Kiebsiella Rhinoscieromatis - an Innocent or a Deadly Organism

Abu Noem Farooqui,Aftab Merchant,Layeeq Ahmed (Department of Medicine, Karachi Medical and Dental College and Abbasi Shaheed Hospital, Karachi.)

Introduction

Kiebsiella rhinoscieromatis is the etiological agent of rhinoscleroma which apparently is the only disease associated with this gram-negative bacillus, The disease is described as a rare form of chronic granulomatous and destructive infection of the nasal passages. It is endemic in Eastern Europe and Central America but is being recognized with increasing frequency in other countries[1] including Pakistan. India and Cli ma[2]. Transmission is believed to be froni person to person in endemic regions. The incubation period is unknown.

The granulomatous inflammation is in response to the presence of Klebsiella rhinoscleromatis within the niacrophages that appear to be unable to kill the organism. This reaction leads to the formation of bulky. soft tissue masses in the respiratory mucosa having potential for local spread. The biopsy specimen of the lesion shows pathognomonic M ikulicz\'s cells (foamy h istiocytes) in the submucosa. Nasal obstruction occurs over a long period of time. caused by tumor like growth. The currently advocated regimen against the microorganism is high dose oral ciprotloxacillin 750 mg bid[3].

The disease although considered as pathology restricted to the upper respiratory tract has the potential of causing life threatening conditions. We are reporting one such case and are eliciting few other cases in which this at icroorganism was responsible for serious clinical consequences. lb the best of our knowledge the case that we have reported is the first ever reported case from Pakistan that describes Krhinoscleromatica causing septicemia.

Case Report

A 26-year-old male admitted to the hospital with complaints of fever for four days. bleeding from gums and bluish red patches over both upper arms for one day.

The patient a non—smoker, non—diabetic, non— hypertensive, driver by occupation. resident of Liaquatabad a lower middle class locality of Karachi, was well four days earlier when he developed fever with rigors. The fever was of' high grade, sudden in onset and continuous. It was associated with nausea. voni iting, myalgia and arthralgia. There were no other associated complaints including cough. upper respiratory congestion, diarrhea, or urinary symptoms. He attended the clinics of two physicians during these four days and one of them prescribed him Ofloxacin 200-mg P0 BID, which he was taking for two days prior to admission. On the day of admission he started bleeding from the gums, developed ecchymotic lesions on both of his upper arms and became drowsy. There was no other site of frank bleeding or ecchyrnosis present over his body. The patient had never experienced an episode of bleeding or unaccounted bruising in the past. There was no history of preceding viral and or respiratory tract infections. Current and recent use of aspirin containing medications, antimalarials or sulphonamides was not present. There was no hospitalization or history' of blood transfusion. Bleeding disorders in the first-degree relatives was also ruled out. And the patient had never traveled abroad in the past.

On examination the patient was febrile (106°F). Tachycardia (pulse 124/rn in, regular) and lachypnoea (resp rate was 26/mm) were present and he was hypotensive (90/60 mml Ig). Anemia, jaundice and dehydration were evident. None of the lymph nodes were palpable. Fresh blood was oozing out of the gums. There was no frank bleeding observed elsewhere on the body. The skin showed two ecchymotic lesions over the anterolateral aspects of both upper limbs. These lesions were irregular in outline and bluish red in appearance. The one on the right arni was 7.5 x 5 ems and the other was 5 x 3,5 ems in

size, The abdomen was flat and soft. None of the viscera including liver, spleen and kidneys were palpable and the gut sounds were audible. Examination of the cardiovascular, respiratory and nervous systems did not reveal any abnormality except increased heart rate, respiratory rate and sorn no lence. His initial investigations showed pancytopenia but subsequent investigations showed neutrophil ic leukoeytosis (total leukocyte count of 19,500 per cmm with 85% neutrophils) and thronihocytopenia (12,000 per cmiii).

Blucose. urea, creatin ine and A PTT were raised. Fibrin degradation product level was inconclusive. Liver function studies showed an elevated conjugated bi lirubin along with raised alkaline phosphatase and disturbed ALT. The lieiiioglobin of' this pat lent was initially near normal but later started decreasing. Bile and urobilinogen were present in the rine. Resides this urine analysis was unremarkable.

Malarial parasite was not seen on the blood film, TYphi dot assay was negative for both lgM and lgG antibodies, Chest X—ray and electrocardiogram were normal. Blood specimens were taken from two different sites and were sent for culture and sensitivity. The details of these investigations are shown in Tables 1 and 2.

Table 1. Hematologic blood values.

Variable	Value		
	Day 1	Day 4	Day 10
Hemoglobin (g/dl)	13.1	10	8.9
Hematocrit (%)	40.2	29.6	27
Total Count			
(per cmm)	1,500	19,400	10.800
Differential Count			
Neutrophils	DLC	85%	68%
Lymphocytes	not	10%	27%
Monocytes	possible	03%	04%
Basophils		02%	01%
Platelet Count			
(per cmm)	12,000	15,000	1,10,000

Table 2. Blood chemical values.

Variable	Value
Random Blood Glucose (mg/dl)	245
Liver Function Test	
Bilirubin (mg/dl)	
Total	8
Conjugated	5.8
ALT	110
Alkaline Phosphatase	675
Prothrombin Time (Control - 13 sec)	15
APTT (Control - 33 sec)	45
FDP (mg/dl)	4
Urea (mg/dl)	175
Creatinine (mg/dl)	3.5
Electrolytes	
Sodium (meq/l)	145
Potassium (meq/l)	4.2
Chloride (meq/l)	102
Bicarbonate (meq/l)	21

The patient was rehydrated and empirical therapy was started with Ceftriaxone 2gm IV qd. Amoxicillin — clavulanic acid 1.2 gm IV q8h and Metronidazole 500mg IV q8h lie was also prescribed dexamethasone 4mg IV q6h and Ranitidine 50mg IV ql2h. Despite the above e forts (lie condition of

the patient improved very slightly. The blood culture and sensitivit report received on (lie third day of admission documented Klebsiella rhinoscleromatis This iii icroorgan ism was shown to be sensitive to am ikacin, ha ipenem. piperaeil I in/tazobac. meropenem and ofloxacin/ciprofloxacin in, The antibiotic regimen was changed to ciprofloxacin 0.2(1 IV I 2h. The patient became a febrile on the fotirth day of' commencement of this treatment. By the end of the first week his total leukocyte count began decreasing while platelet count started improving B (lie end of second week all his investigations were with in the normal range.

Discussion

Klebsiella rhinoscleroment has has long been regarded as a benign organism that does not cause serious systemic effects. However. in our patient this has not been true as this gram-negative bacillus caused disseminated effects. The first ever reported case of disseminated K. rhinoscleromatis infection has been from Florida, United States of America, where in Dee. 1989 a 35-year old obese black American woman presented with nausea, vomiting, diarrhea. fever, cough. and chest pain of two weeks duration. She was pancytopenic and acidotic, with respiratory failure and hypotension. Blood cultures were positive for organisms that were reported to be K. rhinoscleromatis. A diagnosis of septic shock was made. and the patient died 48 hours after admission. At autopsy' she had massive hepatic necrosis with numerous Mikulicz's cells. The lungs, spleen, and bone marrow were also involved[4]. Although Kiebsiella rhinoscleromatis is usually associated with granu lomatous or necrotizing disease of the upper airways. it may cause varying degrees of involvement of the lower respiratory tract. A report describes seven patients in whom K. rhinoseleromatis and IC. ozaenae were recovered from sputum, blood and mixed wound infections. None of these patients had the characteristic clinical manifestations of infection with these species. However, antibiotic sensitivity patterns were unusual in these cases antI included susceptibility to both ampici Ilin and carbenicill in[5]. The currently advocated regimen against the microorganism is either h igh close oral ciprofloxacillin 750mg bid[3] or long terra (two months) treatment with streptomycin, tn met h a prim-su Ifarn eth oxazo le. a qu mo lone, or tetracycline[6]. In Patients with rhinoscleroma. experience show that the organism is diflicult to eradicate completely although seemingly sensitive to common antibiotics in vitro. A prolonged treatment with bactericidal antibiotics is therefore necessary to eradicate it[6-8].

Infection caused by K. rhinoscleromatis has been reported in patients in fected with the human immunodeticienc virus (HIV) and in patients suffering from a major cellular immune deficiency[9] Association has been reported between rhinose Ierom a caused by the bac ill tts EClebsiella rhinoscleroma and rhinosporidiosis caused by' the fungus rhinosporidium seebri[10]. These observations make one curious regarding the role of' cellular immune responses in the pathogenesis of Klebsiella rhinoscleromatis infection. The systemic infection caused by' Klebsiella rhinscleromatis. its unusual presentation in patients in fected with HIV, its association with fungal infection and the prolonged treatment requ red to eradicate th is in fèction raises the question whether in reality it is an innocent or a lethal organism. In our case the patient was found to be HIV negative. his is neutroph ilcount was with in normal range there was rio evidence of any opportunistic fungal infection and in short, there was no sign of imniunodeficiency. We conclude that although K. rhinoscleromatis is a rare cause of septicemia known until now, it can still create disseminated infection in normal healthy adult population.

Reference

1.HOlinger PH. Gelman HK, Wolfe CK. Rhinoscleroma of he lower respiratory tract. Laryagoscope, 1977: 87:1,1-9.

2.Richens J. Rhinoscleroma. In. DJ Weatherall, J.G.G. Ledingham. and DA Warrell Eds. Oxford textbook of Medicine 3rd ed. Oxford University Press, 1996,pp. 7,11,9 568-9.

3.Trautmann M. Held T, Ruhnke M, et al. A case of Rhinoscleroma cured with ciprofloxacin. Infection.

1993: 21;6, 403-6.
4.Porto R. Hevia O, Hensley GT, et al. Disseminated Klebsiella rhinoscleromatis infection. Arch. Pathol. Lab Med., 1989, 113:1381-83.
5.Berger SA. Pollock AA, Richmond AS. Isolation of Klebsiella ozaenae and Klebsiella rhinoscleromatis in a general hospital. Am, J. Clin. Pathol, 1977, 67:499-502.
6.Durand M, Joseph, M, Ann-Sullivan B. Infections of the upper respiratory tract. In: Anthony S.F, Eugene B, Kurt J. et al. eds. Harrison\'s Principles of Internal Medicine. 14th ed. New York, McGraw-Hill, 1998,pp.179-82.
7.Ssali CL. The management of rhinoscleroma. J. Laryngol. Otol., 1975; 89: 91-9.
8.Amoils CP, Shindo ML. Laryngotracheal manifestations of rhinoscleroma. Ann. Otol. Rhinol. Laryngol., 1996; 105: 336-40.
9.Paul C; Pialoux G, Dupont B, et al. Infection due to Klebsiella rhinoscleromatis in two patients infected with human immunodeficiency virus. Clin. Infect. Dis., 1993; 16:441-42.
10.Al Serhani AM, al-Qahtani AS, Arafa M. Association of rhinoscleroma with rhinosporidiosis. Rhinology, 1998; 36:43-45.