

Retraction

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The authors of the above paper have requested to retract the paper due to some inaccuracies, which have been pointed out by fellow microbiologists in Sri Lanka, and also due to authorship disputes.

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Case Report

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Chronic melioidosis causing an inguinal sinus in a Sri Lankan male

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Introduction: Melioidosis is an emerging infection in South Asia. It has a spectrum of presentation causing many difficulties in early diagnosis and the control of infection. Here we report a rare case of melioidosis causing an inguinal sinus in a Sri Lankan male.

Case presentation: This 54-year-old male had recurrent admissions to hospital with multiple acute infections including acute inguinal lymphadenitis, left lobar pneumonia and septic arthritis of the left knee joint. He was treated with short courses of antibiotics. Blood cultures revealed no bacterial growth. Ultrasound scan of the abdomen showed multiple loculated abscess formation typical of melioidosis. Repeat blood culture in selective Ashdown's medium was positive for the growth of *Burkholderia pseudomallei*. He was treated with ceftazidime and imipenem for 4 weeks but was lost to follow-up before starting an eradication regimen. Later, he presented with a large left inguinal sinus and a draining sinus in the left thigh.

Conclusion: Presence of multiple septic foci in this patient within a short span of time was due to discharge of bacilli into the bloodstream from the hidden infection in the spleen. This is a good example of an immune organ itself acting as a source of infection under suboptimal immune response. This case also highlights the importance of full screening and complete eradication of the infection at the initial diagnosis before establishment in relatively avascular tissues.

Keywords: ceftazidime; inguinal; sinus melioidosis.

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Introduction

Melioidosis is an emerging infection in many developing countries including those in South Asia. It is caused by the bacterium *Burkholderia pseudomallei*. It is endemic in southeast Asia and causes outbreaks after monsoons (Dance, 2000). It has a spectrum of presentation ranging from a severe septicaemic illness to chronic progressive infection. Immune-deficient patients with diabetes mellitus, chronic alcoholism, or chronic renal disease are at a higher risk than the healthy population (Currie *et al.*, 2000). Suppurative lymphadenitis caused by melioidosis has been rarely encountered in endemic areas; cervical nodes are the commonest groups of lymph nodes involved (Chlebicki & Tan, 2006). Several cases of melioidosis have been reported in Sri Lanka (Corea *et al.*, 2012), probably due to an increase in international travel to endemic countries. Delay in diagnosis due to the nonspecific nature of its presentation causes a major challenge to physicians. Best clinical judgment and focused microbiological investigations are very important for early diagnosis. Therefore, it is important to identify different patterns of presentations of melioidosis. Here we report a rare case of

melioidosis causing inguinal sinus formation in a Sri Lankan male.

Case report

A 54-year-old male from Ratnapura diagnosed with diabetes mellitus presented to the medical department with a painful lump in the left inguinal region and fever of 1 week's duration. There was no primary focus of infection found in the adjacent draining area. His full blood count showed neutrophil leucocytosis, and erythrocyte sedimentation rate was elevated (96 mmHg/1st hour), indicating an acute inflammatory process. He was started on intravenous cloxacillin 500 mg every 6 h and continued with the oral form for 1 week. His diabetes control was satisfactory. One month later he presented again complaining of left lower pleuritic-type chest pain and fever. On further inquiry, he had got fever on and off together with constitutional symptoms since his previous illness. Chest X-ray was suggestive of left lower lobe consolidation and was treated as left lobar pneumonia with intravenous co-amoxiclav 1.2 g three times daily for 1 week. Three weeks

later, he came back with severe left knee joint pain and fluctuant swelling of the left knee joint suggestive of septic arthritis. After knee joint aspiration he was immediately started on intravenous ceftriaxone 2 g twice daily together with intravenous clindamycin 600 mg three times daily. As on previous occasions, he was getting better on this treatment but never felt completely well.

Investigations

Despite satisfactory control of diabetes, recurrent presentations with acute infections raised the suspicion of an immune deficiency. Therefore, he was investigated for retroviral infection, which was negative. Normal serum globulin levels and cell counts were against an immune deficiency. A two-dimensional echocardiogram was performed to exclude the possibility of discharge of septic emboli from an infected valve; it was normal. His blood cultures were repeatedly negative. An ultrasound scan of the abdomen was carried out owing to complaint of persistent left upper abdominal pain. It revealed multiple small hypo-echoic areas in the spleen suggestive of multiple abscesses (Fig. 1).

Diagnosis

Taking together the clinical history and the pattern of involvement of the spleen, we suspected melioidosis at this stage. Repeated blood cultures were done and selective Ashdown's medium was used based on this strong suspicion; later, one culture became positive for *B. pseudomallei* (Fig. 2). This organism is a biohazard due to the high risk of rapidly fatal Gram-negative septicaemia. Therefore, correct isolation is vital; misidentification of *B. pseudomallei* as *Burkholderia* ssp. by the VITEK 2 system has been reported by Zong *et al.* (2012). *B. pseudomallei* is a highly



Fig. 2. Appearance of *B. pseudomallei* colonies in Ashdown's medium. *B. pseudomallei* colonies are wrinkled and have a metallic appearance. The medium contains crystal violet and gentamicin as selective agents to suppress the growth of other bacteria.

motile organism that grows more quickly under aerobic conditions, it is oxidase-positive and it has a 'safety pin' appearance (bipolar staining).

Treatment

Susceptibility was determined by the broth microdilution method. The organism was sensitive to ceftazidime, imipenem and piperacillin/tazobactam. The patient was started on intravenous ceftazidime 2 g three times daily together with intravenous imipenem 1 g three times daily and continued for 4 weeks according to guidelines published by Timothy *et al.* (2006). Unfortunately, the eradication regimen was not completed as the patient was lost to follow-up.

Outcome and follow-up

Six months later, the patient presented with multiple suppurative inguinal lymph nodes with sinus formation and a large draining sinus in the left mid-thigh. X-ray of the left femur was suggestive of chronic osteomyelitis. This presentation indicated a chronic form of the disease due to dormant bacilli in deep tissues, mostly due to inadequate antibiotic coverage. Excision biopsy of the lymph nodes was positive for the growth of *B. pseudomallei*. The patient was started on intravenous ceftazidime 2 g three times daily based on the sensitivity pattern and continued for 6 weeks. With regular surgical debridement and continuation of antibiotics, inguinal sinuses resolved completely except for the sinus in the thigh.

Discussion

This patient was from Ratnapura District, which is situated in the wet zone of Sri Lanka. He first presented in October,

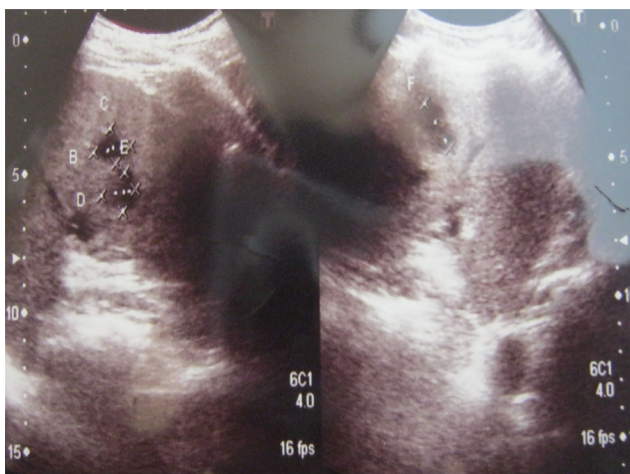


Fig. 1. Ultrasound scan of the spleen. Multiple hypo-echoic areas in the splenic parenchyma are shown. This appearance is suggestive of multiple splenic abscesses characteristic of melioidosis.

at the end of the southwest monsoon (June to October). This was essentially a locally acquired case as this patient had never travelled to any other country. Although data are lacking with regard to the exact locations, most of the other reported cases are scattered around Colombo, Western Province. However, we believe under-reporting due to lack of laboratory facilities may have contributed to this. We suggest that it is important to maintain epidemiological data on melioidosis at a national level through a proper surveillance system.

This case represents a range of presentations of melioidosis in a single patient, causing a diagnostic challenge. Inguinal lymphadenitis with sinus formation is an extremely rare form of chronic melioidosis. Presence of multiple septic foci in this patient within a short span of time was due to discharge of bacilli into the blood stream from the hidden infection of the spleen. This is a good example of an immune organ itself acting as a source of infection under suboptimal immune response. This case highlights the importance of full screening and complete eradication of the infection at initial diagnosis. Once the infection is established in relatively less vascular tissues such as bone, irradiation is extremely difficult. In a clinical scenario where a patient presents with multiple acute septic foci in different sites, melioidosis should be considered as a differential diagnosis alongside infective endocarditis and immune suppression etc. This case also highlights the importance of performing repeated blood cultures in patients with a long-standing fever and patients suspected of having blood-borne sepsis. Bacilli will enter into the bloodstream at different times and may not be captured in all blood samples taken for culture. It is important to use selective Ashdown's medium early in suspected cases of melioidosis. Melioidosis is an emerging infection in South Asia, with increasingly reported cases (Dance, 2000). Still, we believe melioidosis is under-diagnosed and under-reported. This case gives a better understanding of the variable presentations of melioidosis

and the practical difficulties in its diagnosis. This report gives important insights for early diagnosis in resource-poor settings, which will help in better control of this devastating infection in the future.

Acknowledgments

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent form is available for review from the Editor-in-Chief of this journal. We acknowledge the support given by Dr S. K. Jayatilleke in microbiological diagnosis and selection of antibiotics. The authors declare no conflicts of interest.

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