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A CASE REPORT FROM SOUTH OF INDIA DISCUSSES HEMOPHAGOCYTIC LYMPHOHISTIOCYTOSIS IN MELIOIDOSIS CAUSED BY BURKHOLDERIA PSEUDOMALLEI

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ABSTRACT

Burkholderia pseudomallei, a facultative intracellular Gram-negative, saprophytic bacterium frequently found in contaminated soil or water, is the infectious illness that causes melioidosis. South-East Asia, South Asia, Northern Australia, and China are the regions where it is most common. People who work in manual labour, land surveying, fishing, building construction, gardening, and immunocompromised states like diabetes, alcoholism, chronic renal failure, chronic lung disease, and HIV/AIDS are among the populations who are at risk of occupational exposure to wet soil or surface water. We are aware of only few cases of hemophagocytic lymphohistiocytosis caused by melioidosis that have been documented in India.

INTRODUCTION

Melioidosis is a severe systemic infectious disease caused by Burkholderia pseudomallei, a motile gram-negative bacillus with bipolar staining. In endemic places, B pseudomallei, an environmental saprophyte, is frequently found in the soil, groundwater, rice paddies, and ponds. Percutaneous injection, aerosol inhalation, or the consumption of tainted water or food all contribute to the disease's transmission through contact with infected soil or water. Diabetes, Tuberculosis, Pre-term Birth, Chronic Liver Disease, Chronic Renal Disease, Splenectomy, Marasmus, Kwashiorkor, Hypertension, Pesticide Poisoning, Goiter, and Alcoholism are specific risk factors for the development of severe melioidosis.

Subclinical infections, asymptomatic or mild localised abscesses, severe pneumonia, and fulminant sepsis are examples of clinical syndromes. Hemophagocytic lymphohistiocytosis is a rare, even in endemic regions of melioidosis. We present a case of Hemophagocytic lymphohistiocytosis in melioidosis due to Burkholderia pseudomallei.

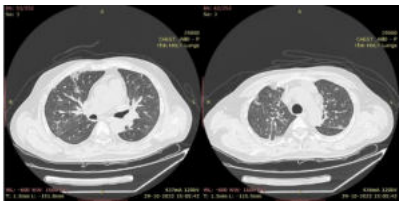
CASE DISCUSSION

A 57-year-old man with type 2 diabetes mellitus and known systemic hypertension presented with complaints of fever lasting seven days, which were followed by dyspnea, generalised body pain, headaches, and dysuria. During general examination, it was found that the patient had a blood pressure of 140/90 mmHg, pulse of 86 beats per minute, respiratiry rate of 17 beats per minute, and a saturation level of 99% on room air. Respiratory examination revealed bilateral basal crepitations and reduced air entry. Organomegaly was absent, and the abdomen was soft with left hypochondriac discomfort. An x-ray of the chest revealed both lung fields to have modest ground glassing and patchy areas of consolidation. A hypoechoeic lesion and moderate splenomegaly were seen on abdominal ultrasound. He exhibited elevated levels of pro-calcitonin (2.85 ng/ml), C-reactive protein (102 mg/l), and ESR (70 mm/hour). The neutrophil leukocytosis in the whole blood count was 11,330/1. Methicillin-resistant, coagulase-negative Staphylococcus aureus (MRCONS) was grown in blood cultures, and candida non-albicans was grown in urine cultures. Clindamycin and Fluconazole were administered intravenously to the patient in accordance with their pattern of antibiotic sensitivity. Despite taking antibiotics and receiving chest physiotherapy, the symptoms and fever remained. Multiple lung nodules, few significant mediastinal lymph nodes, fibro bronchiectatic alterations, and a minor bilateral pleural effusion were all seen on the CT scan of the chest (picture 1). Splenomegaly with poorly enhancing lesions and necrotic adenopathy were seen on the contrast-enhanced CT abdomen (picture 2). We started using empirical antitubercular medications because we suspected widespread tuberculosis. Both the Mantoux test and sputum for acid fast bacilli came back negative. Negative results from sputum culture. Thus, we stopped using anti-tubercular medications.

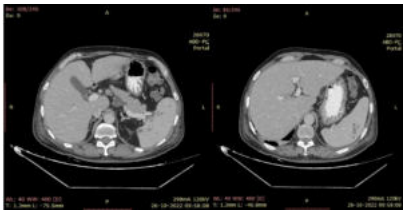
The patient's condition had gotten worse, and inflammatory indicators remained high. At this point, his clinical profile suggested an unusual infection. Retrovirus testing came up negative. Hbsag and anti-HCV tests came out negative. MSAT, Dengue IgM, and IgM Scrub all tested negative. No growth was found in the splenic lesion after a CT-guided aspiration. Due to the possibility of infectious endocarditis, echocardiography was performed, but the results were negative. Three sites of blood were cultured due to the sepsis that was not getting better. On the fifth day, the culture revealed a gram-negative bacillus that was oxidase negative and had an unique earthy odour; this bacillus was eventually identified as Burkholderia pseudomallei(picture 3).

We started the patient on oral cotrimaxazole and intravenous ceftazidime. The patient's condition became better the following week, and the inflammatory indicators returned to normal. During the second week of treatment, the patient's fever spiked unexpectedly and his leukocyte count and thrombocytopenia levels remained abnormal. Triglycerides were determined to be 281 mg/dl, fibrinogen was 1.17 mg/L, CK was 3890 U/L, and serum ferritin was 4170 mcg/L. An aggressive massive histiocytosis encompassing the erythroid and neutrophilic precursor was discovered during a scheduled bone marrow biopsy, pointing to the diagnosis of hemophagocytic lymphohistiocytosis. We started on IV immunoglobulin infusion. But sadly, the patient went into septic shock and passed away.

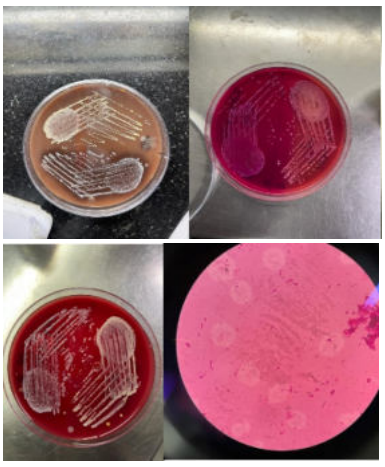
(Multiple lung nodules, free significant mediastinal lymph nodes) Picture 1



(Splenomegaly with poorly enhancing lesions and necrotic adenopathy were seen on the contrast-enhanced CT abdomen) Picture 2



CHOCOLATE AGAR, MAcCONKEY AGAR, BLOOD AGAR, GRAM STAIN Picture 3



DISCUSSION

The primary method of infection transmission is known to be direct inoculation, particularly through skin breaches. Aside from sexual transmission and person-to-person contact, additional prevalent mechanisms of transmission include inhalation, ingestion, and vertical transmission during childbirth (1)(2). Despite treatment, it has a high death rate and is known to lead to reinfection and recurrences. Acute infection (lasting less than two months) and chronic infections are the two main manifestations of melioidosis (more than 2 months). Septicemia, localised infection with or without septicemia, asymptomatic infections, ulcers, pneumonia, visceral abscesses, neurologic infection, and musculoskeletal infections are some of the possible symptoms of melioidosis. Our patient had bilateral lung consolidation, splenic abscess, and bacteremia.

When individuals appear with many abscesses in the right geographic area with a possible exposure history, there should be a strong suspicion that they have melioidosis.When possible, USG/CT scan guided incision and drainage should be used to treat deep abscesses. Blood, pus, wound swabs, urine, and sputum specimens can all be used to isolate the bacterium. Burkholderia pseudomallei should be carefully checked for using the following criteria: colony shape, Gram stain appearance, antibiotic susceptibility pattern, melioidosis antibody level, and PCR. Due to lack of acquaintance with the organism, ignorance of similarities between it and other Gram negative bacteria, and lack of microbiological experience, underdiagnosis has been recorded in numerous cases. Patients with rheumatoid arthritis, lymphadenitis, spondylitis, pneumonia, and visceral abscesses who were later diagnosed with melioidosis received presumptive treatment with antituberculosis medications, especially in tuberculosis endemic areas(3)(4) . There should be a strong suspicion of meliodiosis whenever a patient exhibits symptoms including fever, malaise, abscess, or septicemia. We are aware of very few cases of melioidosis being reported globally.

Melioidosis treatment is divided into two phases: the intense phase and the eradication phase. These stress the need for prompt septicemia treatment, the elimination of the illness, and relapse prevention. The kind, severity, and antimicrobial susceptibility of the illness determine the agent combinations utilised, the length of therapy, and whether adjunct modalities are required. The intensive phase includes of 10 to 14 days (which may be increased to 4 weeks, if clinically required) of intravenous antibiotics, such as intravenous carbapenem (meropenem/imipenem) or intravenous ceftazidime. Co-trimoxazole, doxycycline, or amoxicillin-clavulanate should be administered early during the intensive phase for tissue penetration if there are fluid collections (including skin abscess/septic arthritis), bone, or central nervous system involvement.

After then, the eradication phase should last for a further three to six months while using oral co-trimoxazole (Trimethoprim-sulfamethoxazole), either by itself or in conjunction with oral doxycycline/oral amoxycilin-clavulanate. Significant case deaths, recurrences, and reinfections are brought on by melioidosis.

Since our patient exhibited hemophagocytic histiocytosis of melioidosis along with a splenic abscess and bacteremia, we are reporting this case (5). We want to underline the significance of having a high level of suspicion when diagnosing melioidosis as well as the necessity of having sufficient microbiological knowledge and expertise to appropriately identify the organism. Steroids, cyclosporin, and etoposide injection is part of the standard treatment for HLH. When the bacterial etiology of HLH is discovered, steroids are given in addition to the antimicrobial drug (6).

CONCLUSION

Melioidosis-related hemophagocytic histiocytosis is a very uncommon disorder. We report this case because our patient having visceral signs of melioidosis, including splenic abscesses, hemophagocytic histiocytosis, and bacteremia. We want to emphasise the significance of having a high level of suspicion when diagnosing hemophagocytic histiocytosis in melioidosis, as well as the requirement for adequate microbiological knowledge and competence in correctly identifying the organism, when the patient's condition is not improving.

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