Klebsiella Distinctive Syndrome Presenting with Muscular Abscess and Osteomyelitis: Case Report

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The present article describes a case of 46-year-old man who presented with progressive pain and swelling at the right lower back and buttock for three weeks. The investigation showed an abscess at the right lower back and buttock, osteomyelitis of the iliac bone, and a hepatic abscess. The causative pathogens were confirmed to be *Klebsiella pneumoniae*, both K1 and K2 serotype. This patient was treated with surgical management, percutaneous drainage, intravenous antibiotic, and control of his underlying disease risk factor, which was diabetic mellitus.

Keywords: *Klebsiella* distinctive syndrome, *Klebsiella* liver abscess syndrome, Hypervirulent strain of *Klebsiella* pneumoniae, *Klebsiella* pneumoniae, Liver abscess

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Klebsiella pneumoniae is one of the most common causes of community acquired and hospital acquired infection worldwide. Recently, a distinctive syndrome caused by a hypervirulent strain of K. pneumoniae has emerged and been reported in many countries. This K. pneumoniae strain is characterized with a distinct hypermucoviscosity phenotype, which has been classified as serotype K1 and K2. To date, more than 900 case have been reported. Most of the cases have been reported from Asian countries (Taiwan⁽¹⁻³⁾, South Korea^(4,5), Japan, Singapore^(6,7), Hong Kong^(8,9), and Vietnam⁽¹⁰⁾), but an increasing number of cases have also been reported from North America, Europe, South America, Australia, and Africa. Immunocompetent and immunocompromised host-alike, diabetic mellitus (DM) seems to be the risk factor for this syndrome, and it is considered that

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strict glycemic control might prevent the metastatic complications^(5,6).

For *Klebsiella* liver abscess syndrome (KLAS), an environmental exposure to fecal-oral transmission and gastrointestinal colonization are possible routes of acquisition. After developing a liver abscess, *K. pneumoniae* bacteremia can cause subsequent extrahepatic infections, such as endopthalmitis, meningitis, septic pulmonary emboli, empyema, osteomyelitis, and subcutaneous or muscular abscess.

Nowadays, the antibiotic treatment of choice is the third-generation cepharosporins (ceftriaxone, cefotaxime, or ceftazidime) with a treatment duration ranging between two and six weeks, while percutaneous drainage is also favored for use in welllocalized liver abscess and for deep abscesses of the musculo-skeletal system.

Case Report

A 46-year-old Thai man presented with progressive pain and swelling at the right side of his lower back and right buttock that persisted for three weeks. Two months previously, he was first diagnosed with type 2 DM and was prescribed an oral hypoglycemic drug for controlling his blood glucose level. At that time, his hemoglobin A1c (HbA1c) level was 11.0 mg%. Three weeks before hospital admittance, he developed progressive pain and swelling at his right lower back and buttock coinciding with developing a low-grade fever. Analgesic and



Figure 1. Photo demonstrating marked swelling and a fluctuation at the right lower back and buttock of the patient.

antipyretic drugs were prescribed by the rural hospital, but these did not relieve his symptoms. The patient visited the authors' hospital, and a physical examination showed a high-grade fever (39.5°C), with redness, warmth, marked swelling, and tenderness at his right lower back and buttock (Figure 1). Deep musculoskeletal infection and an abscess were suspected. A computed tomography (CT) scan was performed and demonstrated the presence of a gasforming abscess along the right iliopsoas and erector spinae, right iliacus muscle, measured to be about 9.2×4.7×18.9 cm in AP×W×H. Another lesion at the right gluteus maximus muscle, measured to be about 5.5×14.9×17.7 cm in AP×W×H, also suggested a gasforming abscess. A liver abscess at hepatic segment 4b was measured as 2.5×2.9×2.8 cm. In addition, osteomyelitis was found due to the gas-producing organisms at the right iliac bone (Figure 2).

The laboratory findings included a white blood cell count of 14,900 cells/cu.mm (neutrophil 86.6%, band form 5.2%), glucose 190 mg/dL, HbA1C 9.9%, erythrocyte sedimentation rate (ESR) 73 mm/hour, C-reactive protein (CRP) 21.21 mg/L, creatinine 0.65 mg/dL, liver function test with total bilirubin 0.26 mg/dL, direct bilirubin 0.14 mg/dL, aspartate transferase 10 IU/L, alanine transferase 12 IU/L, alkaline phosphatase 69 U/L, albumin 3.4 g/dL, and globulin 4.3 g/dL.

At first, the patient was empirically treated with intravenous piperacillin-tazobactam (4.5 gm) every eight hours. The operating room was set up for emergency surgical drainage of the patient's right buttock abscess and the intervention radiologist was consulted for percutaneous drainage of the right iliopsoas abscess.

Pus culture from the abscess and two specimens of blood culture grew confirmed *K. pneumoniae* infection with a sensitivity to all antibiotics, including ceftriaxone. Then, the intravenous antibiotic was changed to ceftriaxone (2 gm) daily. Infectious disease specialist and an orthopedist were consulted and both of them agreed with the plan of treatment with intravenous ceftriaxone for four weeks, and then to switch to an oral form of amoxicillin or clavulanic acid and trimethoprim or sulfamethoxazole for another eight weeks. An ophthalmologic examination was performed and showed no evidence of endophthalmitis or other abnormal ophthalmic conditions. An endocrinologist was consulted for glycemic control.

The patient's clinical status was stable six days after admission and the drains were removed 14 days postoperative. The CT scan was repeated and showed a resolution of both abscess areas (the iliopsoas and gluteus maximus muscle). In addition, the patient's blood glucose level was stable at around 100 to 130 mg/dL. The patient was discharged after four weeks of intravenous ceftriaxone (Figure 3).

Discussion

Klebsiella distinctive syndrome caused by a hypervirulent strain of *K. pneumoniae* has incidence mostly in Asia⁽¹⁻¹⁰⁾. Poor glycemic control seems to be a risk factor for this syndrome and has a major role to cause metastatic complications.

In the present report, the authors described a case of *Klebsiella* distinctive syndrome in a 46-yearold Thai man presenting with an abscess at his right lower back and buttock, osteomyelitis of the iliac bone, and a hepatic abscess. The causative pathogens were confirmed to be *K. pneumoniae*, both serotype K1 and K2. The patient was treated with surgical drainage, percutaneous drainage, and intravenous antibiotics with ceftriaxone for four weeks, then switched to an oral form of amoxicillin/clavulanic acid and trimethoprim/sulfamethoxazole for another eight weeks.

The present case might be the first reported case of *Klebsiella* distinctive syndrome in Thailand and the authors believed that the incidence of this syndrome worldwide will significantly increase due to the increase in the number of diabetic patients, which will also increase awareness of this syndrome by doctors and all medical providers.



Figure 2. CT scan whole abdomen of this patient demonstrating: A large gas-forming abscess along the right iliopsoas and erector spinae, right iliacus muscle (left, upper). Another large lesion at the right gluteus maximus muscle (right, upper). A liver abscess at hepatic segment 4b (left, lower). Osteolytic lesion due to a gas-producing organism at the right iliac bone (right lower).



Figure 3. Photo demonstrating the lower back and buttock contour after 1 month of treatment. The patient was discharged home in a stable condition and continued with an oral form of amoxicillin/clavulonic acid and trimethoprim/ sulfamethoxazole for another 8 weeks.

Conclusion

In the present report, the authors describe a case of *Klebsiella* distinctive syndrome in a 46-yearold Thai man with underlying of type 2 DM. The investigation showed an abscess at the right lower back and buttock, osteomyelitis of the iliac bone, and a hepatic abscess. The patient was treated with surgical drainage, percutaneous drainage, antibiotics, and control of his underlying disease risk factor.

What is already known on this topic?

Klebsiella distinctive syndrome or KLAS is a rare disease caused by a hypervirulent strain of *K. pneumoniae* (serotype K1 and K2). To date, more than 900 cases have been reported. Most of the cases have been reported from Asian countries. Immunocompetent and immunocompromised hostalike diabetes mellitus seems to be a risk factor for this syndrome. The environmental exposure to fecaloral transmission and gastrointestinal colonization are

the possible routes of acquisition. After developing a liver abscess, *K. pneumoniae* bacteremia can cause subsequent extrahepatic infections, such as endopthalmitis, meningitis, septic pulmonary emboli, empyema, osteomyelitis, and subcutaneous or muscular abscess.

What this study adds?

This might be the first report in Thailand about the *Klebsiella* distinctive syndrome. In this report, the authors described a case of 46-year-old Thai man presenting with an abscess at the right lower back and buttock, osteomyelitis of the iliac bone, and a hepatic abscess, which were quite uncommon presentation of this syndrome.

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Consent for publication was obtained from the patient in written form.

Conflicts of interest

The authors declare no personal or professional conflicts of interest, and no financial support from any pharmaceutical companies.

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