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Case Report

Recurrent Parotid Abscess; A Diagnostic Dilemma- A Case Report

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Abstract

Meliodosis caused by *Burkhloderia pseudomallei* has recently gained importance as an emerging pathogen in Sri Lanka as increasing number of cases reported mainly from the north and east provinces of the country. It causes a wide spectrum of clinical manifestations like pneumonia, septicaemia, arthritis and abscess in multiple organs potentially mimicking other infections like tuberculosis which is much commoner in a country like Sri Lanka where it is endemic. We describe a 54-year-old male nursing officer who presented with recurrent parotid swelling with lymphadenopathy which was initially diagnosed as extrapulmonary tuberculosis, however poorly responding to standard anti tuberculous treatment. Further investigations isolated *B. pseudomallei* from parotid abscess establishing the diagnosis of Meliodosis.

Keywords: Parotid Abscess; Melioidosis; Tuberculosis

Introduction

Melioidosis is an infection caused by *Burkhloderia pseudomallei*, a facultative gram negative organism, previously known as *Pseudomonas pseudomallei* [1]. The infection is usually acquired by inoculation or inhalation of soil and contaminated water [2]. Therefore certain occupations like farmers, construction workers and military personnel are at higher risk of the disease. Additionally, patients with diabetes, chronic lung, liver and renal diseases are greatly susceptible for severe infection. Globally, cases of Melioidosis have been reported most frequently from Northern Australia and Southeast Asian countries like Thailand, Malaysia and Vietnam [3]. However, it has lately gained importance as an emerging pathogen in Sri Lanka over last decade [2].

This infection can induce a wide spectrum of clinical manifestations varying from acute to chronic involving multiple organs of the body [4]. Therefore, melioidosis can mimic many other common infections and often called 'the great mimicker'. Therefore it has a high tendency to be misdiagnosed in clinical practice leading to delayed institution of proper management resulting a significantly high morbidity and mortality [2,5].

Case presentation

A 54 year old male nursing officer presented initially to National hospital- Kandy, Sri Lanka with left sided facial swelling involving parotid region for one month duration. He noticed that the swelling was gradually enlarging since beginning. However, he denied associated pain, redness, fever or other constitutional symptoms. There was no facial weakness or ear discharges. His past medical history revealed hypertension, dyslipidaemia and type 2 diabetes mellitus requiring insulin therapy for twelve years with satisfactory disease control. There was no past history or contact history of tuberculosis.

An examination revealed minimally tender swelling without erythema or discrete mass in left parotid region. There was no cervical or axillary lymphadenopathy at initial presentation. However, he developed enlarged right sided cervical lymph nodes about 2 weeks later.

Initial investigations revealed white cell count 8000 with normal differential, haemoglobin 12.4 g/dL, platelets 345000/mm,

sedimentation rate 34 mm/hr, C-reactive protein 8 mg/dl (normal < 10). Ultrasound examination demonstrated localized collection of pus in superficial lobe of left parotid gland. However, attempt for aspiration of pus was unsuccessful, due to small collection size. Further evaluation with lymph node biopsy identified suppurative granulomatous inflammation suggesting tuberculosis. Aspiration for acid fast bacilli, tuberculous culture and GeneXpert-MTB/RIF test were negative. Moreover, chest radiograph and Mantoux test failed to reveal any abnormality. Further investigations including HIV serology, lactate dehydrogenase level (LDH), ANA, Toxoplasma antibodies, blood culture, blood picture, serum calcium, 24 hour urinary calcium excretion and echocardiography were within normal limits. Computerized tomography of neck, chest and abdomen showed no evidence of Lymphoma, sarcoidosis or no any other abscess. However, he was commenced on standard regimen of ant tuberculous therapy consisting isoniazid, rifampicin, ethambutol and pyrazinamide as a clinically diagnosed case of extrapulmonary tuberculosis.

The facial swelling and lymph node enlargement gradually resolved over 2 weeks of anti-tuberculous treatment. However, he presented again with recurrence of ipsilateral painful facial swelling associated with fever at two month of anti-tuberculous treatment. A clinical examination revealed tender erythematosus lump localized to left parotid area suggesting a parotid abscess, which was confirmed with ultrasound examination. Investigations performed at current admission noted WBC- 6700, neutrophil 67%, sedimentation rate 40mm/hr and CRP- 5.1 mg/dL. The abscess was managed surgically with incision and drainage. Pus culture yield *Burkholderia pseudomallei*. Serum melioidosis antibody testing using indirect hemagglutination method was reactive with a titre more than 1:1286. Together with microbiological and serological evidence the diagnosis of melioidosis was made. The antituberculous therapy was withheld and he was treated with intravenous meropenem and oral co-trimoxazole according to sensitivity pattern and made a marked clinical improvement.

Discussion

Melioidosis is a disease with protean clinical manifestations ranging from chronic localized infection to acute fulminant septicaemia with disseminated infection leading to formation of abscess in multiple organs. Therefore it is frequently been misdiagnosed and mistreated for other common infections in clinical practice. Hence, it is often called 'the great mimicker' [2,5].

Pulmonary disease is considered the most common form of meliodosis affecting approximately half of the cases [6]. Radiological appearance of pulmonary melioidosis is non-specific and often similar to findings of tuberculosis or community acquired pneumonia [7]. Due to strikingly similar clinical and radiological manifestations, it is often difficult to differentiate meliodosis from other infections like tuberculosis which is much commoner in countries of Indian subcontinent including Sri Lanka. Vidylakshmi., *et al*. published a report highlighting that a number of cases treated as for tuberculosis later diagnosed as melioidosis affecting various organs including lung, joints, spine, lymph nodes, spleen and pericardium [8]. A report by P V Kingsley., *et al*. stated that the volume of melioidosis cases expected form Indian subcontinent is higher than reported, possibly due to under-recognition [2].

Our patient presented with recurrent parotid abscess which was considered as tuberculous in origin due to histopathological evidence of suppurative granulomatous inflammation though microbiological proof was unavailable. Though microbiological identification of Mycobacterium tuberculosis bacilli is required for definitive diagnosis of tuberculosis, it is often impractical especially in cases of extrapulmonary disease. Histopathological evidence of granulomatous inflammation often provides valuable supportive information for diagnosis of extrapulmonary tuberculosis. Therefore, it is common to empirically treat for tuberculosis as clinically diagnosed cases in countries like Sri Lanka where it is endemic, in situations where clinical manifestations are compatible with tuberculosis following exclusion of other possible aetiologies. However, it is not unusual that the diagnosis of tuberculosis is revised due to poor clinical response and proper culprit is recognized later following further investigations. Similar to our case, in the series of 22 patients reported by Vidyalakshmi., *et al*. one case of parotid abscess melioidosis had been mistaken for tuberculosis [8].

Isolation of *Burkhloderia pseudomallei* is considered as the 'gold standard' for diagnosis of melioidosis [2,9]. However, culture often takes time to yield results and has low diagnostic sensitivity [9]. Though *Burkhloderia pseudomallei* can be easily cultured, it can be under-recognized and mistaken for Pseudomonas species especially in non-endemic countries unless high degree of suspicion is maintained [9,10]. Therefore, serological markers have recently gained popularity for expedition of diagnosis [9].

Melioidosis has preponderance for males, which could be due to higher potential for exposure due to occupational and recreational activities. Peak incidence is between 40 - 60 years of age, where the most co-morbidities develop. Diabetes mellitus is considered as the most common co-morbidity associated with melioidosis, though people with chronic lung, renal, liver diseases and malignancies are at higher risk of acquiring the infection [2]. Age, male sex and long standing diabetes were the identifiable risk factors in our patient.

Acquisition of *Burkhloderia pseudomallei* through inoculation or inhalation usually occurs following exposure to soil and muddy surface water [2]. Therefore, history of high risk exposures should be elicited during clinical evaluation of suspected cases. But our patient denied any significant exposure to such environment. However, it should be emphasised that the absence of exposure to soil or contaminated water or evidence of portal of entry via the skin do not rule out the disease [2]. Rarely cases of hospital and laboratory acquired melioidosis have been reported [11,12]. Since our patient was a nursing officer at a surgical ward, healthcare acquired melioidosis should be considered due to the absence of definite high risk exposure, though confirmation of this hypothesis would be extremely difficult.

Conclusion

Melioidosis is an infection with diverse clinical manifestations mimicking many other common infections like tuberculosis especially in countries where tuberculosis is endemic. Accurate differentiation will be limited by limited resources and expertise. Melioidosis has recently being reported in increasing incidence in Sri Lanka. Therefore, high degree of suspicion for melioidosis should be maintained during diagnostic evaluation and follow up of unproven cases of tuberculous to avoid misdiagnosis and mistreatment.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal

Competing Interest

The authors declare that they have no competing interest.

Authors' contribution

DM made the clinical diagnosis and supervised the manuscript drafting. AB, AGRM, SAL and MRAMR drafted the first manuscript, reviewed the literature and involved in direct management of the patient. All authors read and approved the final manuscript.

Authors' information

DM (MD, FRCP, FCCP is a consultant respiratory physicians at Teaching hospital- Kandy. AB (MD, MRCP) and SAL (MD) are senior registrars in respiratory medicine. AGRM (MBBS) is a registrar in medicine, MRAMR is a medical officer.

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