

Available online at www.sciencedirect.com

ScienceDirect





Case Report

Invasive Klebsiella rhinoscleromatis infection leading to ARDS, septic shock, and death: A rare case report *,**,**,**

Manal Merbouh, Phda,b,c,d,*, Ghizlane El Aidouni, Phda,b,c,d, Brahim Housni, Phda,b,c,d

- ^a Anesthesiology and Intensive Care Unit Department, Mohammed VI University Hospital Center, Oujda, Morocco
- ^b Faculty of Medicine and Pharmacy, Mohammed First University Oujda, Oujda, Morocco
- ^cFMP Oujda, LAMCESM, Mohammed First University Oujda, Oujda, Morocco
- ^d Anesthesiology and Intensive Care Unit Department, Faculty of Medicine and Pharmacy, Mohammed First University, Oujda, Morocco

ARTICLE INFO

Article history: Received 18 April 2023 Revised 24 April 2023 Accepted 27 April 2023

Keywords:
Rhinoscleroma
ARDS
Pneumonia
Klebsiella rhinoscleromatis
Septic shock

ABSTRACT

Rhinoscleroma is an uncommon chronic infection caused by Klebsiella rhinoscleromatis, a gram-negative bacillus that affects the upper respiratory tract. Although the nasal cavity is the most affected area, pulmonary involvement has not been previously documented. This case report describes a 61-year-old man followed for nasal rhinoscleroma who was admitted to the intensive care unit with acute respiratory distress and septic shock due to bilateral infectious bronchopneumonia. Despite immediate intubation and intravenous antibacterial treatment, the patient died 10 days later. Klebsiella rhinoscleromatis was identified in a distal pulmonary swab and blood culture. While cases of rhinoscleroma and bacteremia caused by K rhinoscleromatis infection have been documented, the case we present here is the first to report on acute respiratory distress syndrome caused by fulminant pneumonia along with septic shock.

© 2023 The Authors. Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

E-mail address: manal.mrb@gmail.com (M. Merbouh).

Abbreviations: ARDS, acute respiratory distress syndrome; CT, computed tomography; ECG, electrocardiogram; ENT, ears, nose, and throat; HMC, high mask concentration.

Acknowledgments: This article has no funding resources.

^{**} Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

^{*} Ethical approval: The article type (case report), the ethical approval was not necessary.

^{**} Registration of research studies: This is not an original research project involving human participants in an interventional or an observational study but a case report. This registration is was not required.

[†] Provenance and peer review: Not commissioned, externally peer reviewed.

^{*} Corresponding author.

Introduction

Rhinoscleroma is a rare chronic granulomatous infection that affects the upper respiratory tract, usually the nasopharynx, and is caused by gram-negative bacilli [1]. Klebsiella pneumoniae is a bacterial pathogen that causes nosocomial pneumonia and, in some cases, community-acquired pneumonia. However, pneumonia caused by Klebsiella rhinoscleromatis, a subspecies of K pneumoniae, has not been previously documented. This bacterium is commonly found in patients with rhinoscleroma. Here, we present a case report of a patient who developed acute respiratory distress syndrome (ARDS), bronchopneumonia, and septic shock caused by K rhinoscleromatis, which ultimately resulted in death [2].

Case presentation

A 61-year-old man with no significant medical history, followed for nasal cavity rhinoscleroma revealed on histological and immunohistochemical study, was admitted to the intensive care unit with acute respiratory distress and shock. He had not traveled abroad. Upon examination, he was found to be unconscious with a Glasgow Coma Scale score of 10/15, tachycardic at 145 bpm, hypotensive at 86/51 mm Hg, and polypneic at 32 cpm who desaturated at 40% at room air and 78% under high mask concentration 15 L/mn, his body temperature was 38.5°C. The pleuropulmonary examination found diffuse subcrackling rales, the ears, nose, and throat examination found ulcerative lesions of the nasal cavity and the oral cavity. Blood gas found a lactate level 3.24 with a PaO₂/FiO₂ ratio of 105.

Blood tests showed elevated levels of white blood cells to 14,600/µL (4000-10,000/m), C-reactive protein level elevated to 230 mg/L (0-5 mg/L), procalcitonin positive at 2.32 g/L (<0.5 g/L), and lymphopenia at 180/µL (1000-4000/µL), anemia at 8.4 g/dL (13-18 g/dL) with prothombin time at 51% (70%-100%), high level of D-dimers 2.76 mg/L (<0.5 mg/L), and normal renal and hepatic function. The electrocardiogram showed sinus tachycardia without any conduction or repolarization disturbances. The CT scan of the chest showed bilateral infectious bronchopneumonia, moderate bilateral pleurisy, and no signs

of pulmonary embolism (Fig. 1). The transthoracic echocardiogram was normal.

The patient was diagnosed with pneumonia caused by *Klebsiella rhinoscleromat*is, which was complicated by ARDS and septic shock. He was immediately intubated and given vasoactive drugs such as noradrenaline at a rate 0.25 mcg/kg/min. Intravenous antibacterial treatment was initiated using Triaxon 2 g/d and ciprofloxacin 200 mg twice a day. The bacterium was detected in a distal pulmonary swab after intubation and in the blood culture. However, 10 days after admission to the unit, the patient developed organ dysfunction failure and ultimately passed away before adaptation to the antibiogram.

This case report adheres to the SCARE guidelines [3].

Discussion

The Enterobacteriaceae family includes *Klebsiella*, which comprises of 3 species: *K pneumoniae*, *Klebsiella oxytoca*, and *Klebsiella granulomatis*, *K pneumoniae* is the most commonly isolated species in clinical practice and is responsible for causing infections such as pneumonia, urinary tract infections, and biliary tract infections [2].

K pneumoniae has been identified as a cause of both community-acquired and nosocomial invasive infections [4]. This bacterium has 3 subspecies, with K pneumoniae subsp. pneumoniae being the most common and responsible for causing pneumonia and urinary tract infections. The other2 subspecies (K pneumoniae subsp. ozaenae and K pneumoniae subsp. rhinoscleromatis) are responsible for upper respiratory tract infections and are typically found in patients with ozaena and rhinoscleroma [5,6].

Our case report describes a patient with nasal cavity rhinoscleroma who developed pneumonia, ARDS, and septic shock due to *K rhinoscleromatis* whose evolution was death.

This is the first documented case of a patient presenting with septic shock due to pneumonia complicated by ARDS caused by *K rhinoscleromatis*. This subspecies of *K pneumoniae* was initially observed in 1882 as an isolated bacillus in patients with rhinoscleroma [7]. Rhinoscleroma is primarily found in Latin America and Africa, with occasional cases reported in Japan. Additionally, bacteremia caused by

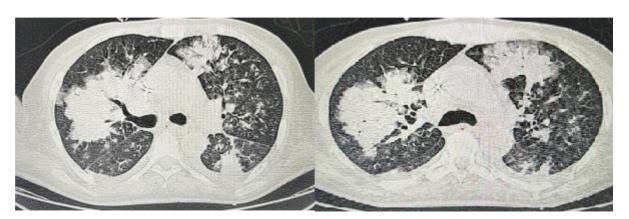


Fig. 1 - Chest CT scan on the first day of admission revealing bronchopneumonia with bilateral pleurisy.

K rhinoscleromatis infection has been reported in 6 other cases, other than upper respiratory tract infections [8,9]. Our case was seen in North Africa, Morocco.

Several treatment options have been reported in the literature for patients with *K rhinoscleromatis*, including combination therapy with ampicillin and gentamicin, combination therapy with cefuroxime and gentamicin, levofloxacin monotherapy, and ceftriaxone monotherapy [8,9]. In our case, the bacterium was found to be susceptible to beta-lactam and ciprofloxacin. Unfortunately, the patient passed away due to the fulminant stage of the disease. Their death may have been caused by the delay in diagnosis and initiation of antibiotic treatment.

Conclusion

Klebsiella pneumoniae is a bacterium that can cause both community-acquired and nosocomial pneumonia. The susceptibility of K rhinoscleromatis to antibiotics may differ between cases, often necessitating treatment with multiple antibiotics. There are documented cases of rhinoscleroma with bacteremia caused by K rhinoscleromatis infection, primarily in the respiratory tract. However, our case represents the first instance of ARDS and septic shock resulting from pneumonia caused by K rhinoscleromatis.

Patient consent

Written informed consent was obtained from the patient's sister for publication of this case report and accompanying

images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

REFERENCES

- [1] Hoffmann E. The etiology of rhinoscleroma. Int Pathol 1967;8:74–7.
- [2] Kumade E, Furusyo N, Takeshima N, Kishihara Y, Mitsumoto-Kaseida F, Etoh Y, et al. A case of lobar pneumonia and sepsis with death caused by invasive Klebsiella rhinoscleromatis infection. J Infect Chemother 2016;22(10):707–11.
- [3] Agha RA, Franchi T, Sohrabi C, Mathew Gfor the SCARE Group. The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines. Int J Surg 2020;84:226–30.
- [4] Ko WC, Paterson DL, Sagnimeni AJ, Hansen DS, Von Gottberg A, Mohapatra S, et al. Community-acquired Klebsiella pneumoniae bacteremia: global differences in clinical patterns. Emerg Infect Dis 2002;8:160e6.
- [5] Hart CA, Rao SK. Rhinoscleroma. J Med Microbiol 2000;49:395e6.
- [6] Shoeib MA. Septal rhinoscleroma. Indian J Plast Surg 2010;43:219e21.
- [7] Brisse S, Grimont F, Grimont PAD. The genus Klebsiella. The prokaryotes, 6. New York, NY: Springer; 2006. p. 159e96.
- [8] Pound MW, Fulton KB, Chima CO. Successful treatment of Klebsiella rhino-scleromatis bacteremia with levofloxacin. Pharmacotherapy 2007;27:161e3.
- [9] Berger SA, Pollock AA, Richmond AS. Isolation of Klebsiella ozaenae and Klebsiella rhinoscleromatis in a general hospital. Am J Clin Pathol 1977;67:499e502.