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Case Report

CASE REPORT

Chronic melioidosis presenting with multiple abscesses

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Abstract

Melioidosis is common in Australia and Southeast Asia and is increasingly recognized in India. It presents in various forms which are difficult to identify and often mimics suppurative infections, tuberculosis, fungal infections, malignancy and systemic rheumatic diseases. Presentation may vary from local disease to disseminated abscesses, pneumonia and sepsis. Disease is common and severe in diabetics. We describe a case of diabetic man presenting with fever, septic shock, peri-articular nodules, lung opacities and multiple abscesses in muscles for the past 3 months remaining undiagnosed. Autoimmune conditions were ruled out and infection with *Burkholderia pseudomallei* was suspected. *Burkholderia pseudomallei* was isolated from blood cultures, confirming the diagnosis. Prolonged treatment with intravenous ceftazidime followed by oral cotrimoxazole led to complete recovery. Awareness of this infection is required by clinicians and microbiologists unfamiliar with the condition to diagnose the disease early to prevent mortality.

INTRODUCTION

Differential diagnoses of disseminated abscess in various organs include common suppurative infections, tuberculosis, fungal infections and systemic rheumatic diseases. Melioidosis is a very important cause of disseminated abscesses, specially, in a diabetic [1]. Melioidosis is frequently reported from India and its presentation is varied [2]. Most cases are diagnosed late due to the lack of awareness and unavailability of proper laboratory facilities. Many cases are mistakenly treated as tuberculosis, or worked up for malignancy. The clinical spectrum of disease ranges from local abscess to severe form, which may cause disseminated abscesses, pneumonia, sepsis or death. Delayed diagnosis contributes to increased mortality and morbidity. We describe a case of a diabetic patient who presented with fever, pyomyositis and lung nodules, not been diagnosed for 3 months. Recommended treatment led to complete recovery. The case highlights the importance of awareness of this infection among clinicians unfamiliar with the condition.

CASE REPORT

A 36-year-old diabetic man from rural part of India presented with high-grade fever, multiple swellings around joints and weight loss of 10 kg for the past 3 months. There was no cough, dyspnoea or abdominal pain. In this period, he had an episode of septic shock from which he recovered, but fever and swellings did not respond to various antibiotics such as cefaperazone/sulbactum, amikacin and levofloxacin. Blood cultures were sterile. There was significant proteinuria and chest CT showed patchy infiltrates in lungs without cavity, nodules or lymphadenopathy. Granulomatosis with polyangiitis was considered as a differential diagnosis, but anti- neutrophil cytoplasmic antibody, anti-

proteinase 3 and anti-myeloperoxidase turned out to be negative. Rheumatoid factor, anti-nuclear antibody and HIV were also negative. Empirical steroids resulted in worsening of symptoms and enlargement of swellings. There was no past history of tuberculosis, recurrent infections, alcoholism or intravenous drug abuse. There was no history of exposure to dust, bird droppings, animal handling or agriculture work. A specific diagnosis could not be made and he was referred to our Rheumatology Center in New Delhi.

On presentation to us, he was cachectic, had bilateral knee joint contractures and came on a stretcher. There were diffuse, multiple, tender and firm nodules deep in muscle plane above left elbow (5 × 5 cm), right wrist (3 × 2 cm), right forearm (7 × 2 cm), right knee (10 × 10 cm) and ankles. Figure 1 shows the large swelling around the right knee. Repeat CT revealed mild hepatosplenomegaly with multiple bilateral lung nodules (Fig. 2a). Musculoskeletal ultrasound showed multiple intramuscular densities. Fine needle aspiration from these lesions was inconclusive and no organism could be detected on gram and acid fast stains. On the basis of above investigations, polymyositis, undifferentiated arthritis, aseptic systemic abscess or other connective tissue disease appeared unlikely. A high dose of insulin (86 IU/day) was needed to control his diabetes. He continued to be febrile and his nodules developed into abscesses in spite of 2 weeks of vancomycin. A large volume of pus (up to 800 ml) was drained from these abscesses.

A review of patient's history revealed that he drank untreated water from a waterfall 1 month before falling sick. As he had diabetes with lung nodules and pyomyositis, an infective aetiology was considered. Repeat blood culture was sterile. Gram/acid fast/potassium hydroxide stains and culture of pus did not show any organism. Melioidosis was suspected and this possibility was discussed with microbiologist. Blood culture incubated in special media isolated *Burkholderia pseudomallei*.

Diagnosis of melioidosis was confirmed and he was administered intravenous ceftazidime for 4 weeks. He showed dramatic improvement; his fever subsided, pyomyositis resolved and nodules in lungs subsided. He then underwent eradication phase with 6 months of oral cotrimoxazole. A repeat contrast CT scan of the thorax showed clearance of lung nodules (Fig. 2b). We managed his diabetes with metformin and he also underwent physical therapy to relieve his contractures. He had complete resolution of all his symptoms and was able to return to his baseline function.

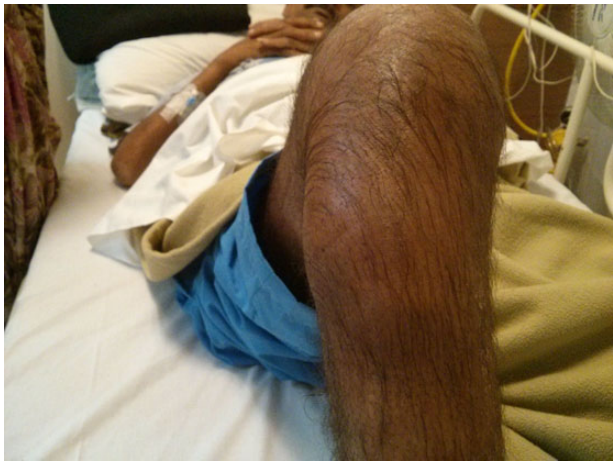

Figure 1: Large abscess over right leg.

DISCUSSION

Melioidosis is caused by gram-negative bipolar, safety pin-shaped bacillus, *B. pseudomallei*. Familiarity with different presentations of this infection is essential for early diagnosis as delayed diagnosis contributes to increased mortality and morbidity. Melioidosis is associated with a range of mortality from 10% [3] to 39% [4]. For those with septic shock, it can rise up to 86% [5]. Globally, it is seen in tropical climates, especially in Southeast Asia (Malaysia, Thailand and Singapore), China, Taiwan and northern Australia [6, 7]. It has been reported anecdotally from all developing countries, although it frequently goes unrecognized. From India, cases are reported frequently from all regions, suggesting that it is becoming an endemic disease, but a large number of cases are not diagnosed [8]. This perceived low prevalence is due to the lack of awareness among physicians, microbiologists and poor laboratory facilities. It is usual for laboratories in non-endemic locations to misidentify the bacterium as commensal. Although there has been expansion of the disease

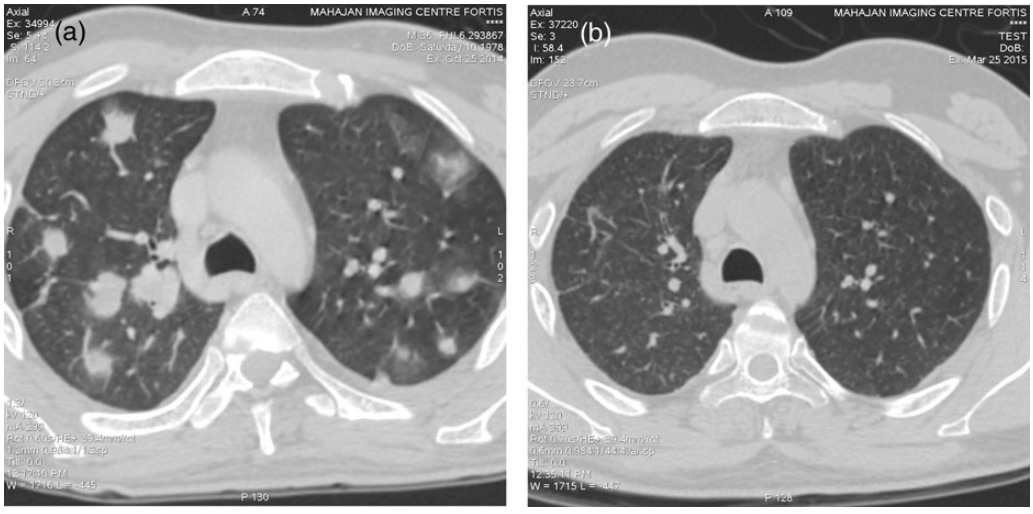

Figure 2: (**a**) CT thorax showing multiple nodules of melioidosis. (**b**) CT thorax showing resolution of nodules after antibiotic therapy.

in tropical countries due to global warming and population movement, many cases are unmasked by improved clinical surveillance and better diagnostic methodology [9].

Melioidosis is transmitted by inhalation of contaminated dust or water droplets, percutaneous inoculation and ingestion of contaminated water. Our patient most probably acquired the infection by ingesting contaminated water. It is a seasonal disease more common in wet season and epidemics occur during tropical monsoonal storms, cyclones, hurricanes and typhoons [10]. There may be a shift from inoculation to inhalation as the predominant route of spread during epidemics [11]. It is a disease of adults with <5% cases belonging to paediatric age group [12].

Risk factors for melioidosis are diabetes mellitus, chronic renal failure, alcohol abuse, thalassaemia, chronic lung or liver disease, malignancy and immunosuppression. Diabetes mellitus is found in up to 60.9% of affected patients [1]. Latent infection is common, with one study reporting a 4% reactivation rate into active illness [13]. This can occur many years after exposure—at one point in time, this was referred to as the Vietnamese time bomb due to its reactivation in returned serviceman from Vietnam [14]. Melioidosis is divided into subclinical, acute and chronic disease. Acute cases are those where symptoms were present for <2 months. The spectrum of clinical presentations ranges from to asymptomatic or minor localized abscess or nodule to severe, fulminant disease (such as shock, multiorgan abscesses and death). Exposure to bacilli most commonly results in subclinical disease. Clinical disease presents most commonly in acute form and only 9% presents as chronic illness [12]. The most common presentation of melioidosis is pneumonia, occurring in more than half the cases [15]. Acute melioidosis commonly presents as pneumonia and may progress to septic shock with mortality in up to 90% of cases [13]. Infiltrates can occur in lungs which coalesce and cavitate and may cause multiple metastatic abscesses. Initially, our patient had chest infiltrates that were non-specific. Chronic disease closely mimics tuberculosis or malignancy [6]. However, melioidosis can present in a wide variety of patterns, ranging from genitourinary symptoms to encephalomyelitis [13].

Treatment has two phases: the intravenous intensive phase for acute disease, followed by eradication phase [16]. Intravenous ceftazidime (2 g, 6 hourly) and meropenem (1 g, 8 hourly) are agents of choice in the intensive phase. Cotrimoxazole is used in the maintenance phase. Treatment duration depends on local versus severe systemic infection. In local or mild disease, intensive phase is of 2–4 weeks followed by 3 months of maintenance therapy. In severe infection including neurological disease, initial intensive therapy is prolonged for 6–8 weeks. It is followed by an eradication phase with oral cotrimoxazole for the next 6 months [17]. Meropenem in double dose is preferred in neurological melioidosis [13]. Our patient responded very well to this treatment regimen and was successfully treated.

Mortality in melioidosis is <10% where resources for rapid diagnosis, early implementation of antibiotics and good intensive care facilities for managing severe sepsis are available [3]. However, such resources are limited in many regions of India leading to underdiagnosis and high mortality.

CONFLICT OF INTEREST STATEMENT

All the authors hereby declare that no conflict of interest exists and there are no financial or personal relationships or affiliations that could influence (or bias) the author's decisions, work or manuscript.

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There is no source of funding or sponsor for this case report.

ETHICAL APPROVAL

Ethical approval from institutional ethical committee has been taken before submitting this article.

CONSENT

Consent from the parents of the deceased patient has been taken for this case report.

GUARANTOR

A.K. agrees to be a guarantor for the accuracy of final manuscript and submission of this article.

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