CASE REPORT

A rare cause of recurrent neck abscess: melioidosis

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Introduction

Melioidosis is an emerging recent infectious disease in Sri Lanka caused by a Gram-negative, oxidase-positive bacillus Burkholderia pseudomallei [1]. The clinical manifestation varies from severe septicemic illness to chronic infection, with high mortality and morbidity among untreated cases [2]. The best clinical assessment and focused microbiological investigations are the cornerstone factors to detect early diagnosis and prompt treatment of melioidosis to prevent mortality and morbidity. Here, we report a case of melioidosis in a young lady who presented with recurrent neck abscess in the Kilinochchi district.

Case Report

A 24-year-old lady presented with fever and right side of the neck swelling for constitutional symptoms of two weeks. She denied any other systemic symptoms, and she had no history of tuberculosis or diabetes mellitus. She was not actively involved in cultivation. However, she had a history of muddy exposure. Upon examination, she had a tender neck swelling without any palpable cervical lymphadenopathy. Her pulse rate was 80/minute regular and her blood pressure was 110/70mmHg. Further systemic examination was unremarkable. Her full blood count showed mild leucocytosis with predominant neutrophils, normochromic normocytic anaemia, normal platelets. Her renal and liver profile was normal. Her erythrocyte sedimentation rate (80 mmHg/1st hour) and C reactive protein (12.2mg/dL) were elevated, indicating an acute inflammatory process. Her chest x-ray was normal. Her ultrasound imaging of the neck favoured the diagnosis of neck abscess. Initially, she was managed with co-amoxiclav 1.2g intravenous three times daily for 10 days along with incision and drainage of the abscess. The pus culture showed no growth. However, she had a persistent fever and neck abscess following a course of antibiotics therapy. Her repeated ultrasound imaging of the neck showed the residual neck abscess. She had persistent leucocytosis with predominant neutrophils and elevated inflammatory markers (ESR-126 mmHg/1st hour and CRP- 10.2 mg/dL). Furthermore, she was managed with intravenous piperacillin-tazobactam 4.5g three times daily along with incision and drainage of the abscess. Furthermore, the trial of antituberculosis treatment was initiated due to persistent fever with high inflammatory markers after expert opinion. However, her X pert (MTB/RIF) for tuberculosis; WHO recommended Rapid Diagnostic (WRD,) using in Sri Lanka (Manufacture details: Cepheid gene Xpert system; GXMTB/RIF-10, Serial no:698209) was negative. On review two weeks and one month later, she had no improvement with therapy. Subsequently, incision and drainage of neck abscess were carried out. The direct smear showed Gram (-) ve, oxidase (+) bacilli and Burkholderia pseudomallei was isolated from pus culture. The melioidosis antibody titre was > 10,240. Melioidosis was the definitive diagnosis and was managed with intravenous Mereponem 1g thrice daily and Cotrimoxazole 1920mg twice daily for two weeks as intensive therapy. She had improved with therapy. Cotrimoxazole 1920mg twice daily for three months was continued with close monitoring of full blood count and liver enzymes every week for one month and then twice monthly for a two-month duration. She recovered with the course of antibiotic treatment for a three-month duration.

Discussion

Melioidosis is a pyogenic infection presenting as acute or chronic infection in humans following percutaneous inoculation which is caused by the Burkholderia pseudomallei. The endemic areas of melioidosis were noted in tropical and subtropical zones of South East Asia and Northern Australia. In 1927, the first case of melioidosis was reported in Sri Lanka by a European tea broker [3]. It is recently noted as an emerging infection in Sri Lanka including Northern Sri Lanka even though Sri Lanka has been considered non-endemic for melioidosis, probably due to an increase in international travel to endemic areas. Diabetes mellitus is a common risk factor for melioidosis which has a positive correlation of 76%. The recognized risk factors for melioidosis are alcohol use, chronic lung disease, chronic renal disease, malignancy, immunosuppression and thalassemia [2]. Our patient had no obvious risk factors for

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melioidosis. The clinical presentations vary from a severe septicaemic illness to chronic progressive infections which include pneumoniae, septicaemia, arthritis, abscess [4]. Pulmonary involvement and deep-seated abscess are the common clinical manifestations. Our patient presented with a recurrent neck abscess. Isolation of the causative bacterium, Burkholderia pseudomallei, in culture is the gold standard for the diagnosis [5]. Isolation of Burkholderia pseudomallei in pus culture favoured the diagnosis of melioidosis in our case. The blood culture may be negative in some cases which may be due to the initiation of antibiotics before blood culture. The melioidosis antibody titre is useful in culture-negative cases. Meropenem is the drug of choice of melioidosis in intensive therapy [6].

However, the combination of Meropenem and Co-trimoxazole is the recommended treatment with a successful outcome in systemic melioidosis. Our patient was managed with intravenous Mereponem 1g thrice daily and Cotrimoxazole 1920mg twice daily for two weeks during intensive therapy. After intensive therapy, the patient should continue on oral cotrimoxazole as the maintenance therapy to prevent relapse for three to six months depending on clinical improvement [6]. Cotrimoxazole 1920mg twice daily for three months was continued for three months duration in our patient. She improved clinically with the course of antibiotics therapy. Several studies have shown the fatalities even after initiation of therapy or due to late diagnosis or presentation.

The delayed diagnosis due to the nonspecific presentation is a clinical challenge to physicians which causes high mortality and morbidity of patients. The best clinical assessment and focused microbiological evaluation are the cornerstone for early diagnosis of melioidosis. Surgeons and physicians are aware of identifying different clinical patterns of melioidosis which is a current and future emerging infection in Northern Sri Lanka.

All authors disclose no conflict of interest. The study was conducted in accordance with the ethical standards of the relevant institutional or national ethics committee and the Helsinki Declaration of 1975, as revised in 2000.

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Learning Points:

- Clinical judgment and focused microbiological investigations are very important key factors for early identification of indexed cases of melioidosis.
- Surgeons should think possibility of melioidosis as one of important differential diagnosis among patient presenting with recurrent abscess in clinical practice as it is an emerging recent infection in Sri Lanka.