Disseminated Melioidosis Presenting as Septic Arthritis

Anjali Rajadhyaksha*, Archana Sonawale**, Shruti Khare***, Chetan Kalal****, Rahul Jankar*****

Abstract
Melioidosis is an infection caused by Burkholderia pseudomallei. The disease is known as a remarkable imitator due to the wide and variable clinical spectrum of its manifestations. Septic arthritis is rare but well-recognized manifestation of this disease.

We report a case of melioidosis in a 52 year male with uncontrolled diabetes mellitus (DM) presenting with a rare combination of septic arthritis and abscesses in the chest wall, liver and subcutaneous tissue. The patient responded to prolonged treatment of intravenous ceftazidime followed by oral co-trimoxazole.

Case Report

Introduction
Melioidosis is a disease of public health importance that is associated with high case-fatality rates in humans. It is endemic in tropical Australia and Southeast Asia with highest number of reported cases from Thailand. Melioidosis is increasingly being recognized as an important cause of life-threatening infections in India. It is underdiagnosed and underreported in India and there is increased need of creating awareness in clinicians, microbiologists and public health professionals in diagnosing melioidosis.1,2

Admission 1
A 52-year-old male, resident of Ratnagiri, was admitted to our institute one year back. Patient was a known case of type II DM on irregular treatment and was apparently well till 1 month back when he developed high grade fever. He had severe pain and swelling in the left ankle joint. Patient was febrile and haemodynamically stable. There was severe tenderness of the left ankle with warmth, erythema, effusion and limitation of active and passive range of motion due to pain and effusion.

Laboratory studies on admission revealed a leukocyte count of 11600/μL with 60% neutrophils, haemoglobin level of 7.6 gm%, platelet count of 2 lakhs/μL and ESR (erythrocyte sedimentation rate) of 30 mm/hr. Fasting and post lunch sugars were 160mg% and 286mg% respectively. Renal and liver function tests, chest X-ray were normal. USG (ultrasonography) of right knee showed loculated collection with thick internal echoes communicating with the joint cavity. USG of lateral aspect of left thigh showed 35ml collection in subcutaneous plane. To look for disseminated infection, USG abdomen was done which showed a collection of 35ml in segment VIII of liver suggestive of abscess. Computed tomography of chest and abdomen revealed hepatic abscess as mentioned above and cold abscess along left lower rib with minimal pleural reaction. The possibility of recurrent disseminated melioidosis as a result of inadequate treatment due to default by patient was considered. Patient was started on intravenous ceftazidime 2gm 8 hourly. Sugar was controlled with 30/70 insulin twice a day subcutaneously.

The aspiration of fluid from knee and hepatic abscess showed purulent material with polymorphonuclear predominance. Culture of the material from both the sites had growth of Burkholderia pseudomallei which was susceptible to piperacillin, ceftazidime, cefoperazone-sulbactam, imipenem, meropenem, cotrimoxazole, chloramphenicol and ciprofloxacin. Patient was given ceftazidime for 4 weeks and was started on oral trimethoprim-sulphamethoxazole after 15 days of discharge on his own and took insulin irregularly.

Admission 2
After 8 months of discharge from first admission
Patient presented with fever without chills for 1 month and right knee pain and swelling. He was febrile with tender, hot swelling of left knee with restricted painful movements. A small tender swelling of around 2X2 cm was noticed on the left thigh on the upper and lateral aspect.

On laboratory examination, haemoglobin was 6.4 gm%, total leucocyte count was 12700/μL with 70% neutrophils. Fasting and post lunch sugars were180mg% and 286mg% respectively. Renal and liver function tests, chest X-ray were normal. USG (ultrasonography) of right knee and leg showed loculated collection with thick internal echoes communicating with the joint cavity. USG of lateral aspect of left thigh showed 5ml collection in subcutaneous plane. To look for disseminated infection, USG abdomen was done which showed a collection of 35ml in segment VIII of liver suggestive of abscess. Computed tomography of chest and abdomen revealed hepatic abscess as mentioned above and cold abscess along left lower rib with minimal pleural reaction. The possibility of recurrent disseminated melioidosis as a result of inadequate treatment due to default by patient was considered. Patient was started on intravenous ceftazidime 2gm 8 hourly. Sugar was controlled with 30/70 insulin twice a day subcutaneously.

The aspiration of fluid from knee and hepatic abscess showed purulent material with polymorphonuclear predominance. Culture of the material from both the sites had growth of Burkholderia pseudomallei which was susceptible to piperacillin, ceftazidime, cefoperazone-sulbactam, imipenem, meropenem, cotrimoxazole, chloramphenicol and ciprofloxacin. Patient was given ceftazidime for 4 weeks and was started on oral trimethoprim-sulphamethoxazole (160/800 mg tab twice a day) at the time of discharge. Insulin was continued with tight sugar monitoring.

Discussion
Melioidosis also known as pseudoglanders or Whitmore’s disease is caused by Burkholderia pseudomallei a gram negative bacillus previously classified under genus pseudomonas. The infection can be acquired by inoculation of environmental organisms through penetrating wounds or into existing skin lesions, inhalation or the aspiration of contaminated water. B. pseudomallei is a containment level-3 pathogen and laboratory-acquired infection is reported3

The infection is more common in males who indulge in agricultural activities. DM increases the risk of infection by 100 folds. Other predisposing factors are renal disease, alcoholism, cirrhosis, chronic lung disease, thalassemia, chronic granulomatous disease, porphyria cutanea tarda, cystic fibrosis, hemosiderosis, splenectomy, aplastic anemia, febrile neutropenia, mycobacterial disease, malnutrition or...
immunosuppression. The disease spectrum is quite variable. The infected individual can remain carrier with totally asymptomatic infection or remain latent for years and manifest whenever cell mediated immunity is suppressed. Melioidosis is known to present as a febrile illness, ranging from an acute fulminant septicaemia to a chronic, debilitating localized infection and abscess formation. There is usually no obvious infected wound or evidence of recent trauma. It can present as acute septicemia with high grade fever, lower respiratory infection and hepatosplenomegaly, disseminated cutaneous and visceral abscesses and shock. Pneumonia is the most common presentation of melioidosis seen in approximately half of all cases. Acute pulmonary syndrome with pneumonitis involves upper lobes of the lung and is confused with tuberculosis. Acute or chronic localized suppurative infection can involve skin, brain, lung, myocardium, liver, spleen, bones, joints, lymph nodes and even the eye.

Jesudasan et al. reported three cases of melioidotic septic arthritis from south India, and three cases imported from Indian subcontinent have been reported in UK. The most commonly affected joints are the knee and shoulder. Generally, septic arthritis arises from hematogenous dissemination of the organism, but it may follow direct spread from other organs or soft tissue infections over joints.3

The diagnosis is by microscopic examination of aspirate which reveals bipolar or unevenly staining Gram-negative rods. It has a low specificity and sensitivity. Bacterial identification by culture is the only accepted ‘gold standard’. Resistance to aminoglycosides and older-generation penicillins and cephalosporins is characteristic.

Intravenous ceftazidime 2gm 8 hourly for 2 to 4 weeks is the treatment of choice. The carbapenem antibiotics (imipenem and meropenem), though slightly less effective, can be given as an alternative. Trimethoprim (8mg/kg/day) and sulfamethoxazole (40mg/kg/day) combination in four divided doses is given for 12–20 weeks after intravenous therapy to eradicate the organism and prevent relapse. The alternative for prolonged treatment is doxycycline or amoxicillin-clavulanate, though less effective.4

Even with optimal treatment, the mortality from acute severe melioidosis is high (30% to 47%). In patients who survive, there is often chronic morbidity resulting both from the disease itself and the underlying conditions.

Documented reports of melioidosis from India have been few and sporadic, the majority being from Christian Medical College, Vellore, Tamilnadu. Lack of awareness, a low index of suspicion and inability of rural population to access health services probably contribute to the paucity of reports from the Indian subcontinent. Sporadic cases have also been reported from Maharashtra, Kerala, Karnataka and Pondicherry.2

Summarising, with the existing geographical and climatic conditions and a susceptible diabetic population, the western coast of South India seems to be an ideal setting for endemicity of melioidosis. The clinical manifestations of melioidosis alone are not diagnostic, and such a diagnosis requires a high degree of suspicion prolonged treatment to avoid chronic debility and mortality.

References