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Case Report

Melioidosis: The Great Mimicker: A Case Report

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Introduction

Melioidosis is a disease endemic to Southeast Asia and northern Australia and is caused by a Gram negative, saprophytic organism, *Burkholderia pseudomallei* [1]. It is commonly seen among agricultural workers. The disease was first described by the pathologist Captain Alfred Whitmore and his assistant C.S.Krishnaswami [2]. Melioidosis emerged as a major public health concern in Thailand in the latter half of the 21st century, accounting for 20% of all community acquired bacteraemias in Ubon Ratchathani, Thailand [3]. Similarly, it has been recognized as the most common cause of fatal bacteraemic community acquired pneumonia at the Royal Darwin Hospital in Northern Australia [3]. The first case of melioidosis in Sri Lanka was reported in 1927 in a European tea broker. Sri Lanka was the third country to report melioidosis in the literature and the first country to detect melioidosis in South Asia [2]. Sri Lanka lies in the endemic belt of the disease. However, cases reported have been largely sporadic. Two clusters have been described in the literature in Batticaloa and Akkaraipattu [4, 5].

Here, we report a case of melioidosis presenting after a short quiescent period of 3 months after an initial presentation of pneumonia. He presented with backache and was later found to have a paraspinous abscess with possible bacteraemic sepsis and ear involvement. The disease course was further complicated by NSAID related nephritis

Case Presentation

Our patient was a 53-year-old farmer from Mahiyanganaya, which is a rural part of the country with a large farming community. He had poorly controlled diabetes on oral hypoglycemic drugs. He gave a history of pneumonia 3 months prior to admission. His previous admission records indicated a protracted disease course of one month. He had

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presented with fever, cough and shortness of breath with pleural effusion. The pleural fluid aspirate, although an exudate, was culture negative. He had been treated for a community acquired atypical pneumonia with empyema, initially with third generation cephalosporins, then IV meropenem and IV levofloxacin due to poor response to the former drugs.

Two months after discharge, the patient had developed gradual onset, mechanical type left lower backache for which he used over-the-counter analgesics. He was admitted to the surgical unit at our hospital and referred to the medical team due to backache associated with high inflammatory markers on a suspicion of myeloma with sepsis. At that time, he had left lower, mechanical type of backache largely localized to the L4, L5 level with right-sided wrist joint pain and swelling and generalized oedema with frothy urine. He denied any fever but had significant constitutional symptoms. He complained of sore throat and voice change with shortness of breath on exertion but not cough or pleuritic chest pain. He also complained of a recent onset hearing impairment and right ear pain with discharge of 2 months duration.

Examination revealed an ill-looking, tachypnoeic patient who maintained saturation above 96% on air. He was tachycardic with a blood pressure just above 100/60mmHg. He was mildly icteric and pale but not febrile. Respiratory system examination was largely unremarkable. Abdominal examination revealed a mild splenomegaly. There was a lump on the back confined to the left lower back at the level of L4/L5 with no spinal tenderness. The lump was around 9×5cm, fluctuant and was lying deep to the skin and subcutaneous tissue with ill-defined margins. Right ear examination revealed a purulent discharge with a perforated tympanic membrane. He had a puffy face with mild lower limb pitting oedema but no evidence of free fluid in the abdomen or chest. Other system examinations were largely unremarkable. Due to the long-standing nature of the illness, evidence of multifocal infections with high inflammatory markers and previous satisfactory response, we started him on IV meropenem with a clinical suspicion of a chronic infection possibly melioidosis.

His investigations revealed high inflammatory markers. Full blood count revealed neutrophil leukocytosis. Serum creatinine was elevated but the patient maintained a satisfactory urine output. Liver functions were deranged with evidence of cholestasis. Ultrasound scan (USS) abdomen showed evidence of splenomegaly but no hepatomegaly or focal liver lesions suggestive of abscesses. Urine showed evidence of nephrotic range proteinuria (Table 1).

Sputum for AFB and Mantoux test were negative. 2D ECHO did not show any evidence of infective endocarditis. Serum ferritin and triglycerides were high, but bone marrow biopsy did not show any haemophagocytes and criteria for haemophagocytic lymphohistiocytosis (HLH) were not fulfilled. As there was bycytopenia and a few criteria to suspect HLH, bone marrow was done in view of excluding HLH and hematological malignancy. There was no evidence of a haematological malignancy in the bone marrow biopsy.

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USS of back revealed echogenic fluid within the L/S paraspinous muscles, measuring approximately 9×6×3 cm suggestive of a parasinous muscle abscess.

All three blood cultures were positive for *Burkholderia pseudomallei*. Culture of ultrasound guided aspirate of pus from the back lump was reported as *Pseudomonas* species, initially. Pus culture from ear discharge was positive for Gram positive cocci.

After confirming the diagnosis of melioidosis, we continued IV meropenem for one month followed by eradication therapy with trimethoprim sulfamethaxazole for a further 3 months. On discharge, his renal and liver function tests had returned to normal. His proteinuria and oedema had completely resolved. We reviewed the patient several times after discharge, and he was in good health with normal biochemistry.

Table 1: Summary of investigations

Investigation	Peripheral hospital	During hospital stay	At discharge
WBC mm^3 Neu Lym Eosinophils Hb Plt MCV	22,000 11.6 439000 	116600 106300 (91.2%) 4200 (3.6%) 1 00(0.1%) 7.5g/dL 43,000 85.4fL	8,000 10g/dl 77,000
ESR	97mm/1st hour	100 mm/ 1st hour	50mm/1st hour
CRP	269	518mg/L	21mg/dl
ALT AST ALP GGT T.Bil D.Bil I.Bil T. protein Alb Glob	32U/L 38U/L 	38.9 U/L 67.2 U/L 1043 U/L 320U/L 62.48mcmol/L 50.52 mcmol/L 12 mcmol/L 5.6g/dL 2.7g/dL 2.9g/dL	34U/L 52U/L 230U/L 130U/L
Serum creatinine BU	1.06 4.6	426mcmol/L 20.56 mmol/L	98.2mcmol/L
Na K	122mmol 4mmol	141mmol 3.5mmol	139mmol 3.7 mmol
INR APTT		1.19 36.4 secs	
➨ RBC ➨ WBC		: NCNC. Few macrocytes , moderate rouleux formation ; Absolute neutrophil leukocytosis. Left shift of	

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➨ Plt		leucocytes. Cytoplasmic vacuolation Mild thrombocytopenia Conclusion: Anemia of chronic disease,with evidence of inflammatory process/infection with mild thrombocytopenia. Could be due to infection	
Rheumatoid factor		621IU/L	
UPCR		2668.2 mg of pro/1g of creatinine	673mg of pro/ig of creatinine
S.Calcium S.Phosphorus		1.91 mmol/L 1.99mmol/L	
Urine full report	Protein: trace Pus cells: 10-20 Red cells : 20-30	Protein :3+ Pus cells : 3-5 Red cells : nil	
Triglycerides		406mg/dL	
Serum.ferritin		3330ng/ml	

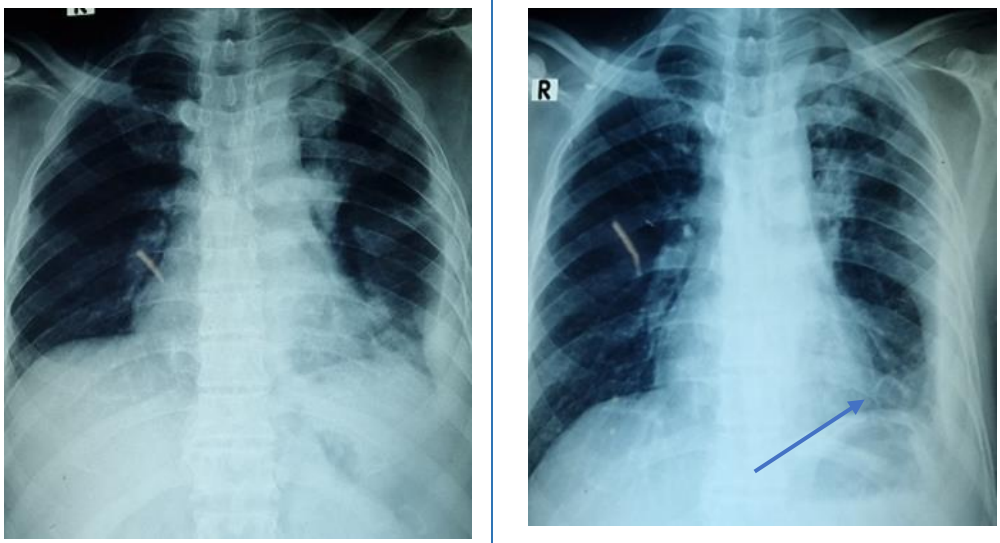

Figure 1: CXR 3/12 prior to admission at peripheral hospital; blue arrow points towards a possible abscess

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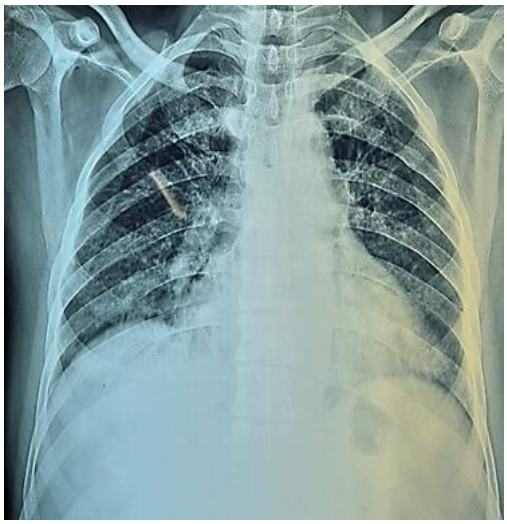

Figure 2: CXR during current admission

Discussion

There are four main presentations of melioidosis, roughly corresponding to the severity of infection. The most severe is acute septicaemia with metastatic lesions in skin, muscle, bone or joints. Localized infection, either acute suppurative or chronic granulomatous and prolonged fever without any apparent site of infection are two other manifestations. The biggest group, by far, is the asymptomatic group, where infection may be subclinical or symptoms arise after a lengthy dormant period, thus justifying its nickname as the Vietnamese time bomb [6]. The latent nature of the infection was elucidated when many veteran American soldiers who fought in the Vietnam war later developed infection after returning to the US. The current record for latency is 26 years [7]. In our case, the patient presented following 3 months of asymptomatic period after his initial presentation of pneumonia, most likely due to inadequate treatment and absence of eradication therapy.

Our patient was treated with meropenem as he had not responded to third generation cephalosporins on his previous admission, and showed a good response. Thus, we could consider melioidosis in the differential diagnosis of patients who do not respond to routine antibiotics but respond to carbapenems [6,8,9]. As described by Dance over 10 years ago, case reports and series may only represent the 'tip of the iceberg' as culture facilities are mostly unavailable in the rural tropics where the disease is prevalent [3,10].

Our case was unique in its presentation, as the patient had been previously partially treated which may have altered the disease course. To our knowledge, this is the first case of melioidosis presenting as a paraspinous muscle abscess in Sri Lanka. However, similar cases with deep muscular abscesses have been reported involving the psoas, gluteus and obturator internus muscles [6].

A case of spinal epidural abscesses which presented with neurological deficits has been reported in India [11]. This patient initially presented with fever and backache similar to ours, and diagnosis was delayed until the patient developed paraplegia. MRI revealed anterior epidural, left paraspinal, left iliac, and sacroiliac abscesses. That patient had to be operated on as the infection was at multiple sites and did not respond to antibiotics alone. We initiated meropenem very early in this patient as we had a clinical suspicion of melioidosis. This may have prevented complications such as epidural abscesses. There are many reported cases of spondylodiscitis due to melioidosis in the literature, most were initially misdiagnosed as tuberculosis [11,12].

Also noteworthy in this patient, was the nephritic picture at outset which was possibly due to treatment related complications. The patient had been using NSAIDs liberally for pain relief which could have precipitated a transient nephrotic-nephritic syndrome that would have been further complicated by sepsis. The clinical picture was thus confused with a possible myeloma causing sepsis.

Our patient also complained of an ear discharge, with hearing impairment. A case report of a patient with chronic suppurative otitis media with cholesteotoma has been reported in Thailand. However, cultures only grew *Pseudomonas aeruginosa* although *Burkholderia* serology proved to be positive [13].

In many cases reported worldwide, the diagnosis had been delayed for many reasons including a low level of awareness and delay in obtaining a tissue diagnosis [13]. This may be due to a lack of facilities for culture or the misidentification as *Pseudomonas* due to a lack of microbiological experience. In our case, the pus culture from abscess was initially identified as *Pseudomonas*. Upon communicating our clinical suspicion to the microbiology team, an accurate diagnosis of melioidosis was obtained by blood culture. Therefore, communication may be the key in obtaining a proper microbiological diagnosis in cases of suspicion. A correct microbiological diagnosis is important as effective treatment with a standard antibiotic regime is required to prevent inadequate treatment and relapse as seen in our patient.

Our patient was treated for 4 weeks with intravenous antibiotics followed by a 3-month course of eradication therapy and is doing well to date. Yet the importance of follow up needs to be emphasized due to the heterogenous nature of the disease course.

Conclusion

Melioidosis is an emerging disease in Sri Lanka that might have varied presentations including those that are similar to much commoner endemic diseases such as tuberculosis. Patients may have a protracted or relapsing disease course due to inadequacy of initial treatment or delay in the diagnosis. Our case highlights the variability of disease presentation in melioidosis and the importance of microbiological confirmation as the disease needs long term treatment. We emphasize the need for a high degree of suspicion and the need to liaise with the clinical microbiologist which might hasten accurate microbiological identification. Early and adequate treatment of the

disease with induction and eradication is mandatory to prevent relapses and fatal complications. Partial treatment may lead to alteration of disease presentation which may give rise to a diagnostic challenge.

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