



Unusual Presentation of Emphysematous Pyelonephritis (EPN): "Double trouble" A Case Report

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Authors' contributions

This work was carried out in collaboration among all authors. Author AMM designed the study, performed the statistical analysis, wrote the protocol, and wrote the first draft of the manuscript. Authors SS A/L CS and AH managed the analyses of the study. Author AMM managed the literature searches. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Introduction: Emphysematous Pyelonephritis (EPN) is a possibly life-threatening condition that is usually present as sepsis. Delay in identifying the disease may lead to detrimental outcomes, even mortality. The present study reports a case of incidental finding of an EPN in a perforated viscus patient. Studies also advocate the minimally invasive approach of percutaneous drainage with antibiotics in asymptomatic EPN patients. Historically, EPN was managed by nephrectomy or open drainage along with antimicrobial therapy but resulted in high mortality of 40-50%. Introduction of percutaneous drainage had shown better outcome compared to nephrectomy.

Case Presentation: A 63-year-old gentleman, with underlying Diabetes Mellitus (DM), initially presented with a sudden onset of severe generalized abdominal pain and distension. Examination revealed peritonitic abdomen. Erect CXR revealed air under the diaphragm suggestive of perforated hollow viscus. Patient underwent exploratory laparotomy with intraoperative findings of

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perforated prepyloric gastric ulcer that managed with modified Graham patch. Besides, there was also huge retroperitoneal mass. Post operatively, patient subjected for abdominal and pelvic CT that revealed right EPN. It was managed with antibiotic and percutaneous drainage. Patient was responded and recovered.

Conclusion: EPN is a fatal disease that requires early detection with a high index of suspicion particularly in patients with signs of sepsis and pyelonephritis. Although it is rare, in subjects with pneumoperitoneum and the presence of pathology over renal area, EPN should be one of the differential diagnosis. In this case, it is possible that the presence of EPN poses stress to the patient leading to development of perforated viscus.

Keywords: Emphysematous pyelonephritis; diabetes mellitus; nephrectomy; mortality.

1. INTRODUCTION

EPN is defined as a severe gas forming infection of renal parenchyma and its neighbouring structure [1]. It is a possible life-threatening condition that is usually present as sepsis. Delay in identifying the disease may lead to detrimental outcomes, even mortality. However, in rare cases, its presentation diverts its normal clinical pictures, masquerading the true pathology. We present a case of incidental finding of an EPN in a perforated viscus patient. We advocate the minimally invasive approach of percutaneous drainage with antibiotics in asymptomatic EPN patients. However, serial radiological reassessment should be carried out to see the resolution of the abscess collection.

2. CASE REPORT

A 63-year-old gentleman, with underlying Diabetes Mellitus (DM), initially presented with a sudden onset of severe generalized abdominal pain and distension. He was tachycardic and dehydrated. Abdominal examination revealed distended abdomen with peritonism. Laboratory investigation showed leucocytosis with WBC of $23.9 \times 10^9/L$ and acute kidney injury (AKI) evidenced by elevated urea of 11.8 mmol/L and creatinine of 131 $\mu\text{mol/L}$. The urine examination showed red blood cells 3+, leucocyte 3+. We proceeded with an Erect chest X-ray showed air under the diaphragm. Our initial diagnosis was perforated hollow viscus with differential diagnosis of perforated gastric ulcer (PGU) /perforated duodenal ulcer (PDU). His diabetes was managed with insulin infusion (actrapid infusion) which was later replaced with basal bolus insulin after the blood sugar was optimized. He was then subjected for operation where he underwent exploratory laparotomy with modified Graham Patch repair for perforated prepyloric gastric ulcer measuring 2cm as shown in Fig. 1. Intraoperatively, there was a huge retroperitoneal mass measuring 8cm x 8cm over the right side,

fixed, non-pulsating with no obvious signs of inflammation.

Postoperatively, the patient had a speedy recovery and was discharged home well.

He was then planned for contrast enhanced abdominal and pelvic CT to investigate the retroperitoneal mass. Abdomen and pelvic CT revealed features suggestive of right EPN with large perinephric abscess measuring 9.2x6.7x10.9cm and bilateral ureteric calculi as portrayed in Fig. 2. We proceeded with ultrasound guided percutaneous drainage and thick pus was aspirated and sent for cultures. The pus culture grew Extended Spectrum β -Lactamase Klebsiella Pneumoniae. He had uneventful recovery afterwards, however the pigtail catheter meant for pus drainage prematurely dislodged. A repeat ultrasound revealed residual decreasing size collection. He was planned for conservative management with antibiotics (initially he was started with intravenous cefoperazone and metronidazole and completed for one week). It was later changed to cefuroxime based on its pus culture and sensitivity for another 2 weeks. He had frequent visits to surgical outpatient clinic. His latest visit was unremarkable and showing good recovery.

3. DISCUSSION

EPN is defined by severe necrotizing and gas forming infection of the renal parenchyma and its surrounding structure namely perinephric tissue and collecting system [1,2]. The first documented case of EPN was described in 1898 and the term EPN coined in 1962 to correlate between acute kidney infection with gas forming condition [1].

The incidence for female is more common than men, with a mean age of 40-50 years old [2]. It had been reported that left kidney is affected more than right without apparent explanation

[2]. Usually, it presented with sepsis and pyelonephritis signs such as fever with rigor, lumbar pain and urinary tract infection symptoms [3]. However, in some extreme cases, peritonitis with pneumoperitoneum suggested of perforated viscus had been reported [4,5]. This results from the extension of the disease into the peritoneum. This unique presentation is a red-herring resulting in a different approach in managing the disease. However, in our case the patient presented with peritonitis secondary to perforated gastric ulcer with incidental finding of EPN. This caused a diagnostic dilemma and difficulty in managing this case.

Most EPN is associated with immunocompromised state particularly diabetes mellitus where it occurrence in diabetic patients

accounts for more than 95% of all EPN cases [6,7]. Besides, urinary tract obstruction also predispose patients to develop EPN [6].

Since 1984, multiple classification systems developed in order to aid in managing the disease as illustrated in Table 1. The latest composition by Huang et al had provided objective means in treating EPN [7].

EPN is a devastating condition that bears high risk of morbidity and mortality mainly due to septic complications. Variety of risk factors identified to prognosticate the outcome of the illness. The meta-analysis had concluded several factors including patient who was treated conservatively, type 1 EPN, bilateral EPN and patient with low platelet (thrombocytopenia) [8].

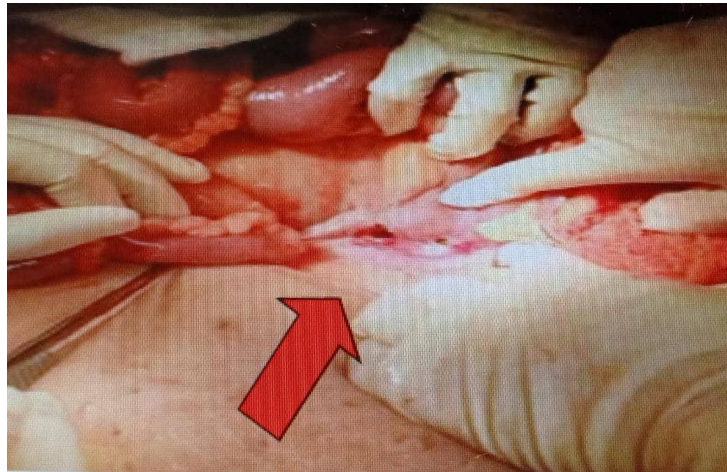


Fig. 1. Perforated prepyloric gastric ulcer

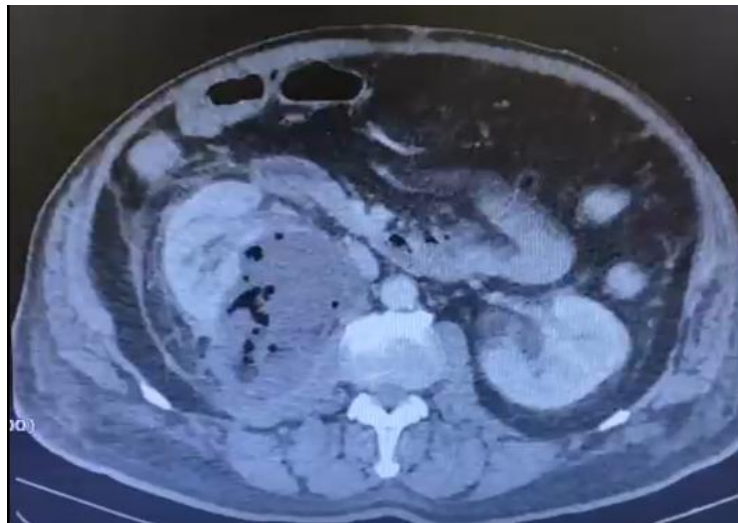


Fig. 2. Right EPN with large perinephric abscess

Table 1. Classification of EPN

Author	Radiological modality	Class
Michaeli et al [4]	Plain x-ray with intravenous urography	I : Gas in the parenchyma or perinephric tissue II : Gas in the kidneys and it surroundings III : Gas through fascia or bilateral kidney involvement
Wan et al [5]	CT scan	I : Renal necrosis with presence of gas but no fluid II: Renal or perinephric fluid collection associated with gas in the collecting system.
Huang et al [6]	CT scan	1. Gas only in collecting system 2. Gas only in renal parenchyma 3A. Perinephric space involvement 3B. Pararenal space involvement 4. Bilateral kidney involvement or solitary kidney with EPN

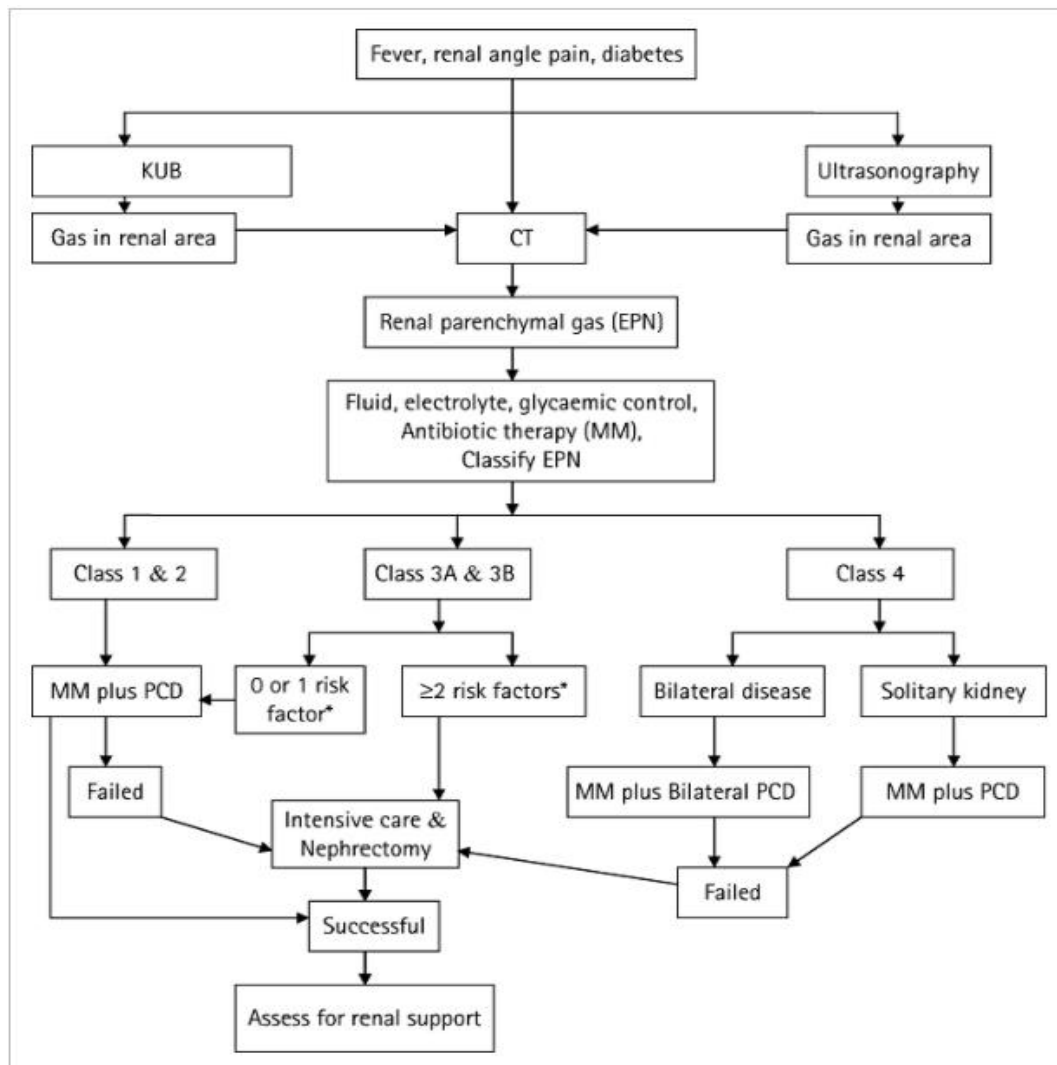


Fig. 3. Management algorithm of EPN. (Risk factors: Diabetes, thrombocytopenia, acute kidney injury, altered sensorium, shock) [7]

The diagnosis of EPN is radiological supported by the presenting history and laboratory investigations [5]. Although plain radiograph may show presence of air near the kidney, the gold standard modality in diagnosing EPN is undoubtedly CT scan [9]. CT scan yields 100% accuracy compared to plain radiograph which only positive up till 69% of the cases [9].

Historically, EPN was managed by nephrectomy or open drainage along with antimicrobial therapy but resulted in high mortality of 40-50% [4]. After the introduction of minimally invasive percutaneous drainage, the management shifted towards percutaneous drainage, and this method proved to yield better outcome. A systematic review advocated percutaneous drainage as the initial management of EPN whereas it is associated with reduced mortality compared to emergency nephrectomy or medical management however, nephrectomy may be required in some cases not responding to the aforementioned treatment [9]. Fig. 3 demonstrates the management algorithm extracted from Huang et al. [7].

Referring to our case, as the patient was asymptomatic, percutaneous drainage with medical management were the treatment of choice. Radiological imaging repeated after one month showed improvement. Chen et al had concluded that imaging should be repeated after 4-7 weeks and from their cohort, the treatment lasted for approximately 3 months [10]. Based on study by Chen MT et al, their patient was followed up till 10 years with mean of 5 years (follow up till clinically and radiologically resolved) showed no recurrence or complications [10]. Our patient is still on regular follow up and planned to be followed up till complete clinical and radiological resolution. In cases which do not respond to percutaneous drainage, nephrectomy should be considered [6]. As EPN is a rare occurrence, there is limited consensus pertaining to the treatment of incidental finding of EPN during an exploratory laparotomy. However, a safer approach must be undertaken to avoid causing more harm to the patients particularly in asymptomatic patients. Open drainage with posterior approach (flank incision) is one of the methods of choice [11].

4. CONCLUSION

EPN is a fatal disease that requires early detection with a high index of suspicion

particularly in patients with signs of sepsis and pyelonephritis. Although it is rare, in subjects with pneumoperitoneum and the presence of pathology over renal area, EPN should be one of the differential diagnosis. In this case, it is possible that the presence of EPN poses stress to the patient leading to development of perforated hollow viscus.

CONSENT

As per international standard or university standard, patients' written consent has been collected and preserved by the authors.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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