; dijabetes melitus, tip 1; emfizem; pijelonefritis; lečenje, ishod.

Introduction

Emphysematous pyelonephritis (EPN) is a rare type of severe kidney infection characterized by gas formation in the renal parenchyma, collecting system, or surrounding perinephric tissues [1]. Emphysematous cystitis (EC) is a rare, potentially fatal condition characterized by the presence of gas in the bladder wall and lumen [2]. Bilateral EPN combined with EC in the same patient is a very rare case. It is caused by gram-negative microorganisms, most commonly *Esche*

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richia coli (*E. coli*), and other anaerobes such as *Proteus spp* and *Klebsiella* [3]. Kelly and MacCallum [4] reported the first case of gas-forming renal infection in 1898. EPN is commonly associated with poorly controlled diabetes mellitus (DM) and urinary tract obstruction or polycystic kidneys [5, 6]. The gold standard for diagnosis is computed tomography (CT). Treatment of EPN with EC can be conservative, based on appropriate antibiotic therapy, or invasive, including emergency nephrectomy or percutaneous drainage [7].

Huang and Tseng [5] created a classification system based on the CT scan: class 1 – gas in the pyeloureteral system only; class 2 – gas forming in the renal parenchyma without extension to parararenal tissues; class 3 – accumulation of gas to perirenal tissues; class 4 – solitary kidney with EPN or bilateral EPN.

We present a patient with poorly controlled DM who acquired EC and bilateral EPN. Written informed consent for participating in the study was obtained from the patient.

Case report

A 38-year-old female patient with type 1 DM experienced malaise, suprapubic pain, fever, and flank pain for 4 days before admission to the Department of Urology, General Hospital of Novi Pazar, Serbia, in September 2023 (day 0). She had been suffering from DM, and she had been receiving insulin therapy. She did not consume alcohol or cigarettes.

A physical examination on admission revealed the following: blood pressure of 110/80 mmHg; body temperature of 37.5 °C, heart rate of 80 beats *per* minute, Glasgow Coma Scale (GCS) of 15 (the highest possible GCS score of 15 means fully awake, responsive, and without any problems concerning thinking ability or memory), and respiration rate of 17 breaths *per* minute. Bilateral costovertebral angle tenderness was present without palpable masses. The remainder of the physical examination was unremarkable.

At the time of the patient's admission, laboratory analyses showed raised inflammatory markers: C-reactive protein (CRP) 398.7 mg/L [normal range (NR) < 5 mg/L], leukocytes 29.5 × 10^9 /L (NR 4.0–10.0 × 10^9 /L) and neutrophils 92% (NR 34–71%). Laboratory analyses also showed a raised blood glucose of 24.1 mmol/L (NR 2.6–6.1 mmol/L), values of glycated hemoglobin A1c (HbA1c) of 12.1%, serum creatinine 154 µmol/L (NR 53–124 µmol/L), and blood urea 9.6 mmol/L (NR 2.5–8.3 mmol/L). Hypoalbuminemia was detected at 22 g/L (NR 35–55 g/L). Urine analysis confirmed the presence of glycosuria, ketonuria, and pyuria.

An abdominal ultrasound (US) showed evidence of gas on both kidneys (Figure 1) and the bladder wall (Figure 2). An urgent CT scan of the abdomen and pelvis was performed. CT confirmed the presence of extensive gas accumulation in both kidneys (Figure 3), with gas inside and around the bladder wall (Figure 4). According to Huang and Tseng [5], this EPN with EC was categorized as class 4.

The patient was hospitalized and started on empirical antibiotics along with other supportive measures. The patient's blood and urine cultures were negative. On the second day of hospitalization (day 2), due to the complexity of the treatment, we decided to refer the patient for further treatment to the Clinic for Urology at the Clinical Center of Serbia, Belgrade, Serbia. The patient was treated with intravenous (i.v.) fluids, antibiotics (metronidazole, vancomycin, and meropenem), and rapid-acting insulin. In addition, the electrolyte imbalance was treated. No other invasive form of treatment was needed. Urine cultures were repeated several

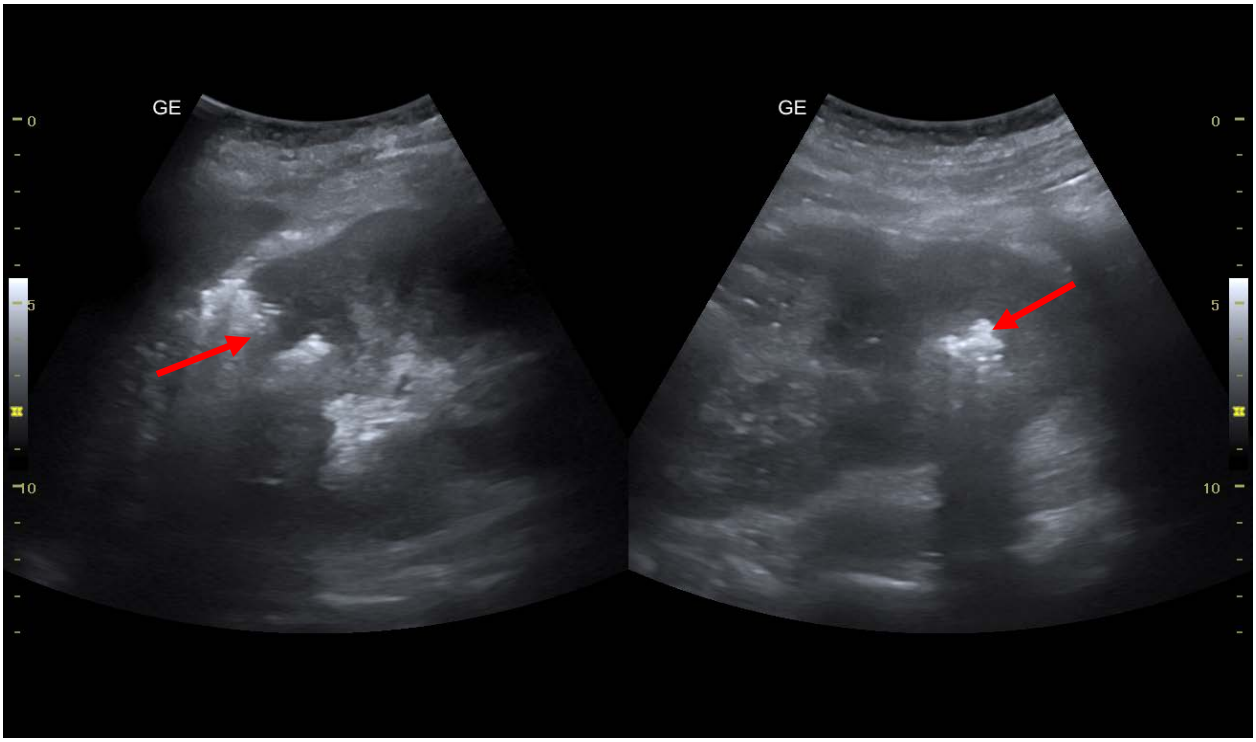

Fig. 1 – Ultrasound of the abdomen showed the presence of gas accumulation in both kidneys (arrows).

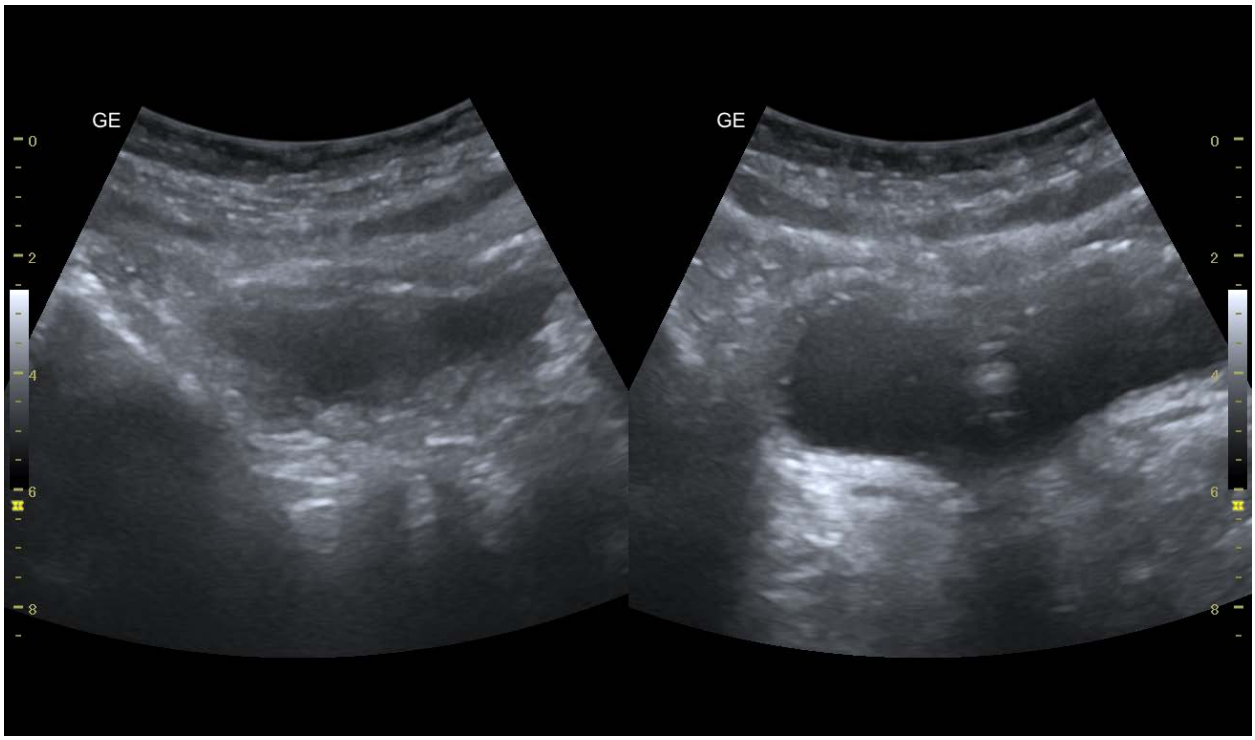

Fig. 2 – Ultrasound of the abdomen showed gas accumulation in the bladder wall (acoustic shadowing around the bladder wall).

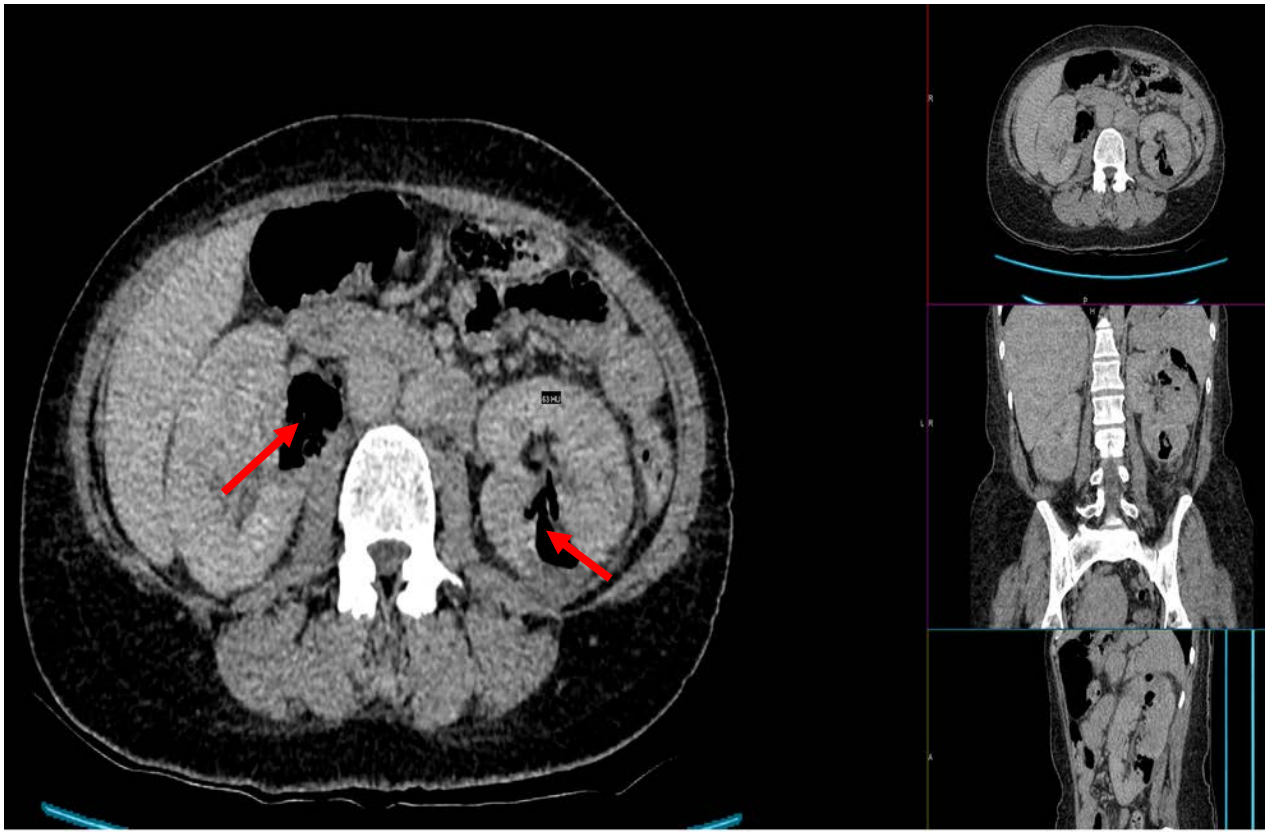

Fig. 3 – Computed tomography showing diffusely enlarged kidneys and gas in the kidneys bilaterally, indicative of bilateral emphysematous pyelonephritis (class 4 emphysematous pyelonephritis) (arrows).

times during hospitalization and were always negative. Likewise, the blood culture was also negative. Throughout the hospitalization, the patient remained hemodynamically stable with preserved diuresis. On day 15 after admission, the patient was afebrile. On day 25 of admission, a second CT scan was performed, which showed a complete resolution of the right renal and bladder emphysema and the presence of a small gas accumulation in the left renal parenchyma. After 28 days of inpatient care, the patient was discharged with oral antibiotics. US of the abdomen 7 days after discharge showed a complete resolution of right renal and bladder emphysema and the presence of a small amount of gas in the upper pole of the left kidney (Figure 5). Laboratory analyses had normalized by discharge (leucocytes 9.3 x 10^9/L, urea 7,1 mmol/L, creatinine 89 μmol/L, and CRP 4 mg/L).

Laboratory analyses performed 20 days after discharge showed increased CRP 13 mg/L and leukocytes 11.2 × 10^9/L (other values were expected for DM patients) (Table 1).

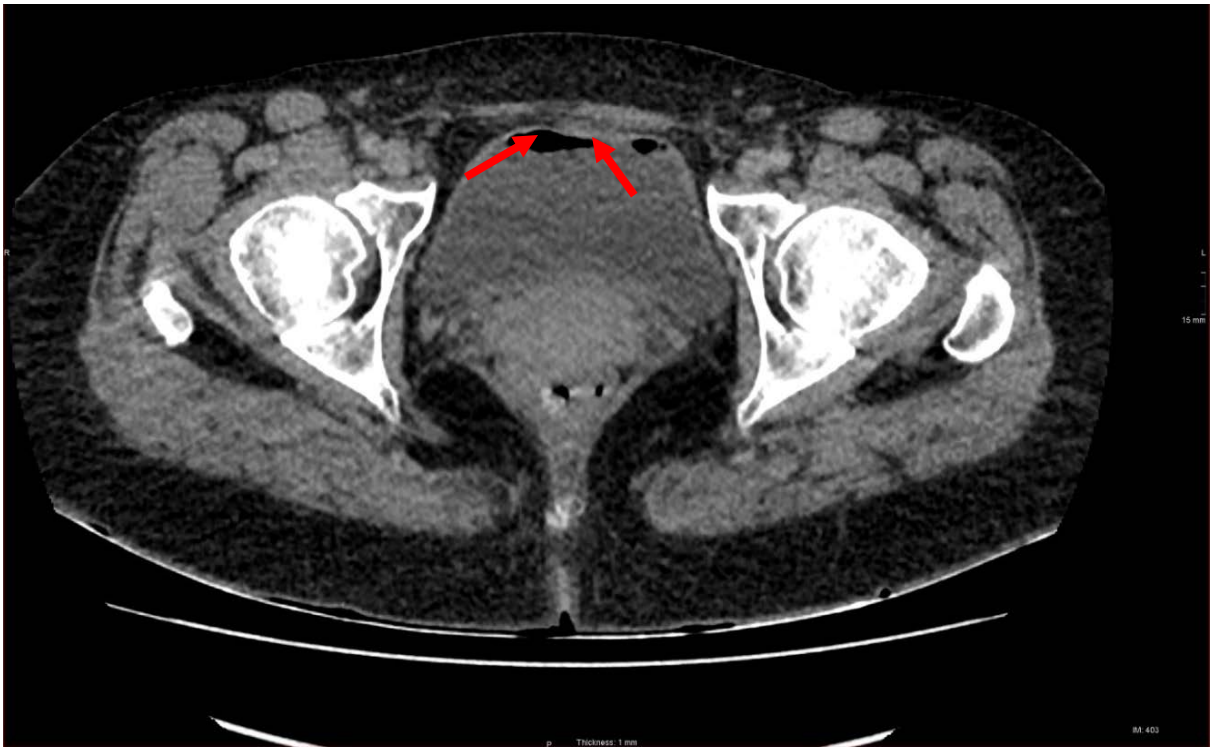

Fig. 4 – Computed tomography image of gas accumulation in the bladder wall (arrows).

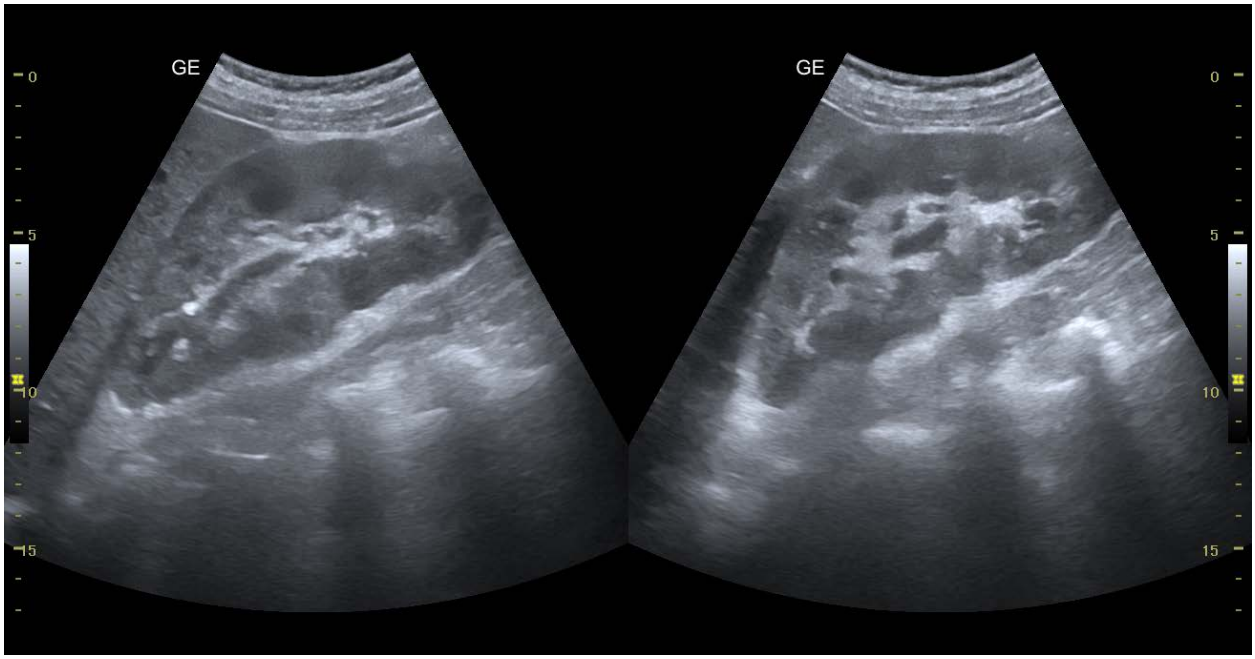

Fig. 5 – Ultrasound imaging – disappearance of gas in the right renal parenchyma four weeks after initiation of treatment, and a small amount of gas in the left kidney.

Table 1

Patient laboratory test results 20 days after discharge from the hospital

Parameters	Values
White blood count, × 10^9 /L	11.2
C-reactive protein, mg /L	13
Serum creatinine, μmol/L	65.5
Serum urea, mmol/L	4.0
HbA1c, %	7.7
Hemoglobin, g/L	117
Serum glucose, mmol/L	11.8
Serum albumin, g/L	43

HbA1c – glycated hemoglobin A1c.

Discussion

Descriptions of patients with bilateral EPN and EC in the literature are scarce. It is believed that EPN is more common in females because they have urinary tract infections more commonly [8]. The most common risk factor for the development of EPN associated with EC is DM. Stapleton [9] reports that a high glucose concentration in the tissue contributes to the growth and proliferation of gas-producing bacteria. Moreover, a high tissue glucose concentration inhibits the function of leukocytes and impairs the response to infection. Our patient had a poorly controlled DM with an HbA1c value of 12.1% on admission. Due to the fermentation of glucose and lactate, gas is produced, which is then accumulated at the site of inflammation. As mentioned above, EPN is caused by gram-negative microorganisms, most commonly *E. coli*, and other anaerobes such as *Proteus spp* and *Klebsiella* [3]. In our case, no definitive causative organism was identified from urine or blood culture. We assume it is because our patient used antibiotics prior to urine collection, which suppresses bacterial growth and leads to false-negative results.

In patients with EPN or EC, the mortality rate is between 8.7% and 21%. This infection is life-threatening [10, 11]. Lu et al. [12] reported that high mortality was present in EPN patients who require emergency hemodialysis, have present shock on initial presentation, have altered mental status, severe hypoalbuminemia, inappropriate antibiotic therapy, or polymicrobial infection. Likewise, inadequate empirical antibiotic and polymicrobial infections were associated with high mortality. Our patient did not require emergency hemodialysis because she did not have altered mental status or shock on the initial presentation. In a series of 19 patients with EPN, Khaira et al. [13] also reported that shock was an independent poor prognostic risk factor.

Over the years, the treatment of EPN with EC has evolved from an invasive to a conservative approach. The major treatment methods are conservative management, percutaneous drainage, or emergency nephrectomy.

Angulo et al. [14] suggested that conservative treatment or percutaneous drainage should be performed in patients with EPN in a solitary kidney, bilateral EPN, localized EPN, or if the patient cannot tolerate general anesthesia. In a study conducted in Taiwan, ten patients with EPN received only antibiotics, and the mortality rate was 20% (two patients died) [12]. Our patient was diagnosed with class 4 disease, leukocytosis, azotemia, and hypoalbuminemia on admission. Aboumarzouk et al. [15] show that patients with EPN and EC have an overall mortality rate of about 18%. Furthermore, this study shows that conservative treatment and percutaneous drainage are associated with significantly higher survival rates than emergency nephrectomy. Emergency nephrectomy should be applied when the patient does not improve despite previous treatment methods. Empirical antibiotics are the first line for the treatment of EPN with EC [13]. Our patient was managed in accordance with the current evidence-based protocols, where she was treated with empirical antibiotics and other supportive therapy. The most successful treatment was a combination of i.v. antibiotics and nephrostomy insertion, with a mortality rate of around 13% [16].

Conclusion

The best diagnostic method for emphysematous pyelonephritis and emphysematous cystitis is computed tomography, which also aids in classifying emphysematous pyelonephritis. The conservative management of our patient was successful. We emphasize that timely diagnosis and specific antibiotic treatment can prevent the need for surgery. The risk of mortality was reduced because our patient did not manifest shock, mental disorder, thrombocytopenia, or polymicrobial infection.

Conflict of interest

The authors declare no conflict of interest.

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