



Inflammation and infection

Complicated case of bilateral emphysematous pyelonephritis and emphysematous cystitis successfully treated with transurethral drainage and ureteral stents

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ABSTRACT

Emphysematous pyelonephritis and emphysematous cystitis are intractable diseases. Eight cases of bilateral emphysematous pyelonephritis and emphysematous cystitis have been reported, but no treatment has been established. An 88-year-old female was admitted with traumatic subarachnoid hemorrhage, and on the fourth day of hospitalization, she developed fever and septic shock. A computed tomography scan revealed bilateral emphysematous pyelonephritis and emphysematous cystitis. The patient was treated with bilateral double-J stents and an indwelling urethral catheter. This is the first report of bilateral emphysematous pyelonephritis and emphysematous cystitis treated conservatively with drainage and an internal stent, which may be a treatment option.

1. Introduction

Separate diagnoses and treatments for emphysematous pyelonephritis and emphysematous cystitis have been established, although the treatment of emphysematous pyelonephritis combined with emphysematous cystitis has not been well reported.^{1,2} We report a case in which a patient with bilateral emphysematous pyelonephritis and emphysematous cystitis, which were thought to be related to vesicoureteral reflux (VUR), was successfully treated with drainage and ureteral stenting.

2. Case presentation

An 88-year-old female patient was admitted to our hospital with left-sided head contusions after falling in a facility. Her Glasgow Coma Scale score was 14 (E4V4M6), and her body temperature was 37.1 °C. Blood tests revealed the following results: white blood cell count 10,100/μL, neutrophils 82.1 %, blood urea nitrogen 25.2 mg/dL, creatinine 1.13 mg/dL, and hemoglobin A1c 6.5 %. Head computed tomography (CT) was performed to diagnose traumatic subarachnoid hemorrhage, and

the patient was admitted to the neurology department. She had a history of diabetes mellitus and familial VUR.

On day 4 of admission, her level of consciousness decreased to a Glasgow Coma Scale score of 10 (E2V3M5), with a blood pressure of 69/45 mmHg, body temperature of 38 °C, and gross hematuria. The patient was referred to the urology department, where emphysematous pyelonephritis and emphysematous cystitis were detected on abdominal CT. CT showed bilateral emphysema and a dilated renal pelvis, and emphysema was evident in the bladder, including the bladder diverticulum, despite the placement of a urinary catheter (Fig. 1). Blood tests and urinary findings were as follows: white blood cell count, 2500/μL; C-reactive protein, 25.04 mg/dL; blood urea nitrogen, 51.4 mg/dL; creatinine, 1.71 mg/dL; procalcitonin, 53.82; and urinary leukocytes and urinary occult blood, 3+. Considering the limitations of conservative treatment, she underwent bilateral stenting (right 6 Fr 26 cm, left 6 Fr 24 cm), which was changed to an 18 Fr urethral catheter. Oxygen and noradrenaline were administered. At the time of ureteral stent placement, the bladder was erythematous and markedly swollen subcutaneously due to emphysema, and it appeared as if the ureter had not

Abbreviations: VUR, Vesicoureteral reflux; CT, computed tomography.

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penetrated the wall (Fig. 2). Right and left pyeluria were observed with dark brown urine, and *Klebsiella pneumoniae* was detected in two sets of blood cultures. The patient was treated with the antibacterial meropenem for seven days, followed by augmentation for seven days, while the urinary catheter remained in place.

The patient was transferred to a rehabilitation hospital 35 days after stenting, where her general condition improved relatively well, although her food intake was poor. Considering her fitness, we decided on a policy of periodic ureteral stent and urethral catheter replacement. Three months after the first ureteral stent placement, the patient was admitted for the second time for stent replacement. VUR was prominent; however, bladder erythema improved, and there was no evidence of right or left pyeluria (Fig. 3). Seven months after the initial diagnosis, the patient was unable to eat, and she died.

3. Discussion

We found that bilateral emphysematous pyelonephritis and emphysematous cystitis can cooccur in patients with VUR and that ureteral stents may be effective if the emphysema is confined to the renal pelvis. The patient had hematuria and fever after admission, and since she did not have fever at the time of admission, VUR was considered. The patient was admitted with traumatic subarachnoid hemorrhage, which led to emphysematous cystitis, which in turn led to bilateral emphysematous pyelonephritis. The patient was elderly, had poor appetite, and may have had diabetes mellitus as an underlying disease. She had a family history of VUR and a urinary tract infection that had been neglected. The present case of emphysematous pyelonephritis and emphysematous cystitis was the ninth reported case of bilateral emphysematous pyelonephritis and cystitis at autopsy.^{1,2} Neurogenic bladder and VUR should be considered when there is significant reflux of retrograde contrast and bilateral findings.

Conservative treatment with ureteral stents and urethral catheters may be effective for treating emphysematous pyelonephritis and emphysematous cystitis triggered by VUR. A ureteral stent was considered effective, as the patient had hematuria at the time of fever, with blackish-brown drainage at the time of stenting. In general, emphysematous pyelonephritis has a mortality rate of 18%–21%.^{2,3} Class 4 emphysematous pyelonephritis has the worst prognosis, with a 50% mortality rate, based on the classification outlined by Huang et al.^{4,5} Predictors of mortality include low albumin, shock, bacteremia, need for hemodialysis, and multidrug bacilli, while poor conversion factors include shock and multidrug bacilli at initial presentation.³ When shock coexists, the mortality rate can increase to 54%.⁶ When a patient is in shock, conservative treatment with invasive procedures may be considered at the appropriate time.⁶ On the other hand, emphysematous cystitis has a mortality rate of 7%–7.4%, and treatment includes drainage and appropriate antimicrobial agents.^{7,8} When emphysema lesions involve other organs, the mortality rate increases to 14%–15%.^{1,7} There are few case reports of the cooccurrence of emphysematous

pyelonephritis and emphysematous cystitis, with eight cases of emphysematous pyelonephritis and bilateral emphysematous cystitis previously reported. Despite the combination of adverse conditions of bilateral emphysematous pyelonephritis and shock, the autopsy case was able to improve the patient's condition.^{5,6} The large amount of dark brown pyuria collected at the time of ureteral stenting suggested that the intervention was successful.⁸ There are two possible reasons for this improvement. First, the disease onset occurred while the patient was hospitalized, where blood glucose control and early intervention were possible.

Second, as the patient was diagnosed with class 4 emphysematous pyelonephritis with underlying VUR; the lesion was relatively confined to the renal pelvis, and the urethral catheter itself was effective, which may have been the reason for the improvement. Bilateral emphysematous pyelonephritis combined with emphysematous cystitis may be easier to treat than high-grade emphysematous pyelonephritis, depending on the status of the emphysema, as in this case.

Here, we report a case of bilateral emphysematous pyelonephritis and emphysematous cystitis complicated by drainage, including ureteral stenting. Depending on the site of inflammation and the underlying diseases, cases of combined emphysematous pyelonephritis and emphysematous cystitis can be treated. However, in this case, the patient was in a general condition that did not allow her to eat, and although she was treated and discharged from the hospital, she eventually died. It is important to inform families and patients that the combination of emphysematous pyelonephritis and emphysematous cystitis may result in a poor prognosis.

4. Conclusions

Here, we report a case of bilateral emphysematous pyelonephritis and emphysematous cystitis treated with drainage and ureteral stents. In complicated cases, treatment based on the underlying disease may be effective.

CRedit authorship contribution statement

Yoshihiro Kawaguchi: Writing – original draft, Visualization, Project administration, Methodology, Formal analysis, Data curation, Conceptualization. **Yoshikado Miyagawa:** Writing – review & editing, Supervision, Conceptualization. **Shigehisa Mizuta:** Writing – review & editing, Data curation, Conceptualization. **Kosuke Ueda:** Writing – review & editing, Supervision. **Kiyooki Nishihara:** Writing – review & editing, Supervision. **Makoto Nakiri:** Writing – review & editing, Supervision. **Tsukasa Igawa:** Writing – review & editing, Supervision.

Informed consent statement

Written informed consent was obtained from the patient for the publication of this case report.

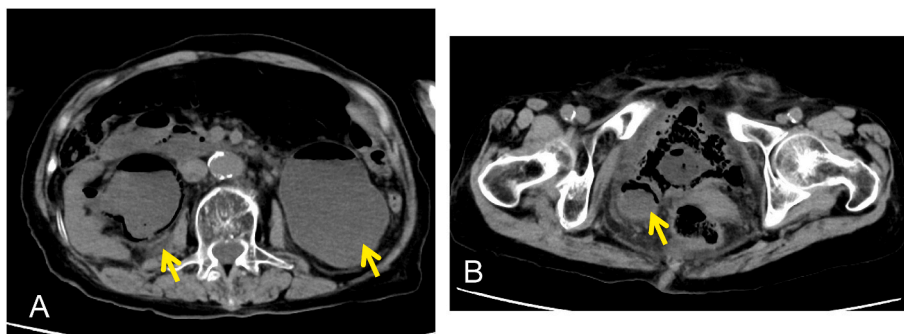


Fig. 1. A: Emphysema lesions in the bilateral renal pelvis with evidence of hydronephrosis. B: Emphysema lesions (arrows) around the cuff of the urethral catheter and the bladder diverticulum.

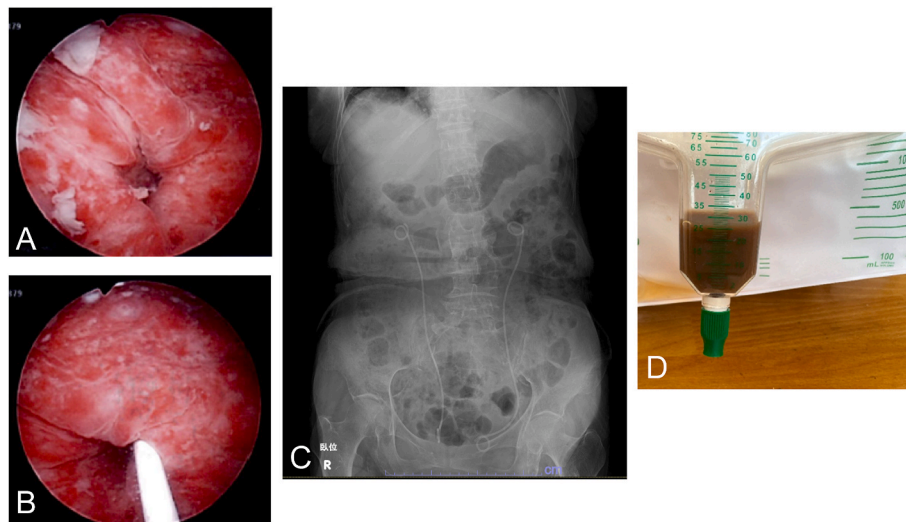


Fig. 2. A: Bladder erythema at the time of stent placement. The figure shows the right ureteral opening. The mucosa is markedly erythematous with edema. B: The left ureter was emphysematous and difficult to identify. C: Kidney ureter bladder findings. Bilateral 6-Fr 26-cm and 6-Fr 24-cm ureteral stents were also used. D: Drainage from the bilateral renal pelvis was performed, and dark brown urine was continuously drained. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

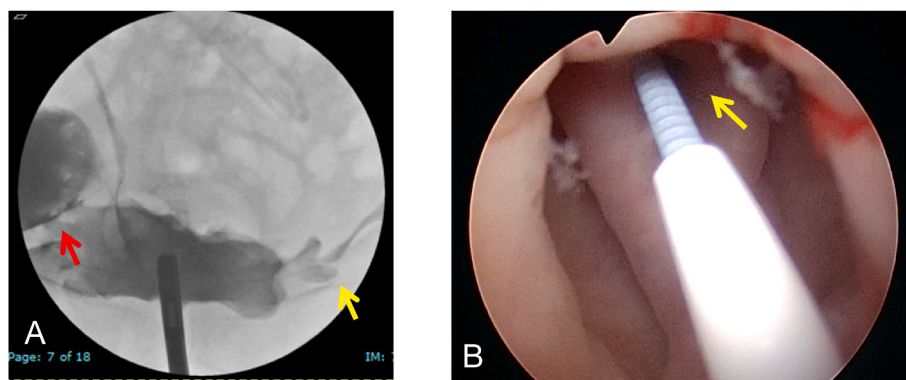


Fig. 3. A: Injection of 20–30 cc of contrast medium into the bladder revealed reflux into the left and right ureters. B: The left ureteral opening (yellow arrow) does not appear to penetrate the bladder wall. The red arrow indicates a bladder diverticulum. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

Data availability statement

The data are available from the corresponding author upon request.

Declaration of generative AI in scientific writing

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Declaration of competing interests

None.

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