

Emphysematous Pyelonephritis and Hiccups, a Case Report

Muhanna Kazempour,¹ Mahbobeh Oroei,² Minoosh Shabani,³
Taraneh Faghihi Langroudi⁴

¹Department of Internal Medicine, Loghman-Hakim Hospital, Shahid Beheshti of Medical Science, Tehran, Iran

²Community Medicine Specialist, Hearing Disorders Research Center, Loghman-Hakim Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran

³Infectious Disease and Tropical Medicine Research Center, Shahid Beheshti University of Medical Sciences, Tehran, Iran

⁴Department of Radiology, Shahid Beheshti of Medical Science, Tehran, Iran

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The initial manifestation of emphysematous pyelonephritis (EPN) with hiccups is extremely rare. We report a 47-year-old diabetic man with complaint of persistent hiccups as the first manifestation. Class 3B EPN was confirmed based on findings of Contrast-enhanced abdominal multi-slice CT scan. There were no neurologic deficits, Medications that lead to hiccups and gastrointestinal dysfunction. Our patient was treated with antimicrobial therapy and double J stenting. His hiccups completely resolved in one week and had no relapse.

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INTRODUCTION

A hiccup is an involuntary, spasmodic contraction of the diaphragm, that is usually transient and harmless.¹ The hiccups that last for 48 hours are likely considered a serious problem and should be comprehensively evaluated. Hiccups can be due to gastrointestinal problems but sometimes it may be side effect of drugs such as corticosteroids, benzodiazepines, and barbiturates.² There are some reports about prolonged hiccup (lasting \geq 48 hours) or intractable hiccup (lasting \geq one month)³ in the patients with acute neurological disorders.^{1,4-7}

Emphysematous pyelonephritis (EPN) is a life threatening urologic disorder that is not common but is mostly presented in diabetics, urinary tract obstruction, end stage renal disease and immunosuppression. Contrast-enhanced CT is a main diagnostic method as it shows presence

of the gas in the parenchymal of renal.⁸ The clinical manifestation of EPN is similar to acute pyelonephritis, which includes fever, nausea, vomiting, abdominal pain or an abdominal mass with tenderness, pyuria, and leukocytosis. Here, we report a rare case with a persistent hiccup.¹⁰

CASE PRESENTATION

A 47-year-old man with diabetes mellitus was referred to Shahid Modarres hospital of Shahid Beheshti University of Medical Sciences in April 2018. He was suffering from prolonged hiccups for 1 week that had disrupted his sleep and daily activities. We found no clinical complains except hiccup and mild right flank pain from 4 days. He had no history of alcohol consumption. He had no symptoms of heartburn or regurgitation. He had a 2-year history of renal stone and type two

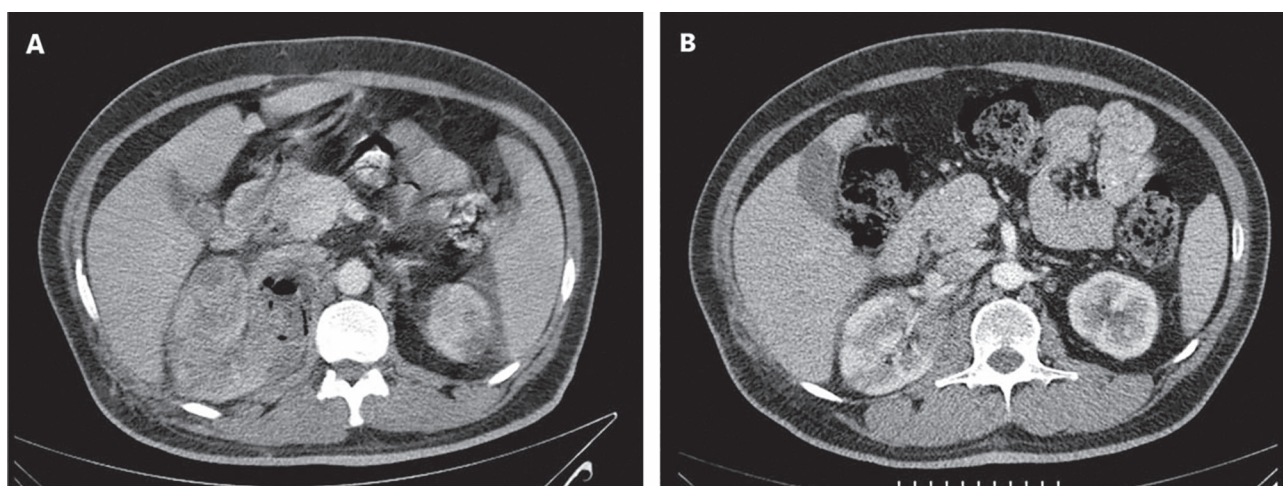
diabetes mellitus and was taking oral anti-diabetic drugs. He had not any known severe disease such as advanced malignancy. On physical examination, his vital signs were stable, including a blood pressure of 140/85 mmHg, pulse rate of 91 beats/min, temperature of 37.1 °C, and respiratory rate of 18 breaths/min. There was no evidence of thyromegaly and lymphadenopathy in head and neck examination, as well as infection or foreign body in ear. Neurologically, he was awake and no other abnormalities were found. No mass was detected in palpation of abdomen. Chest X-ray and ECG were normal. The first blood test revealed leukocyte count: 9,900/ μ L (normal range: 3,600 to 10,000 / μ L), platelet count: 122,000 / μ L (normal range: 150,000 to 400,000 / μ L), hemoglobin level: 13.4 g/dL, erythrocyte sedimentation rate (ESR): 90 and C-reactive protein (CRP) with +1 reaction. Biochemistry blood tests included blood urea nitrogen: 51 mg/dL, creatinine: 1.9 mg/dL, sodium: 130 mg/dL, potassium: 3.7 mg/dL, and HbA1C: 10. The urinary analysis had many bacteria, 8 to 10 WBC and 6 to 8 RBC, so urine culture was requested. Ultra-sonography of abdomen showed a hetroechoic finding in right kidney bridge.

Reflux esophagitis LA class B was detected in upper GI endoscopy. In order to rule out hiccup related to central nervous system (CNS), brain magnetic resonance imaging was conducted and the result was normal.

We prescribed oral pantoprazole (40 mg/d), oral chlorpromazine (25 mg t.i.d), and gabapentin

(100 mg t.i.d) to relieve hiccups. After 2 days of admission, the patient was alert but his vital signs changed, including temperature of 39.5 °C, respiratory rate: 40, pulse rate: 140, blood pressure: 140/70 mmHg. Result of urine culture showed escherichia coli. We found the following changes in second blood sample; leukocyte count: 15,200 / μ , platelet count: 100,000 / μ , hemoglobin level: 12.4 g/dL, ESR:115, CRP: +2, and serum ferritin: 1259 ng/mL. Based on the findings of kidney ultrasound and urine culture, abdominopelvic CT was requested. In order to decrease risk of contrast nephropathy, 100 mL normal saline/h and oral N acetyl cysteine (1200 mg) were administered for 12 hours pre- and post- procedure. Contrast-enhanced abdominal multi-slice CT scan (MSCT) using 64-slice scanner (Brilliance® 64; Philips Medical Systems) with administration of intravenous contrast agent (100 mL ultravist 370 mg) and oral contrast (40 mL of meglumine compound 76%) was performed and the findings were indicative of emphysematous pyelonephritis (Figure 1-A). Class 3B EPN was confirmed based on classification of CT severity in the study of Huang *et al*.⁹ Our patient had no signs or symptoms of septic shock and did not require intensive care unit admission. Blood culture and urine culture were positive for escherichia coli.

The initial management for EPN was included parenteral antibiotics based on the report of antibiogram, meropenem (1 g tid) and Amikacin (500 mg bid) for two weeks and bilateral double J stent insertion. The hiccup of the patient was stopped



A, It shows contrast enhanced CT scan before treatment (enlargement of right kidney associated with heterogeneous parenchymal density and some small intra-renal and peri-renal abscess formations and some intra-parenchymal and peri-nephric air bubbles are indicative of emphysematous pyelonephritis). **B**, It demonstrates contrast enhanced CT scan after treatment (significant decrease in renal parenchymal heterogeneity and disappearance of intra-renal and peri-renal air bubbles after treatment).

after a week. In order to control diabetes, insulin therapy was considered for the patient.

The patient was discharged with sterile culture of urine and blood, other investigations showed leukocyte count: 9,400 / μ L, platelet count: 396,000 / μ L, hemoglobin level: 13.6 g/dL. Oral Levofloxacin (750 mg/d) was prescribed for 2 weeks. The patient was followed and found well with these laboratory data, ESR: 50, CRP: +2, negative urine culture, BUN: 43, Cr: 1.39, sodium: 135, and potassium: 3.6. Double J stenting was removed after 4 weeks. According to positive acute phase reactants, oral levofloxacin was continued for 2 weeks. Acute phase reactants decreased (ESR: 4, CRP: negative, and ferritin: 255), and also urine culture was negative. Contrast-enhanced CT scan (2 months after diagnosis) depicted remnant of inflammatory tissues in right kidney (Figure 1-B). In this following period, there was no relapse of hiccup and the occurrence of neurologic symptoms. The patient was followed for one year and had no relapse.

DISCUSSION

EPN is a gas-producing, necrotizing infection involving the renal parenchyma and surrounding tissues that it is associated with high mortality and morbidity.⁸ In the majority of cases of EPN, patients with long-standing diabetes and poor glycemic control are affected. The main treatment is prompt antibiotic therapy with or without percutaneous drainage and in the absence of response, nephrectomy should be considered.¹⁰ This disease has no specific sign or symptom. However, the most common of clinical findings are fever, flank pain, dysuria, and costo-vertebral angle tenderness.⁹ We reported a man with diabetes mellitus and EPN whose first clinical manifestation was hiccup. One of the main common reasons of hiccup is gastric distension.¹¹ Our patient neither had any gastrointestinal symptom, nor used drugs that induce hiccup.¹² In terms of neurological conditions, he was stable and well.

Hiccup rarely indicates a disease. It is usually considered less important in clinical practice. However it is important in some patients as a side effect of some drugs like chemotherapy.¹²

When a patient with hiccups has no history of alcohol abuse and is not febrile, reflux esophagitis, esophageal cancer, Hodgkin's lymphoma, and

bronchogenic cancer should be considered; so we performed CXR and upper GI endoscopy. But in a patient with hiccups and fever, some other differential diagnosis should be considered such as pneumonia with pleurosis, pericarditis sub diaphragmatic abscess and peritonitis. The interesting thing about this patient with pyelonephritis is that fever developed after one week of hiccups.¹³

Based on our literature review, we found no report from occurrence of hiccup in kidney infection especially EPN. Some infections such as syphilis, pneumonia, pleurisy, myocarditis, and empyema may be caused chronic hiccup due to stimulation or damage of phrenic nerve as the afferent pathway for hiccup.¹⁴ In this report, perhaps para-renal fluid collection and peri-nephric fat stranding have led to stimulate hiccup afferent pathway and occurrence of hiccup.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

PATIENT CONSENT

The patient kindly provided written informed consent for publication.

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Correspondence to:
Muhanna Kazempour
Department of Internal Medicine, Loghman- Hakim Hospital,
Shahid Beheshti University of Medical Sciences, Kamali St.,
South Karegar Avenue, Tehran, 1333431151, Iran \

Tel: 0098 21 5541 6130
Fax: 0098 21 5541 6130
E-mail: muhannakazempour@gmail.com

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