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Case of a Lung Mass due to Melioidosis in Mexico

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

ABEF 1 Kimberly K. Truong
BEF 2 Samer Moghaddam
BEF 3 Samer Al Saghbini
AEG 2 Bahman Saatian

1 Department of Medicine, University of California Irvine, UC Irvine Medical Center, Orange, CA, U.S.A.
2 Department of Medicine, Division of Pulmonary and Critical Care, University of California Irvine, UC Irvine Medical Center, Orange, CA, U.S.A.
3 Department of Medicine, Division of Infectious Disease, University of California Irvine, UC Irvine Medical Center, Orange, CA, U.S.A.

Corresponding Author: Bahman Saatian, e-mail: bsaatian@uci.edu
Conflict of interest: None declared

Patient: Female, 70
Final Diagnosis: Melioidosis
Symptoms: Chills • fever • neck pain • night sweats
Medication: —
Clinical Procedure: Incision and drainage • endobronchial ultrasound guided biopsy
Specialty: Infectious Diseases

Objective: Rare disease

Background: Melioidosis, an infection caused by the gram-negative bacterium *Burkholderia pseudomallei*, is an important cause of pneumonia, skin infection, sepsis, and death in Southeast Asia and Australia, but is exceedingly rare in North America. Pulmonary melioidosis typically presents as acute bacterial pneumonia or cavitary lung lesions resembling tuberculosis.

Case Report: We report melioidosis in a 70-year-old active smoker from Mexico with no history of travel to disease-endemic areas. The patient presented with a left supraclavicular abscess and a non-cavitary, left lung mass encasing a pulmonary vein. Incision and drainage of the patient's subcutaneous abscess isolated *B. pseudomallei*, and fine-needle aspiration of enlarged mediastinal lymph nodes revealed the presence of intracellular gram-negative bacilli with no evidence of malignancy. Biochemical tests determined that the strain the patient acquired from Mexico is identical to only 1 other isolate from Thailand.

Conclusions: This report highlights the blurring epidemiological borders of this organism, its rare presentation mimicking lung malignancy, and an aggressive antimicrobial treatment that resulted in resolution of the patient's symptoms.

MeSH Keywords: *Burkholderia pseudomallei* • Melioidosis • Non-Cavitary Lung Mass

Full-text PDF: http://www.amjcaserep.com/abstract/index/idArt/893182

Background

Melioidosis is a disease caused by the environmental gram-negative bacterium *Burkholderia pseudomallei* [1,2]. It is an important cause of localized skin infection, community-acquired pneumonia, sepsis, and mortality in Southeast Asia and northern Australia, but is extremely rare in North America. In northeastern Thailand, melioidosis accounts for 20% of community-acquired sepsis, with a case-mortality rate of 30–50% [3,4]. The incidence rate of melioidosis in northern Australia is 19.6 cases per 100,000 population per year [5]. In contrast, the U.S. has, on average, zero to five cases reported annually in people with a history of travel or emigration from endemic areas [6]. Pulmonary melioidosis typically presents as acute bacterial pneumonia or cavitary lung lesions mimicking tuberculosis [7]. We report a rare manifestation of pulmonary melioidosis in a healthy individual from Mexico without exposure to disease-endemic areas.

Case Report

A 70-year-old healthy woman visiting from Mexico presented with fevers, chills, an enlarging left neck mass, and 15-lb weight loss for 3 weeks. The patient denied cough, hemoptysis, dyspnea, night sweats, prior tuberculosis exposure, or travel outside of Mexico and the United States. She has a 7.5 pack/year smoking history. She lives in an adobe mud house and is in daily contact with environmental dirt and water to make mud stoves.

On admission, she was afebrile with blood pressure 122/77 mmHg, heart rate 66 beats per minute, respiratory rate 16 breaths per minute, and oxygen saturation 95% on room air at rest. On exam, there was an erythematous, tender left supraclavicular mass measuring 3.5×4.5×3.5 cm (Figure 1). WBC count was 8800 cell/mm^3 with 69% neutrophils; bacterial and fungal blood cultures and fungal serology, including Coccidioides, *Cryptococcus neoformans*, and Histoplasma serologies, were negative. QuantiFERON®-Tuberculosis Gold test and HIV test results were negative. Chest x-ray revealed a left suprahilar opacity concerning for lung mass, and subsequent chest computed tomography (CT) revealed a 2.5×3.0 cm noncavitary left suprahilar mass extending into the aortopulmonary window and encasing a left superior pulmonary vein with prominent regional lymphadenopathy (Figure 2). The supraclavicular mass was drained, and subculture led to an identification of *Burkholderia pseudomallei* by automated VITEK 2 (bioMérieux; Durham, NC) instrument and culture (Figure 3). Cytology

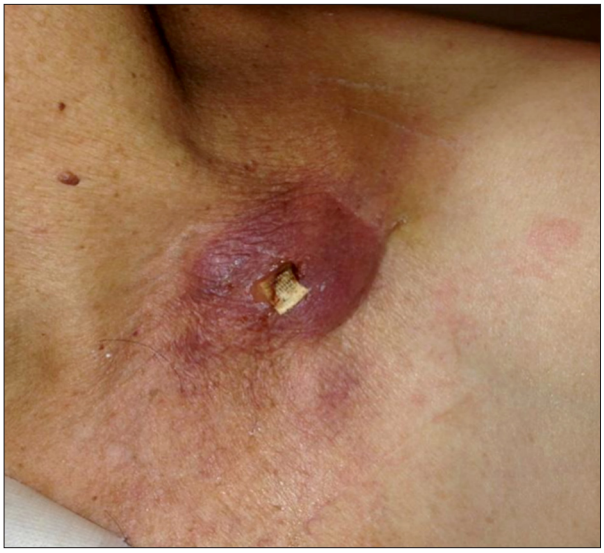

Figure 1. Left supraclavicular mass status post-incision and drainage.

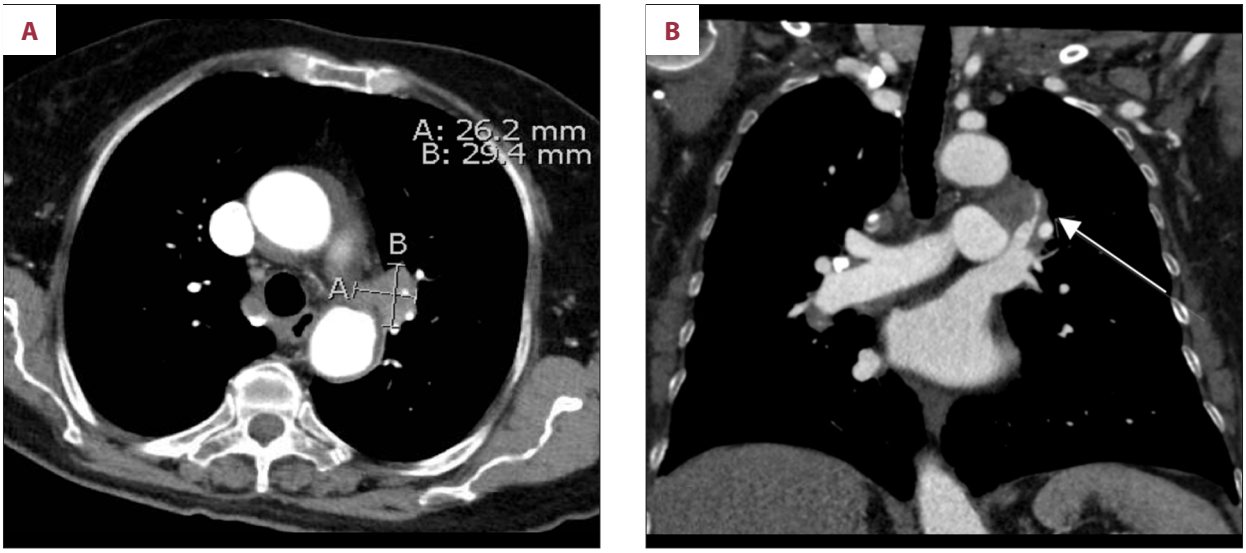

Figure 2. CT of chest with intravenous contrast in (**A**) axial view and (**B**) coronal view showing 2.6 cm by 2.9 cm hilar mass.

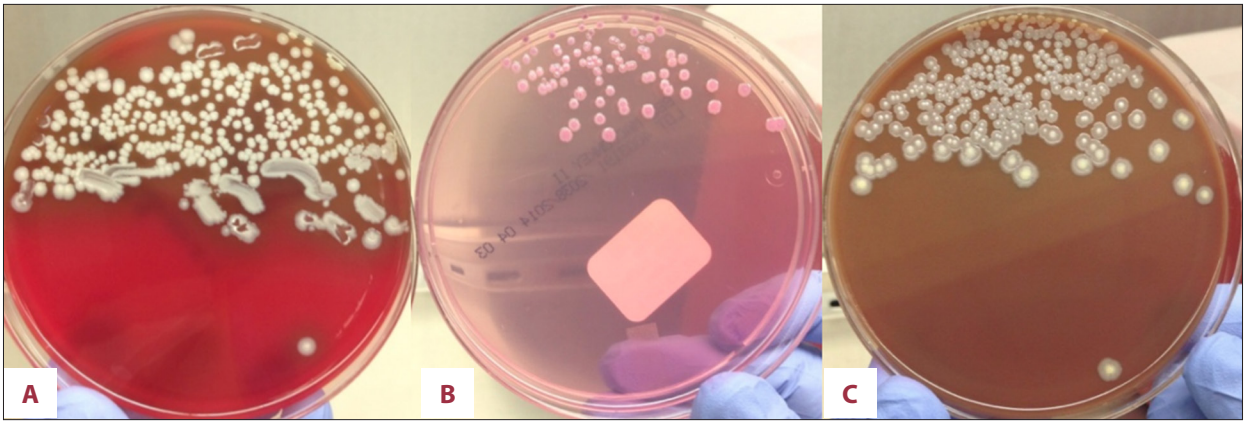

Figure 3. *Burkholderia pseudomallei* colonies growing on (**A**) sheep blood agar, (**B**) MacConkey agar, and (**C**) chocolate agar.

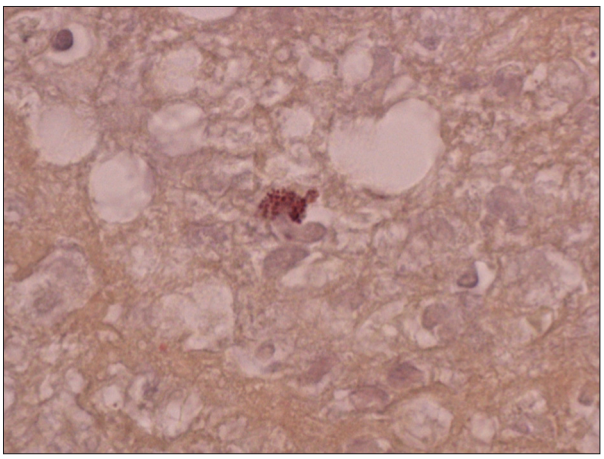

Figure 4. Brown-Hopps stain of lymph node needle aspiration showing intracellular gram-negative organism.

of endobronchial ultrasound-guided (EBUS) transbronchial needle aspiration of mediastinal lymph nodes showed mixed lymphoid population with no malignant cells. Lymph node aspirates did not grow *B. pseudomallei,* likely due to antibiotic use and inadequate sampling. However, Brown-Hopps staining of the lymph node aspirates were positive for rare intracellular gram-negative bacilli (Figure 4) supportive of *B. pseudomallei*.

The patient was started on intravenous meropenem for 14 days followed by oral trimethoprim/sulfamethoxazole (TMP/SMX) and doxycycline for six months. At the 2-week follow-up appointment, the patient reported resolved symptoms and decrease in the size of the supraclavicular mass. The patient was lost to follow-up after returning to Mexico 1 month later, and a follow-up chest CT to assess resolution of lung mass was impossible to obtain.

The original isolate was forwarded to the Orange County Public Health Laboratory and the Centers for Disease Control and Prevention for molecular subtyping using internal transcriber spacer and multilocus sequencing. The internal transcriber spacer sequence type G is consistent with the organism having a Western Hemisphere origin, while the multilocus sequence (ST951) is identical to only one other isolate from Thailand (strain 1133a). The ST951 strain is a single locus variant with associations to Puerto Rico, Martinique, Kenya, Papua New Guinea, Cambodia, and Vietnam.

Discussion

Melioidosis is a rare disease in North America. Symptomatic infection is associated with type 2 diabetes, alcoholism, chronic lung disease, renal disease, and liver disease, which this patient did not have [7,8]. Pneumonia is the most common presentation of melioidosis and is involved in approximately half of all cases. Acute pulmonary melioidosis often presents as an acute bacterial pneumonia highly associated with sepsis and death. Subacute or chronic pulmonary melioidosis typically presents as a cavitary lung lesion resembling pulmonary tuberculosis, with concurrent subcutaneous and visceral organ abscesses. Given the patient's subcutaneous abscess and lung involvement, her presentation is most compatible with subacute or chronic melioidosis.

B. pseudomallei can be transmitted by direct inoculation, ingestion, and inhalation. The size of the inoculation of microorganism is responsible for disease pattern and disease severity [8]. This patient most likely acquired her infection in Mexico, as she was at an increased risk from daily exposure to local soil and ground water. Currently this is the second confirmed reported case of melioidosis originating from Mexico [1,2].

A striking feature about this case is the pulmonary infection presenting as an encasing, non-cavitary, hilar lung mass radiographically mimicking lung carcinoma. Radiographically, the most prominent findings of pulmonary melioidosis are localized patchy alveolar infiltrates (37.5%), fibroreticular infiltrate (15.3%), pulmonary nodule (8.3%), and lung abscess

(6.9%) [9]. Only 1 other case in Thailand reported pulmonary melioidosis presenting as lung mass mimicking lung cancer [10]. This patient underwent a protracted diagnostic work-up due to the atypical character of the lesion, including a complete left pneumonectomy, before confirmation of *B pseudomallei* in pleural fluid.

B. pseudomallei exhibits intrinsic resistance to various antibiotics, including penicillins, third-generation cephalosporins, quinolones, macrolides, aminoglycosides, and rifampins [8]. Current literature suggests 2 phases of antibiotic regimen: intensive-phase antibiotic regimen and eradication-phase (also known as maintenance phase) antibiotic regimen. During the intensive phase, ceftazidime (50 mg/kg up to 2g IV every 6 hours) or meropenem (25 mg/kg up to 1 g IV every 8 hours) is suggested. During the eradication phase, trimethoprim/sulfamethoxazole (TMP-SMX) at 250/1200 mg every 12 hours for at least 3 to 6 months is recommended. Doxycycline has been used in conjunction with TMP-SMX for eradication therapy. This patient received 14 days of intravenous meropenem followed by oral TMP/SMX and doxycycline for 6 months with subsequent resolution of symptoms at 2-week follow-up.

Conclusions

As geographic boundaries are becoming less clear, it is important for physicians to maintain clinical suspicion for melioidosis in patients with underlying risk factors and travel history to endemic areas. Early diagnosis and appropriate antibiotic treatment can prevent progression of the disease and reduce mortality rate.

Acknowledgement

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Conflict of interest

Authors reported no conflict of interest.

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