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Clinical report

Septicemia due to Klebsiella rhinoscleromatis: a rare case

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Background: *Klebsiella rhinoscleromatis* is the etiologic bacterium of rhinoscleroma. While rare, it has been implicated as a cause of septicemia. Only five cases of septicemia due to *K. rhinoscleromatis* have been reported previously.

Objective: Report a case of *K. rhinoscleromatis* septicemia in Thailand and literature review.

Results: K. rhinoscleromatis septicemia was observed in a 71-years-old-female diabetic patient suffering with acute fever.

Keywords: Bacterial infection, gram-negative bacteremia, Klebsiella, rhinoscleroma, rhinoscleromatis

Klebsiella rhinoscleromatis infection is relatively rare, and clinically presents with usually rhinoscleroma (RS). It is a chronic, slowly progressive granulomatous disease affecting any portion of the respiratory tract and particularly the nasal mucosa [1]. In humans, *K. rhinoscleromatis* is predominantly isolated from nasal discharge and infected tissues [1, 2], and only rarely from blood. Although *K. rhinoscleromatis* is a proven cause of rhinoscleroma, it is rarely implicated as a cause of septicemia. Only five human cases with *K. rhinoscleromatis* septicemia have been reported previously [3-7]. We report a case of septicemia due to *K. Rhinoscleromatis* in a diabetic patient in Thailand.

Case report

A 74-years-old Thai female was admitted to Hat Yai Hospital for evaluation of four-hour fever. She has a history of type 2 diabetes mellitus, hypertension, and hypercholesterolemia. She had been treated for many years with enalapril maleate, simvastatin, hydralazine, metformin, glipizide, pioglitazone and aspirin. Her glucose control was poor, hemoglobin A1C value 9.9%. At her initial physical examination, she was alert and cooperative. Her temperature was 38.5°C, pulse 110/minute, and blood pressure 100/60 mmHg. She had no clinical evidence of compromised immunity or clinical signs of localized infection. Physical examination was unremarkable. The leucocyte count was 18x10³ cells/mL with 92% segmented neutrophils, the hematocrit level was 37%, and the platelet count was 181x10³ platelets/mL. The blood glucose level was 145 mg/dL. Renal and liver function tests were within normal ranges. Urinanalysis revealed glycosuria without significant sediments. A chest radiograph was normal.

Primary bacteremia without localized infection was suspected, and one hour after admission, she was administered 2 gm of intravenous ceftriaxone. Two blood culture samples from peripheral lines were obtained using BacT/Alert FA bottles (bioMériex, Durham, USA), and after incubation for one day, both blood culture bottles grew gram-negative bacilli. Samples from both bottles were sub-cultured onto 5% (vol/vol) human blood agar, MacConkey agar, and chocolate agar at 35°C with 5% CO₂. Slow-growing mucoid colonies were recovered from the plates. Klebsiella rhinoscleromatis was then isolated. Disk diffusion tests for antibiotic susceptibility were performed on chocolate agar incubated at 37°C in 5% CO₂. The strain was susceptible to ceftriaxone, ceftazidime, gentamicin, imipenem, ertapenem, tazobactam/piperacillin, and sulfamethoxazole/ trimethoprim but resistant to ampicillin, ciprofloxacin and levofloxacin using standardized disk agar diffusion techniques. After K. rhinoscleromatis was reported,

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the patient was sent for a complete laryngoscopic upper respiratory tract examination. It revealed no abnormalities. A repeat blood culture on day 7 of hospitalization was negative. Intravenous ceftriaxone was continued until day 14 of hospitalization. These drugs proved efficacious and clinical improvement became rapidly evident. She fully recovered and was discharged from hospital 15 days after admission. She recovered fully and was discharged from hospital 15 day after admission.

Discussion

Klebsiella rhinoscleromatis is a gram-negative intracellular facultative encapsulated bacterium, often referred to as Klebsiella pneumoniae subsp. Rhinoscleromatis. It belongs to the family Enterobacteriaceae [2]. There is no clear understanding of the natural habitant of K. rhinoscleromatis, and little is known about the transmission of this organism [2]. K. rhinoscleromatis was first described in 1882 by Von Frisch. He also determined its role as the causative agent of RS [8], which is an uncommon, chronic and progressive granulomatous disease. It predominantly affects the upper respiratory tract, particularly the nasal cavity, and may spread to the tracheobronchial tree [1]. A clinical diagnosis of RS is highly nonspecific, and is based on such factors as presence of a nasal mass, chronic nasal discharge, airway obstruction, and chronic rhinitis. A specific diagnosis needs to be confirmed by bacterial culture and examination of the particular histopathological features [1, 9]. K. rhinoscleromatis usually infects the respiratory tract [1], and septicemia caused by this organism has only rarely been reported, with only five human cases previously described in the English-language literature [3-7].

Table 1 summarizes the main features of the case reports of septicemia from *K. rhinoscleromatis*, including the present case.

In case 1 [3], the patient was an elderly diabetic patient having symptoms of vomiting, rigor, and suprapubic pain. Blood cultures grew *K. rhinoscleromatis*, while a urine culture was positive for *Klebsiella pneumoniae*, *E. coli*, and *Proteus mirabilis*. Case 2 [4] was a female child presenting with purpura fulminans complicated with disseminated intravascular coagulopathy. In case 4 [6], the female patient had severe clinical features of *K. rhinoscleromatis* septicemia and died after progression to septic shock and respiratory failure. These three cases appear to be the result of community-acquired bacteremia. Two other cases of *K. rhinoscleromatis* septicemia have been reported during hospitalization, with the first case in patient suffering from tracheal obstruction caused by RS [5], and the second case in a patient who developed septicemia with unknown primary site after 14 days of hospitalization [7]. However, only one of these patients was reported to have RS, as confirmed by a positive blood culture [5]. Obtaining blood cultures in patients with RS may promote early diagnosis of septicemia due to *K. rhinoscleromatis*.

In our case, we describe the sixth case of septicemia resulting from *K. rhinoscleromatis* infection. The patient was a poorly controlled diabetic who developed acute fever. *K. rhinoscleromatis* was documented from blood cultures, and she was subsequently diagnosed with bacteremia of unknown primary site. We believe that diabetes may have played an important role in the virulence of her *K. rhinoscleromatis* infection. Interestingly, there was no evidence of RS in our case.

Clinical experience with the use of antimicrobials in the treatment of K. rhinoscleromatis infection is limited to information derived from patients with RS. These patients were sensitive to a wide range of antibiotics, including ciprofloxacin, ofloxacin, ceftriaxone, and sulfamethoxazole/trimethoprim [1, 10]. Since there is little information concerning septicemia resulting from this organism, the selection of antimicrobials should be considered in light of an agent's in vitro activity, bioavailability, toxicity, and cost. Among the cases reviewed herein, cephalosporin plus gentamicin, and the quinolone group, particularly levofloxacin and ciprofloxacin were drugs of choice. Nevertheless, two cases resulted in death. In our case, the patient was successfully treated with ceftriaxone. Early empirical treatment with antibiotics should be considered to cover common causes of bacteremia in diabetic patients such as E. coli and Klebsiella [11, 12] before antibiotic susceptibility tests become available.

In conclusion, *K. rhinoscleromatis* septicemia should be suspected in elderly diabetic patients who present septicemia even though direct evidence of rhinoscleroma has not been found. Obtaining blood cultures from patients with RS may detect early detection of bacteremia if present. Obtaining blood cultures from patients with RS may detect bacteremia of this organism early.

Table 1	. Demograph	ic and c	linical cha	racteristics of repo	rted cases of Klebsiella rhinoscleroma	<i>tis</i> septicemia in	the literature.		
Case	Country [reference]	Year	Sex/age in years	Predisposing factors	Clinical manifestations	Rhinosclero- ma (infected area)	Treatment (duration, days)	Antibiotic susceptibility	Outcome
	USA [3]	1979	M/71	DM	Vomiting, rigor, suprapubic pain	No	Ampicillin, Gentamicin (14)	NA	Survived
7	USA [4]	1989	F/35	Alcoholism	Fever, vomiting, nausea, diarrhea, cough, chest pain, septic shock, acute respiratory failure	No	Cefazolin, Gentamicin → Cefoxitin (NA)	NA	Died
3	Israel [5]	2000	M/42	Heavy smoker	Chronic nasal discharge, upper airway obstruction	Yes (Trachea)	Ciprofloxacin (14)	NA	Died
4	Nigeria [6]	2002	F/3.5	NA	Fever, vomiting, diarrhea, DIC, purpura fulminans	No	Cefuroxime, Gentamicin (15)	NA	Survived
S	USA [7]	2007	F/71	DM COPD Heart fàilure	Left sided weakness, slurred speech, hospital acquired fever	No	Levofloxacin (21)	Sensitive to sulfamethoxazole/trimethoprim, levofloxacin, cefazolin, gentamicin, and tetracycline Resistant to (NA)	Survived
This report	Thailand	2010	F/71	D M H T Hyperlipidemia	Fever	No	Ceftriaxone (14)	Sensitive to ceftriaxone, ceftazidime, gentamicin, imipenem, ertapenem, tazobactam/piperacillin, and sulfamethoxazole/trimethoprim Resistant to ampicillin, ciprofloxacin and levofloxacin	Survived

F=female, M=male, DM=diabetes mellitus, HT=hypertension, COPD=chronic obstructive pulmonary disease, DIC=disseminated intravascular coagulopathy, NA= not applicable, → indicates changing antibiotics

Vol. 4 No. 6 December 2010

Klebsiella rhinoscleromatis septicemia

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