



## Case of the Month

# 'Case of the Month' from University of Michigan, Ann Arbor, MI, USA: emphysematous pyelonephritis following ureteroscopy in a solitary kidney

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## Case Presentation

A 46-year-old man with diabetes mellitus and morbid obesity (body mass index 38 kg/m<sup>2</sup>) was referred to our clinic with asymptomatic right hydronephrosis; 5 months previously he presented with chronic urinary retention and renal impairment (GFR 59 mL/min/1.73 m<sup>2</sup>). Past history included bilateral megaureter with tapering and re-implantation in childhood. CT showed right hydroureteronephrosis and non-obstructing small renal calculi. The left kidney was atrophic resulting in functionally solitary right kidney. Cystoscopy and retrograde access to the right ureter was attempted by his local urologist and failed.

Urodynamic studies at our centre revealed an atonic bladder. He was managed with regular intermittent catheterisation. A dynamic renogram showed right renal obstruction, despite the patient making adequate urine and having a stable GFR. A percutaneous nephrostomy (PCN) was organised to allow for evaluation of the ureter. However, due to the coronavirus disease 2019 (COVID-19) pandemic this was delayed. He then developed left flank pain. A CT showed right distal ureteric calculi (largest 8 mm). A right PCN was placed followed by a right antegrade ureteroscopy (URS) and laser lithotripsy 4 weeks later. Multiple stones proximal to a ureteric orifice stricture in a megaureter were fragmented and a 7-F JJ stent was placed. A second-look retrograde URS was performed 6 weeks later. The patient was treated with preoperative antibiotics for bacteriuria, per our institutional protocol. Small fragments in the distal ureter were treated and a persistent stricture was confirmed. An 8-F Silhouette stent was placed (Applied Medical, Rancho Santa Margarita, CA, USA). The patient was discharged the same day, with a plan for reconstructive ureteric surgery.

The following day (postoperative day [POD] #1), the patient presented to his local hospital with a fever, dyspnoea and in septic shock. After negative severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) testing, he was transferred to our institution. CT showed an appropriately positioned JJ stent, mild right hydronephrosis,

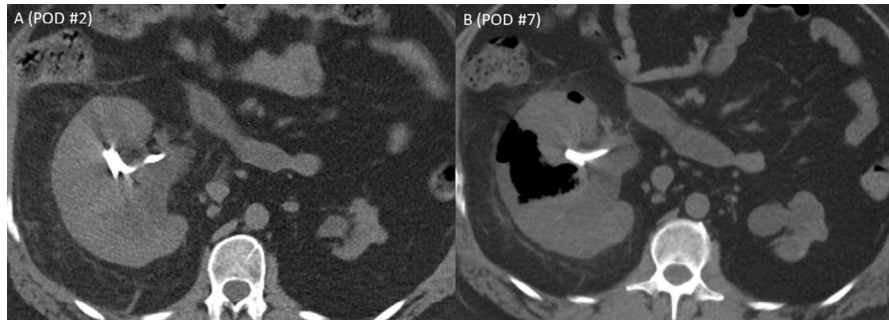
and a small focus of gas in the collecting system thought to be from the recent instrumentation (Fig. 1A). He was in septic shock with multi-organ dysfunction and required intensive care unit (ICU) admission; ICU care included systemic antibiotics (piperacillin-tazobactam), vasopressors, mechanical ventilation, and continuous renal replacement therapy. He improved and was extubated on POD #4. Blood cultures grew *Klebsiella aerogenes*, resistant to piperacillin-tazobactam. Renal ultrasonography was performed on POD #6 due to persistently positive blood cultures. This showed possible emphysematous pyelonephritis (EPN), which was confirmed on non-contrast CT (Fig. 1B).

## Management Considerations

All treatment options including medical management, PCN or percutaneous drain (PCD) placement, and emergent nephrectomy in case of clinical deterioration were discussed. At the time of EPN diagnosis, despite the striking CT findings, he had no flank pain or tenderness that would be expected of pyelonephritis and was off vasopressors and extubated. He required renal replacement therapy for acute kidney injury. Antibiotic therapy was changed to cefepime due to the resistance profile of the *Klebsiella* isolate.

The dilemma we had was whether to place a PCD or PCN (or both) into the right kidney. He had a functionally solitary kidney, which allowed for accurate assessment of urine formation and excretion. There was good urinary drainage via the JJ stent and minimal hydronephrosis. The parenchyma had multiple foci of gas, but no fluid collection. The risk of a severe bleeding complication from PCD placement in an improving patient with morbid obesity and solitary kidney had to be carefully considered. After multi-disciplinary consultation, the consensus was to continue management with systemic antibiotics and the JJ stenting, and place a PCD if the clinical condition changed. An open surgery team was coordinated in case of the need to perform salvage nephrectomy.

**Fig. 1** (A) Initial CT when hospitalised with septic shock showing small locules of gas in the collecting system in the right kidney and the JJ stent *in situ*. (B) Follow-up CT scan showing EPN with gas in the parenchyma of right kidney.



## Treatment Course

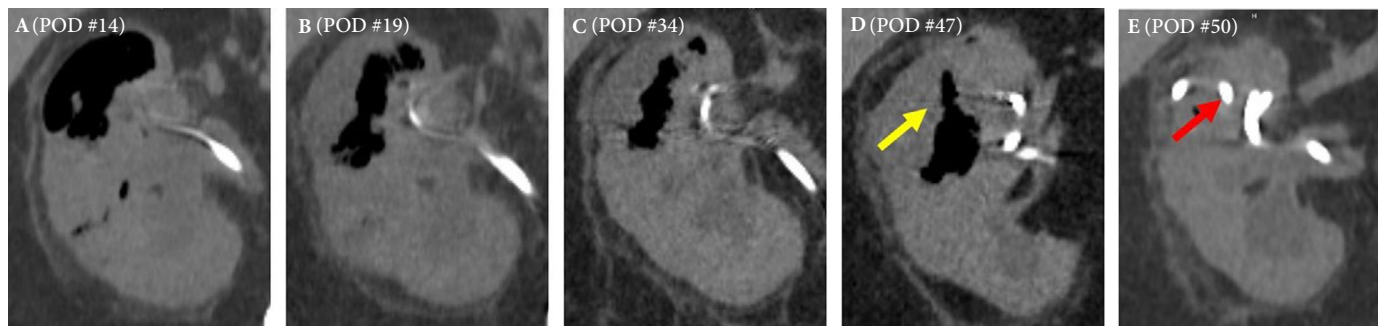
The patient was transferred out of ICU on POD #9. Additional CTs were obtained to assess gas appearance, especially as the patient complained of no pain or flank tenderness. These showed minimal change (Fig. 2). Just prior to discharge, on POD #22, he developed anaemia and melena, requiring blood transfusion. Gastroscopy revealed a duodenal ulcer and *Helicobacter pylori*, which was managed medically. Haemodialysis was discontinued on POD #23. The GFR stabilised at 17 mL/min/1.73 m<sup>2</sup>. He was ultimately discharged on POD #30 on a long-term course of intravenous ertapenem with the plan for serial scans to assess EPN resolution. On POD #43 he had right flank pain for the first time. A CT on POD #47 gave concern for a developing 6.2-cm right upper pole abscess. He underwent uncomplicated PCD placement on POD #48 with drainage of purulent material (*K. aerogenes*). The drain was removed 14 days later. He recovered well thereafter, returning back to work. On POD #89 he underwent a ureteric stent exchange without complication.

## Discussion

Sepsis after URS for urinary stones is uncommon, but can be fatal. In 1737 patients undergoing URS in 11 practices from the Michigan Urological Surgery Improvement Collaborative, the hospitalisation rate for infection-related complications was 2.4%, and the mortality rate was 0.2% [1]. Risk factors included comorbidity, large stone size, and history of recurrent UTIs. EPN specifically after URS has not been well documented. Our present patient had the risk factors of morbid obesity, diabetes, obstructive uropathy, and bacteriuria. Despite URS mitigated with preoperative antibiotics, the patient developed sepsis.

Emphysematous pyelonephritis is a rare but potentially life-threatening necrotising infection of the renal parenchyma characterised by imaging findings of gas in the renal collecting system, parenchyma, or perinephric tissue. Clinical presentation is variable but typically includes fever, flank pain, and vomiting. A significant number of patients will present with sepsis and bacteraemia. Risk factors include female gender, diabetes, and urinary obstruction [2]. Huang

**Fig. 2** Serial CTs of EPN managed with antibiotic therapy and JJ stenting. Inpatient scans showing (A) Stable appearance of gas in parenchyma and (B) Improving appearance of gas in parenchyma. Outpatient scans showing (C) continued parenchymal gas and (D) subsequent gas with concern for developing abscess (yellow arrow). Inpatient scan showing (E) percutaneous drain placement (red arrow) demonstrating resolution of abscess and gas.



**Table 1** Summary of three large case series of EPN management and outcomes.

Author	Country	N	Patients undergoing treatment modality, n (%)						Survival, n (%)	Comments
			Antibiotics alone	Percutaneous drainage	JJ stent	JJ and drain	Open drainage	Salvage nephrectomy	Emergent nephrectomy	
Huang and Tseng 2000 [2]	Taiwan	48	5 (10)	41 (56)	–	–	–	8 (16)	2 (4)	In Class 1 and 2 EPN, all patients treated with a PCD or JJ stent survived
Lu <i>et al.</i> 2016 [4]	Taiwan	51	10 (20)	39 (76)	1 (2)	–	1 (2)	5 (9.8)	1 (2)	If Class 2–4 EPN and risk factors, carbapenem was recommended as initial therapy
Sokhal <i>et al.</i> 2017 [5]	India	74	12 (16)	44 (59)	28 (38)	10 (14)	2 (3)	18 (24)	4 (5)	16% had ureteric calculi; 24% had renal calculi

and Tseng [2] developed a classification system for the severity of EPN: Class 1, gas in the collecting system only; Class 2, parenchymal gas only; Class 3A, extension of gas into perinephric space; Class 3B, extension of gas into pararenal space; and Class 4, EPN in a solitary kidney, or bilateral disease.

Management of EPN has evolved. Parenteral antibiotics targeting gram-negative organisms should be initiated in all cases, as these are the most common organisms to cause EPN. Appropriate empiric therapy includes a  $\beta$ -lactam/ $\beta$ -lactamase inhibitor or a third–fourth generation cephalosporin. However, some Enterobacteriaceae (including *K. aerogenes*) may produce an AmpC  $\beta$ -lactamase, which can lead to development of  $\beta$ -lactam resistance; this was suspected in the present case. Cefepime or a carbapenem are preferred therapy in the setting of a known AmpC-producing organism. A switch to a carbapenem or fluoroquinolone may be required in this situation and infectious diseases consultation is encouraged. Because EPN is a deep infection, prolonged therapy (4–6 weeks) is required. Our present patient was treated with 6 weeks of ertapenem and transitioned to prophylactic ciprofloxacin until stent exchange.

Historically, EPN was managed with emergent nephrectomy and antibiotic therapy. Ahlering *et al.* [3] reported on 13 patients, of which 12 (92%) underwent emergent nephrectomy. The mortality rate was 42%, and the authors suggested early nephrectomy to reduce mortality. However, contemporary data has shown that EPN can be managed successfully with antibiotics and endourological or percutaneous drainage without nephrectomy in the majority of cases (Table 1). Lu *et al.* [4] reported an overall survival of 90% in 51 patients. PCD was needed in 73%. One patient with EPN after URS with JJ stenting was treated successfully with antibiotic therapy. Sokhal *et al.* [5] reported on 71 patients with a survival of 92%; 38% were managed with a JJ stent. In their assessment, JJ stenting or PCD was based on clinical and radiological extent of disease. The presence of internal echoes in the collecting system with obstruction was an indication for PCD.

Our present case highlights the challenge of managing a patient who developed EPN during the course of treatment for septic shock after URS. Initial therapy must take into consideration the patient's clinical status, and emergent nephrectomy may be required in critically ill patients as a life-saving strategy. Our present patient had a JJ stent with good urinary drainage established; therefore, PCN was not thought to add benefit. PCD was strongly considered, but not performed initially due to improving clinical condition with appropriate antibiotics and supportive care, and lack of localising collection on imaging. The occurrence of gastrointestinal bleeding highlights the cascade of events that can occur in patients who develop complications.

Nephrectomy was not favoured as it would have rendered the patient functionally anephric. Nonetheless, an open surgical team was coordinated in case of need for salvage nephrectomy, as this is an operation with high mortality. We found that with antibiotic therapy and JJ stenting, the emphysematous appearance improved minimally over time. Eventually, the phlegmon coalesced into a collection suitable for drainage.

## Conclusion

The management dogma of EPN has changed from emergent nephrectomy to a kidney-sparing approach with antibiotic therapy and percutaneous and/or endourological drainage depending on clinical and radiological features. Our present patient was managed in a multi-disciplinary manner with medical therapy and JJ stenting. Serial CTs showed minimal resolution of the emphysematous changes, which eventually progressed to abscess formation that was successfully drained. Recent studies demonstrate the overall survival of EPN is as high as 92%, but nephrectomy is needed in those who do not respond. PCD in a solitary kidney has risks and in patients who are clinically improving with a stent can be forestalled. If there is any deterioration, drainage must be promptly provided.

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## Conflict of Interest

None.

## References

- 1 Cole A, Telang J, Kim TK et al. Infection-related hospitalization following ureteroscopic stone treatment: results from a surgical collaborative. *BMC Urol* 2020; 20: 176
- 2 Huang JJ, Tseng CC. Emphysematous pyelonephritis: clinicroadiological classification, management, prognosis, and pathogenesis. *Arch Intern Med* 2000; 160: 797–805
- 3 Ahlering TE, Boyd SD, Hamilton CL et al. Emphysematous pyelonephritis: a 5-year experience with 13 patients. *J Urol* 1985; 134: 1086–8
- 4 Lu YC, Hong JH, Chiang BJ et al. Recommended initial antimicrobial therapy for emphysematous pyelonephritis: 51 cases and 14-year-experience of a tertiary referral center. *Medicine* 2016; 95: e3573
- 5 Sokhal AK, Kumar M, Purkait B et al. Emphysematous pyelonephritis: changing trend of clinical spectrum, pathogenesis, management and outcome. *Turkish J Urol* 2017; 43: 202–9

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Abbreviations: EPN, emphysematous pyelonephritis; ICU, intensive care unit; PCD, percutaneous drain; PCN, percutaneous nephrostomy; POD, postoperative day; URS, ureteroscopy.