A rare cause of hepatic abscess in a Mexican fisherman

Una causa rara de absceso hepático en un pescador mexicano

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ABSTRACT

Melioidosis is a disease of low incidence in Latin America caused by the Gram-negative bacillus *Burkholderia pseudomallei*. This disease is endemic in Southeast Asia and Northeastern Australia. The main risk factor is diabetes mellitus. Pulmonary followed by intra-abdominal manifestations is the most common form of clinical presentation, where the liver is the most affected organ. We present a case of a Mexican male who had hepatic melioidosis secondary to skin dissemination. The microbiological diagnosis was made according to recommended standards (2017 Clinical and Laboratory Standards Institute) and corroborated by the 16S rRNA analysis. The patient received antimicrobial treatment with meropenem and trimethoprim/sulfamethoxazole, achieving a clinical, biochemical, and radiological cure.

Key words: Melioidosis. Hepatic abscess. *Burkholderia pseudomallei*. 16S rRNA.

RESUMEN

La melioidosis es una enfermedad de baja incidencia en América Latina ocasionada por el bacilo gramnegativo *Burkholderia pseudomallei*. Esta enfermedad es endémica en el sureste de Asia y noreste de Australia. El factor de riesgo mayormente asociado es la diabetes mellitus. La forma más frecuente de presentación de esta infección es la pulmonar, seguida de la intra abdominal, donde el hígado es el órgano mayormente afectado. Presentamos el caso de un hombre mexicano que tuvo melioidosis hepática secundaria a diseminación cutánea. El diagnóstico microbiológico se realizó con los estándares recomendados por el Clinical and Laboratory Standards Institute 2017 y corroborado mediante el análisis del ARN ribosomal 16s. El paciente recibió tratamiento antimicrobiano con meropenem y trimetoprima-sulfametoxazol, logrando cura clínica, bioquímica y radiológica.

Palabras clave: Melioidosis. Absceso hepático. *Burkholderia pseudomallei*. ARNr 16s.

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Date of reception: 14-02-2019

Date of acceptance: 04-02-2020

DOI: 10.24875/HMCM.20000186

Available online: 02-04-2020

Hosp Med Clin Manag. 2020;13:26-9

INTRODUCTION

Melioidosis is caused by the Gram-negative bacillus *Burkholderia pseudomallei*. The disease was first reported in 1912 by Alfred Whitmore and C.S. Krishnaswami among morphine addicts; it clinically resembled glanders, and one patient was documented with presenting multiple abscesses at morphine injection sites and lobar pneumonia. Initially, *B. pseudomallei* was classified in the *Pseudomonas* genus; however, due to its unique features, such as survival capacity and biochemical characteristics, in 1992, it was reclassified in the *Burkholderia* genus¹.

This saprophytic microorganism is found in freshwater surfaces and soil, mainly in regions with tropical climates, being endemic in Southwest Asia and North Australia. The contact with soil-contaminated surfaces increases the risk of acquiring this disease, such as among rice harvesters in Oriental countries². Diabetes mellitus is the most important non-occupational risk factor for developing the disease³, and the increasing number of people living with diabetes mellitus will probably raise the number of reported melioidosis cases in the near future.

The disease occurs after direct contact with a contaminated source, especially through soil dust inhalation, ingested water, or through a skin wound. Pulmonary melioidosis is the most common form of presentation, following contaminated dust inhalation, which may produce mild bronchitis to severe pneumonia. This is the reason why *B. pseudomallei* has been listed as a potential bioterrorism agent by the US Centers of Disease Control and Prevention, due to its high pathogenicity. Melioidosis may affect any organ, including spleen, skeletal muscle, bones, and joints⁴. A key component of *B. pseudomallei* pathogenesis, which differentiates this bacterium from other microorganisms, is its survival ability and latency capability in the human body⁵.

Eleven autochthonous cases of human melioidosis have been documented in Mexico, although in none of them, the accurate biochemical or molecular protocol to diagnose *B. pseudomallei* infection was followed^{6,7}. We present the first case of *B. pseudomallei* infection diagnosed following the appropriate diagnostic protocol. It is important to stress that this case occurred in a non-endemic area, where clinical suspicion is low.

CASE PRESENTATION

We present a case of melioidosis in a 59-year-old fisherman of the Mexican southwest, with uncontrolled type 2 diabetes mellitus (glycated hemoglobin 8.4%), who had a traumatic injury in one leg while fishing on the sea. The skin wound evolved into a fistula which resolved spontaneously in the following weeks. Four months later, he started with persistent fever and was evaluated by a physician, although no potential cause could be identified. On hospital arrival, 6 months after the initial injury, physical examination was remarkable for a heart rate of 110, temperature 38.8°C, hepatodynia, and hepatomegaly. Blood test results showed white blood cells 10.3×10^3 cells/mm³, gamma-glutamyl transpeptidase 158 IU/L (9-48 units/L), C-reactive protein 3.9 mg/dL (0-1 mg/dL), and serology for HIV was non-reactive. Contrast computed tomography (CT) evidenced multiple hypodense images with a "honeycomb" pattern (Fig. 1a-c). Then, a CT-guided drainage of a hepatic abscess was performed.

The liver abscess sample was cultured in 5% sheep blood agar and MacConkey agar, growing purple and dry colonies of variable shapes and irregular borders (Fig. 2). The Gram stain showed "hook pin"-shaped Gram-negative bacilli (Fig. 3). Biochemical tests recommended by the Clinical and Laboratory Standards Institute (CLSI, 11th ed.) were performed. The bacteria displayed cytochrome oxidase activity, reduced nitrates to molecular nitrogen, and grew at 42°C. Sanger sequencing was used to determine the sequence of 16S rRNA gene and compared it with the GenBank database through BLAST program (GenBank accession number AB626115.1). The strain was ultimately identified as *B. pseudomallei*. Broth microdilution susceptibility was performed for ceftazidime, imipenem, trimethoprim/sulfamethoxazole (TMP/SMX), and doxycycline, with the isolate being susceptible to all of them. The patient received meropenem (at a dose of 1 g every 8 h) plus TMP/SMX IV (15 mg/kg of TMP per day divided in 3 doses) for 21 days for the initial intensive treatment; after hospital discharge, he continued with TMP/SMX PO. After completing 6 months of treatment, the patient was asymptomatic and the CT scan revealed complete resolution of the hepatic lesions.

CONCLUSIONS

Melioidosis is a rare disease in Mexico, with a probably underestimated magnitude due to a low suspicion index in a non-endemic region, and to an error of up to 35%

Figures 1. Tomography of the abdomen in coronal (A), sagittal (B), and axial (C) planes with contrast medium in venous phase, where multiple rounded hypodense lesions with a honeycomb pattern are documented in segments V and VI, displaying a subtle peripheral reinforcement.

Figure 2. MacConkey agar showing the growth of rough, raised, pinkish, non-fermenting lactose colonies.

Figure 3. Peripheral blood Gram stain with short Gram-negative bacilli, with sharp points that mimic a "hook pin."

of the semi-automated methods for the identification of *B. pseudomalle*⁸. To the best of our knowledge, this is the only documented case which complied with the biochemical diagnosis recommendations by CLSI and corroborated with the molecular analysis of 16S rRNA. Among the 11 reported cases of melioidosis acquired in Mexico, the most frequent host risk factor for the disease was type 2 diabetes mellitus (3 of 11 cases); other factors were alcoholism in one patient and immunosuppressive treatment in a kidney transplant recipient^{6,7,9-12}.

The most frequent epidemiological risk factor is visiting a tropical area by the sea, many times also associated with natural disasters such as hurricanes^{10,12,13}. Melioidosis mortality rate is high, between 20% and 60%, despite appropriate treatment. In Mexico, from the 11 previously reported patients, 3 (27%) died.

No resistance to the first-line recommended antimicrobials against this disease has been documented in Mexico, consistent with international reports, where more

than 90% of the isolates were susceptible to meropenem, TMP/SMX, and ceftazidime^{14,15}. The established treatment in our case was based on international consensus, with an initial intensive therapy period of 21 days and an eradication therapy phase of 6 months¹⁶, achieving the patient's cure. We consider that in non-endemic areas such as Mexico, increasing awareness and greater surveillance are needed to identify melioidosis cases.

CONFLICTS OF INTEREST

The authors declare no conflict of interest.

ETHICAL DISCLOSURES

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this study.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

REFERENCES

- Whitmore A, Krishnaswami C. An account of the discovery of a hitherto undescribed infective disease occurring among the population of Rangoon. *Indian Med Gaz.* 1912;47:262-7.
- Limmathurotsakul D, Wongrattanacheewin S, Teerawattanasook N, Wongsuvan G, Chaisuksant S, Chetchotisakd P, et al. Increasing incidence of human melioidosis in northeast Thailand. *Am J Trop Med Hyg.* 2010;82:1113-7.
- Chaowagul W, White NJ, Dance DA, Wattanagoon Y, Naigowit P, Davis TM, et al. Melioidosis: a major cause of community acquired septicemia in northeastern Thailand. *J Infect Dis.* 1989;159: 890-9.
- Lim KS, Chong VH. Radiological manifestations of melioidosis. *Clin Radiol.* 2010;65:66-72.
- Cheng AC, Currie BJ. Melioidosis: epidemiology, pathophysiology, and management. *Clin Microbiol Rev.* 2005;18:383-416.
- Wiersinga WJ, Currie BJ, Peacock SJ. Melioidosis. *N Engl J Med.* 2012;367:1035-44.
- Martínez-Hernández L, González-Hijar A, Valdéz-Vázquez R, García-López S, González-Chon O. Melioidosis: reporte de caso y revisión de la literatura. *Neumol Cir Torax.* 2013;72:291-8.
- Boroel-Cervantes C, Ibarra-Valdez M, Miranda-Pacheco S, Sánchez-Camarena E. Melioidosis: reporte del primer caso en el estado de Sonora, México. *Rev Med Inst Mex Seguro Soc.* 2017;55:257-9.
- Podin Y, Kaestli M, McMahon N, Hennessy J, Ngian HU, Wong JS, et al. Reliability of automated biochemical identification of *Burkholderia pseudomallei* is regionally dependent. *J Clin Microbiol.* 2013;51:3076-8.
- Barnes PF, Appleman MD, Cosgrove MM. A case of melioidosis originating in North America. *Am Rev Respir Dis.* 1986;134:170-1.
- Truong KK, Moghaddam S, Al Saghibini S, Saatian B. Case of a lung mass due to melioidosis in Mexico. *Am J Case Rep.* 2015;16:272-5.
- McCormick JB, Weaver RE, Hayes PS, Boyce JM, Feldman RA. Wound infection by an indigenous *Pseudomonas pseudomallei*-like organism isolated from the soil: case report and epidemiologic study. *J Infect Dis.* 1977;135:103-7.
- Cheng JW, Hayden MK, Singh K, Heimler I, Gee JE, Proia L, Sha BE. *Burkholderia pseudomallei* infection in US traveler returning from Mexico, 2014. *Emerg Infect Dis.* 2015;21:1884-5.
- Dutta S, Haq S, Hasan MR, Haq JA. Antimicrobial susceptibility pattern of clinical isolates of *Burkholderia pseudomallei* in Bangladesh. *BMC Res Notes.* 2017;10:299.
- Hassan MR, Vijayalakshmi N, Pani SP, Peng NP, Mehenderkar R, Voralu K, et al. Antimicrobial susceptibility patterns of *Burkholderia pseudomallei* among melioidosis cases in Kedah, Malaysia. *Southeast Asian J Trop Med Public Health.* 2014;45:680-8.
- Currie B. Melioidosis: the 2014 revised RDH guideline. *North Territ Dis Control Bull.* 2014;21:4-8.