

## Case Report

## Emphysematous pyelonephritis initially presenting as a spontaneous subcapsular hematoma in a diabetic patient

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## A B S T R A C T

## Article history:

Received 19 March 2014

Received in revised form

29 April 2014

Accepted 9 May 2014

Available online 8 July 2014

## Keywords:

Emphysema

Hematoma

Pyelonephritis

Emphysematous pyelonephritis (EPN) is a life-threatening infection characterized by the formation of gas. Complications of EPN include septic shock, acute renal failure, and disseminated intravascular coagulation. Spontaneous subcapsular hematoma (SCH) has also been reported as a rare complication of EPN, although there have been no reports to date of this occurring prior to the presentation of EPN. We report a case of EPN that initially presented as spontaneous SCH. The patient was admitted for left flank pain, and initial computed tomography revealed SCH without any air shadows. Laboratory findings and clinical symptoms suggested the presence of urinary tract infection and the patient was started on antibiotics. Fever developed 24 hours after admission. On follow-up computed tomography 7 days later, EPN was newly observed, and a percutaneous drain was inserted. Blood, urine, and drainage fluid cultures all revealed growth of extended-spectrum  $\beta$ -lactamase-negative *Escherichia coli*.

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## Introduction

Emphysematous pyelonephritis (EPN) is a severe necrotizing, gas-forming infection of the renal parenchyma and surrounding tissue. It commonly occurs in patients with uncontrolled diabetes mellitus or obstruction within the urinary tract [1]. EPN has been associated with a high mortality rate, usually due to sepsis, and has therefore been treated with early, aggressive therapy such as nephrectomy. With improvement of minimally invasive intervention strategies, antibiotics, and general supportive care, however, there has been a paradigm shift in the treatment of EPN towards an approach that preserves renal function [2].

Spontaneous subcapsular hematoma (SCH) has been reported in association with malignant and benign tumors, vascular disorders and approximately 7% of cases are associated with kidney infection [3]. There have been a few reported cases of EPN with spontaneous SCH both locally and internationally [4–8]. However, all of these cases initially presented with EPN and had a subsequent diagnosis of SCH. In this report, we describe a case of EPN that initially presented as a spontaneous SCH in a diabetic patient that was successfully treated with percutaneous drainage and antibiotics.

## Case report

A 57-year-old woman with a history of diabetes mellitus and hypertension presented to our emergency room with 2 days of sudden-onset left flank pain. She had no history of recent trauma, hematological abnormalities, liver disease,

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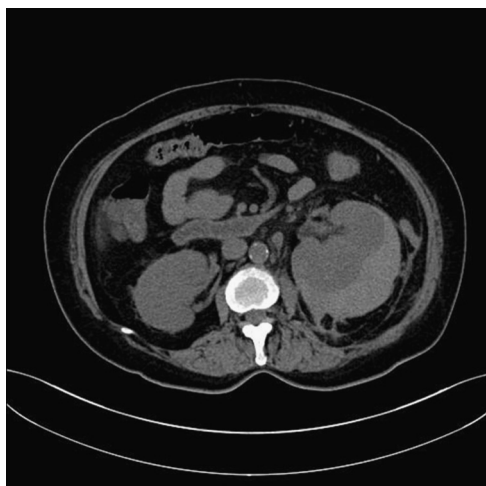
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or kidney disease, with the exception of acute pyelonephritis (APN) that required hospitalization 4 years previously. Her complete blood count and blood chemistry levels taken 6 months prior to this presentation were all within normal limits. Her medication list included anti-diabetics (metformin 850 mg, glimepiride 2 mg, and vildagliptin 50 mg), antihypertensives (cilnidipine 10 mg), and acetaminophen (650 mg 3 times daily) for intermittent leg pain. Her vital signs at the time of admission were as follows: blood pressure 96/52 mmHg, heart rate 118 beats/min, respiratory rate 28 breaths/min, and body temperature 37.3°C. On physical examination, she was mentally alert, but appeared acutely ill. Palpation of her abdomen revealed diffuse abdominal tenderness, especially on the left side, with severe left costovertebral angle tenderness. Her laboratory data showed a white blood cell count of 23,010/ $\mu$ L (82% neutrophils and 12% band-neutrophils), hemoglobin of 11.9 g/dL, platelet count of 76,000/ $\mu$ L, high-sensitivity C-reactive protein of 29.89 mg/dL, urea nitrogen of 48.3 mg/dL, creatinine of 3.44 mg/dL, and prothrombin time (international normalized ratio) of 1.32. Hemoglobin A1c level was 10.1%, indicating that her diabetes was poorly controlled. Urinalysis demonstrated 10–19 white blood cells and many red blood cells per high-power field. Protein (2+) and occult blood (4+) were also detected, and her urine specific gravity was 1.02. Spot urine chemistry showed a protein/creatinine ratio of 1.79 (g/g), and the fractional excretion of sodium was calculated to be 0.52%. A noncontrast computed tomography (CT) scan of the abdomen revealed a crescentic high-density fluid collection of 2.2 cm in diameter surrounding the left kidney with extension into the infrarenal space. These findings were consistent with a diagnosis of SCH (Fig. 1).

The patient was afebrile on presentation, although her initial laboratory results suggested the presence of urinary tract infection. Therefore, blood and urine culture were obtained and empiric antibiotic treatment was initiated (intravenous ceftriaxone 2 g/d). Additionally, she was given intravenous fluids to maintain her hydration status, as well as insulin for glucose control. A urologist was consulted regarding surgical management of her SCH. However, because the patient's vital signs were stable after initial medical management, it was decided instead to observe her closely while treating conservatively with antibiotics, hydration, and bed

rest. The patient developed a fever of 38.2°C within 24 hours of admission, but it subsided within 1 day. On Day 4 of admission, she continued to have pain in her left flank, left lower quadrant, and left upper quadrant. Both urine and blood cultures revealed growth of extended-spectrum  $\beta$ -lactamase (ESBL) negative *Escherichia coli*. Additional laboratory studies showed a white blood cell count of 12,790/ $\mu$ L, platelet count of 130,000/ $\mu$ L, hemoglobin of 9.5 g/dL, and creatinine of 1.11 mg/dL. On hospital day 7, a repeat CT scan was performed with the use of contrast dye enhancement. Newly noted air shadows, consistent with EPN of the left kidney with parenchymal necrosis were observed (Fig. 2). There was a subtle interval increase in the size of the SCH of the left kidney and an interval increase in perirenal and infrarenal haziness and fluid collection.

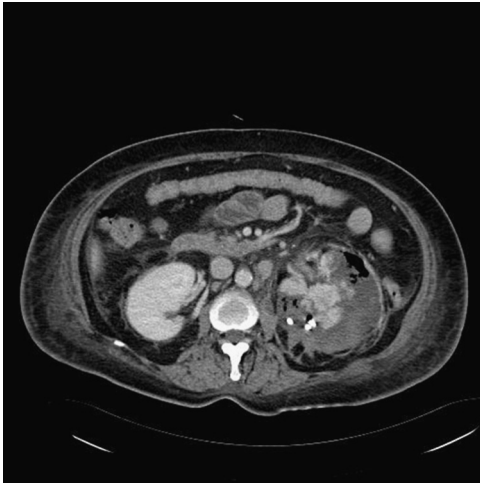
Percutaneous drainage (PCD) of both the intrarenal air and perinephric hematoma was performed using an 8.5 Fr pigtail catheter. Although her clinical symptoms and laboratory values showed improvement, imaging studies showed progression to EPN. Therefore, the patient's antibiotics were changed to intravenous piperacillin/tazobactam (Tazocin) 4.5 g every 8 hours. The initial pigtail drainage was dark and bloody in color, with approximately 150 mL of fluid collected on the 1<sup>st</sup> day. Culture of the drainage fluid also revealed growth of ESBL-negative *E. coli*, which was the same pathogen identified in the blood and urine cultures. A diagnosis of EPN with sepsis due to ESBL-negative *E. coli* and concomitant SCH was made. The patient did not have any subsequent fever, her platelet count was within normal limits (405,000/ $\mu$ L), and her left flank pain improved over time. A follow-up CT scan performed on hospital day 12 showed mildly decreased EPN and a smaller hematoma on the side where the catheter had been placed, but mildly increased hematoma and air shadows on the side without drainage (Fig. 3). Therefore, an additional catheter was inserted. Follow-up CT scan performed on hospital day 29 showed decreased air on the right anterior side but residual air on the posterior side (Fig. 4). We subsequently removed the anterior catheter and repositioned the posterior catheter so that the tip was located within the air pocket in the left posterior pole. The patient's clinical symptoms and laboratory data continued to show improvement and she was



**Figure 1. Initial non-contrast abdominal CT scan.** A crescentic high-density fluid collection of 2.2 cm in diameter surrounding the left kidney with extension into the infrarenal space is noted.



**Figure 2. Follow-up contrast enhanced CT scan on hospital day 7.** New air shadows consistent with emphysematous pyelonephritis of the left kidney, as well as a subtle interval increase in the size of the subcapsular hematoma are noted.



**Figure 3. Follow-up CT scan performed on hospital day 12.** The scan shows mildly decreased emphysematous pyelonephritis and a smaller hematoma on the side in which the catheter had been placed, but a slightly larger hematoma and increased air shadows on the side without drainage.



**Figure 4. Follow-up CT scan performed on hospital day 29.** The scan shows decreased air compared to the CT obtained on hospital day 7. The overall size of the kidney as well as the subcapsular hematoma has also decreased, and much of the renal parenchyma is spared.

discharged home on hospital day 34 with oral antibiotics (ciprofloxacin 500 mg every 12 hours).

At her outpatient follow-up appointment 11 days after discharge, almost no drainage was observed from the pigtail catheter. The catheter was removed, and follow-up CT scans showed almost no air pockets and markedly decreased hematoma. The patient was placed on oral antibiotics for 1 more month.

## Discussion

Spontaneous SCH without a history of trauma or malignancy is rare, and SCH due to infection accounts for <7% of cases. CT scans are most useful for the diagnosis of SCH and evaluation of the underlying disease. However, if no mass or probable underlying cause is identified initially, a serial CT scan or renal angiography may be performed for detection of

vascular malformations [9]. Our patient had no history of trauma, malignancy, or vascular disease, and was diagnosed with spontaneous SCH due to EPN on a follow-up CT scan.

To the best of our knowledge, this is the first case of EPN initially presenting as spontaneous SCH. This early onset of bleeding imposed further difficulties in diagnosing and treating the patient, because infection is rarely the cause of spontaneous SCH. Although the etiology of our patient's early bleeding remains unclear, it may have been caused by thrombocytopenia secondary to disseminated intravascular coagulation, which has been suggested as a poor prognostic factor in severe cases of EPN [10]. Sepsis and disseminated intravascular coagulation may have occurred as complications of the rapidly progressing EPN, resulting in bleeding before air shadows could be detected on the CT images.

There are several approaches to treating EPN with spontaneous SCH, including medical management with antibiotics, PCD, or surgical management with nephrectomy or open-surgical drainage. The mortality associated with EPN has previously been reported to be 40–50% [11], but Somani et al [12] reported that the mortality decreased significantly with the use of PCD. Koh et al [13] suggested that if fever and septic conditions persist after sufficient antibiotic therapy and PCD, elective nephrectomy should be performed. Our patient had residual gas and a hematoma after 1 month of treatment; however, nephrectomy was not indicated as the patient did not show signs of sepsis or ongoing bleeding. Bok et al [14] reported a case of EPN that responded to antibiotic treatment alone. Intrarenal gas persisted in this patient for > 1 month, but resolved after 2 months treatment. You et al [8] reported a case of APN with SCH that was successfully treated with intravenous antibiotics alone, and Kim et al [7] and Choi et al [6] reported cases of APN with SCH that were successfully treated with PCD and antibiotics. Our patient also showed rapid improvement of clinical symptoms and decreased intrarenal air and hematoma with appropriate infection control and drainage.

In conclusion, our case was a rare example of EPN initially presenting as spontaneous SCH that was successfully treated with PCD and antibiotic therapy. Even though infection is a rare cause of SCH, a high index of suspicion must be maintained as timely diagnosis and treatment may be crucial in preserving renal function in the patient.

## Conflict of interest

All authors declare no conflict of interest.

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