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Disseminated septicaemic melioidosis: an unusual presentation of masticator space infection

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Abstract
Melioidosis is an infectious disease caused by a saprophytic bacterium, *Burkholderia pseudomallei*. It is endemic to Southeast Asia and Northern Australia. The spectrum of melioidosis in humans varies from sub-clinical to overwhelming protean manifestations resembling other acute and chronic bacterial infections. Disseminated septicaemia melioidosis presenting as a masticator space infection is reported here. This is germane to those treating diabetic patients with deep neck infections living in, or having visited, areas endemic for *B. pseudomallei*.

Key words: Melioidosis; Neck; Diabetes Mellitus

Introduction

Melioidosis is an infection caused by *Burkholderia pseudomallei*; an organism widely distributed in the soil and water of the tropics. The disease is endemic to Southeast Asia and Northern Australia but is often missed because facilities for diagnostic microbiology are scarce[1] and in the West it could be missed for the lack of familiarity. Common serious infections reported include acute pneumonitis with variable radiographic findings and septicaemia.[3,4] In Northeast Thailand, melioidosis is a major cause of community-acquired septicaemia (20 per cent).[2,3] Less severe forms present as localized infections of the skin, joints, pleural spaces, lymph nodes, liver, spleen, kidneys, pericardium, urogenital organs and muscles.[4] We report a case of disseminated septicaemic melioidosis which presented as a masticator space infection.

Case report

A 33-year-old female presented with a high-grade fever, cheek swelling and trismus after having had a tooth extracted four weeks earlier. She was a diabetic controlled with an oral hypoglycaemic drug. Aspiration of the swollen soft tissue of the left cheek yielded no pus. The computer tomography (CT) of the masticator space is shown in Figure 1. She was admitted for blood cultures (three specimens) and parenteral antibiotic treatment – penicillin to start. The blood culture of the first specimen grew *B. pseudomallei*. The serologic test revealed an indirect haemagglutination antibody (IHA) titer of 1:640. Abdominal ultrasound was performed and a small splenic abscess was detected at the lower pole of the spleen.

The antibiotic was changed to ceftazidime (100 mg/kg/day) and co-trimoxazole (trimethoprim-sulfamethoxazole) [TMP-SMZ], 8 and 40 mg/kg/day. A few days after the change in antibiotic, the patient's clinical condition improved remarkably and the fever abated. She continued under parenteral antibiotics for two more weeks and was then put on a 12-week maintenance therapy comprising oral co-trimoxazole and doxycycline.

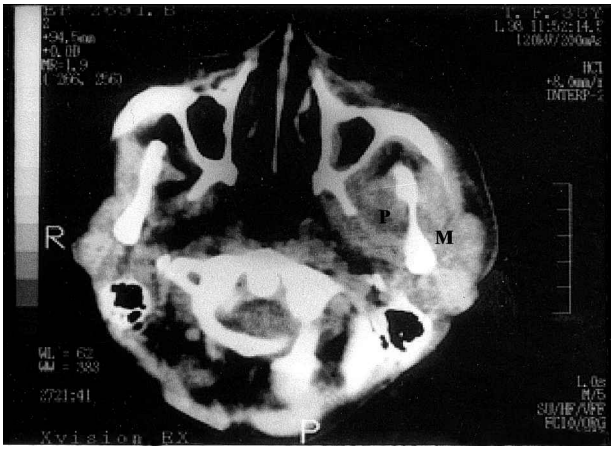

Fig. 1
Computed tomography scan in the axial plane demonstrates generalized swelling of the masseter muscle (M) and pterygoid muscle (P).

Discussion

Melioidosis was first described in Rangoon by Whitmore and Krishnaswamy in 1912.[5] The spectrum of melioidosis in humans varies from the sub-clinical to overwhelming protean manifestations resembling other acute and chronic bacterial infections.[4] Melioidosis has been reported in the parapharyngeal space.[6] Disseminated septicaemic melioidosis presenting as a masticator space infection, has not been reported previously.

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A definitive diagnosis of active melioidosis can be determined by specimen culture but only suggested by positive serology.[4] *B. pseudomallei* is a small, motile, Gram negative, obligately aerobic, non-spore-forming bacillus that shows characteristic bipolar staining on culture media.[4] Of the various serologic methods currently available, IHA has received the most attention because of its simplicity and low cost.[7] In Thailand, the diagnosis of active melioidosis uses a cut-off IHA titre of ⩾1:80, yielding a sensitivity of 90 per cent and a specificity of 88 per cent.[8]

Our patient came from an endemic area and had diabetes – the most common underlying disease associated with melioidosis.[9] Her blood culture yielded *B. pseudomallei* – the serologic test titre was 1:640. This finding suggests the possible mode of infection to the masticator space was from haematogenous spread precipitated by the tooth extraction.

Melioidosis is a serious infection with a high rate of relapse.[10] Before the introduction of ceftazidime, the conventional treatment for melioidosis was a combination of intravenous chloramphenicol, tetracycline and trimethoprim-sulfamethoxazole (TMP-SMZ). The mortality associated with severe melioidosis treated with the four drug combination was >70 per cent. The addition of ceftazidime reduced the mortality to 50 per cent among septicaemic patients surviving at least 48 hours.[11] Following the use of intravenous ceftazidime, these four drugs are still needed for a 10- to 18-week oral maintenance follow-up regime.[12]

Although melioidosis is a major cause of community-acquired septicaemia in Northeast Thailand, the clinical manifestation of a masticator space infection is rare. In our patient, a predisposing factor was likely her underlying diabetes mellitus, while associated factors included her being from an endemic area and having a dental infection following an extraction.

Melioidosis is a serious infection, but with early diagnosis and appropriate use of antibiotics morbidity and mortality are reduced. Melioidosis should be suspected in the diabetic patient with a deep neck infection, especially if the patient comes from (or has been visiting) an endemic area. Notwithstanding the longstanding awareness of melioidosis, the fact of extensive travel even by those with underlying diseases such as diabetes mellitus makes review of this presentation relevant.

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