

Temporomandibular joint septic arthritis: A rare manifestation of melioidosis

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Melioidosis is a community-acquired infectious disease that is caused by the Gram-negative bacillus, *Burkholderia pseudomallei*. It is endemic in Southeast Asia countries, and its clinical spectrum is broad, often mimicking other illnesses, which make diagnosis challenging. The hallmark of its presentation is formation of abscess most commonly in the lungs. The overall mortality rate owing to melioidosis is extremely high especially in the bacteremia form. A 54-year-old woman presented with fever, limited mouth opening, and painful left preauricular swelling for 3 months. Premorbidly, she was well without underlying medical illnesses particularly diabetes or autoimmune disease. Local examination revealed a tender diffuse swelling at the left preauricular region. Her mouth opening was limited to two fingerbreadths. Hematological test showed raised total white cells, erythrocyte sedimentation rate and C-reactive protein. Computed tomography and MRI showed presence of significant left masseteric collection with erosion of left temporomandibular condyle. Other incidental findings from the imaging were multiple liver abscesses and right lower lung abscess. Her indirect immunofluorescence enzyme-linked immunosorbent assay and enzyme-linked immunosorbent assay diagnostic test was positive with significant titre ratio of 1 : 320, which established the diagnosis of melioidosis. Full recovery was attained following surgical drainage of the abscess, together with antibiotics for 6 months (intravenous ceftazidime for a month, followed by oral augmentin for 5 months). Melioidosis involving the TMJ joint is a rare entity. This is the second reported case of melioidosis affecting TMJ joint worldwide, from extensive literature search. This case highlights the importance of suspecting melioidosis in individual presenting with abscess at uncommon and atypical site. Enzyme-linked immunosorbent assay is a rapid test as compared to the gold standard blood culture, and it helps in reaching the diagnosis early with its advantages of high sensitivity and specificity.

Keywords:

enzyme-linked immunosorbent assay, melioidosis, temporomandibular joint

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Introduction

Melioidosis is a community-acquired infectious disease that is caused by the Gram-negative bacillus, *Burkholderia pseudomallei*. It is endemic in Southeast Asia countries and its clinical spectrum is broad often mimicking other illnesses, which make diagnosis challenging. The hallmark of its presentation is formation of abscess most commonly in the lungs. The overall mortality rate owing to melioidosis is extremely high especially in the bacteremia form.

Case report

A 54-year-old woman presented with fever, limited mouth opening, and painful left preauricular swelling for three months. Premorbidly, she was well without underlying medical illnesses particularly diabetes or autoimmune disease. Local examination revealed a tender diffuse swelling at the left preauricular

region. Her mouth opening was limited to two fingerbreadths. Hematological test showed raised total white cells, erythrocyte sedimentation rate, and C-reactive protein. Computed tomography (Figs 1 and 2) and MRI (Fig. 3) showed presence of significant left masseteric collection with erosion of left temporomandibular (TMJ) condyle. Other incidental findings from the imaging were multiple liver abscesses and right lower lung abscess. Her indirect immunofluorescence enzyme-linked immunosorbent assay and enzyme-linked

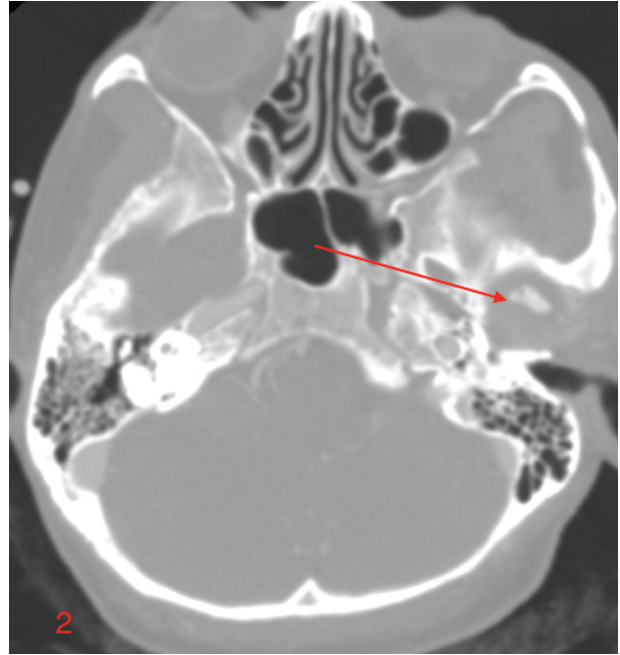
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Figure 1



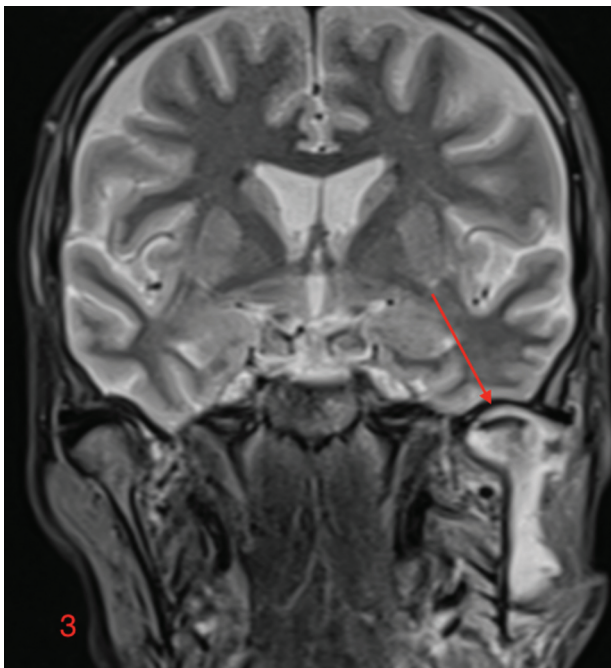
Computed tomography scan showed a collection of abscess at the left temporomandibular joint (arrow).

Figure 2



Computed tomography scan (bone window) showed severe destruction of left temporomandibular joint (arrow).

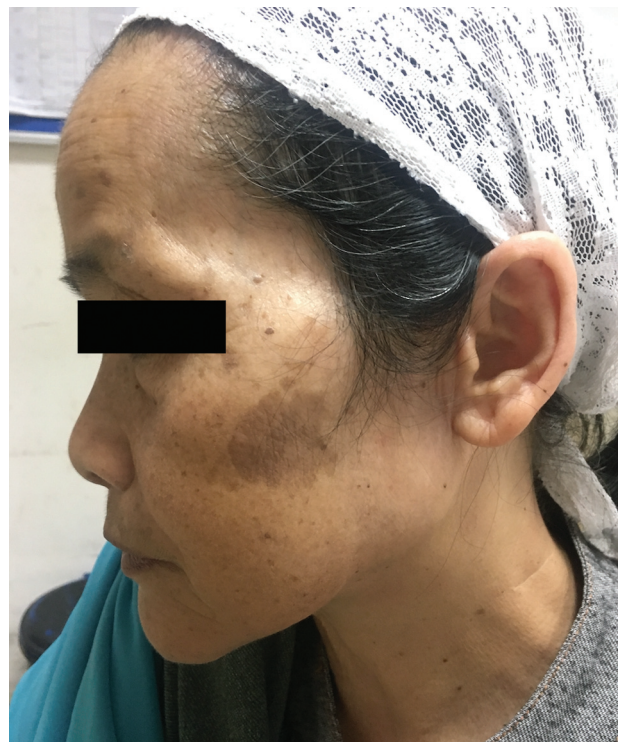
Figure 3



MRI showed inflammation and destruction of left temporomandibular joint (arrow).

immunosorbent assay (ELISA) diagnostic test was positive with significant titer ratio of 1 : 320, which established the diagnosis of melioidosis. Full recovery was attained following surgical drainage of the abscess, together with antibiotics for 6 months (intravenous ceftazidime for a month, followed by oral augmentin for 5 months). She recovered fully without any trismus (Fig. 4).

Figure 4



Picture showed total resolution of the disease after completion of antibiotic course.

Discussion

Septic arthritis of TMJ is an uncommon reported entity in adults. Most common pathogens have been

Staphylococcus aureus, *Haemophilus influenzae*, and *Streptococcus* spp. Major route of spread is usually by hematogenous dissemination from a distant focus. Direct extension from contiguous sites and direct inoculation of microorganisms from penetrating wound are other alternative pathways [1].

Abscess formation is the trademark of *B. pseudomallei* infection. In our report, we believed that hematogenous dissemination and direct spread from adjacent masseteric muscle abscess is the most plausible explanation for the TMJ melioidosis. The clinical presentation of TMJ melioidosis includes preauricular swelling, pain, and other signs of inflammation, which are not different from that owing to common bacterial pathogens. The diagnosis of melioidosis is always challenging owing to its variable manifestations. It is a systemic disease, which affects any visceral organ, and which imitates the great masquerader – tuberculosis [2].

Bacterial identification by culture is accepted as the gold standard. However, its main drawback is that it takes at least 48–72 h to obtain result, which maybe too late for successful treatment, as a high percentage of patients died within 24 h of admission for acute septicemia related to melioidosis [3]. Hence, serological test such as immunofluorescent antibody test (IFAT) and ELISA are effective tools in supporting the diagnosis especially with the presence of other clinical features such as pneumonia, multiple liver, and other soft tissue abscesses [4]. A single high antibody titer is adequate to support the diagnosis of acute melioidosis from ELISA test. Besides, serology test shows high sensitivity and specificity for as high as 92 and 90% [5]. In comparison with bacterial culture as 'gold standard' tool for diagnosis, it carries low

sensitivity (60%) [6]. This could be explained in our report where she was given a course of augmentin before admission. Bacterial load might be reduced leading to a negative blood and pus culture result.

Conclusion

Melioidosis involving the TMJ joint is a rare entity. This is the second reported case of melioidosis affecting TMJ joint worldwide, from extensive literature search. This case highlights the importance of suspecting melioidosis in individual presenting with abscess at uncommon and atypical site. ELISA is a rapid test as compared with the gold standard blood culture, and it helps in reaching the diagnosis early, with its advantages of high sensitivity and specificity.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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