Infected Submandibular lump: Expect the unexpected

Abstract
Melioidosis is an emerging infectious disease, caused by the gram-negative saprophyte Burkholderia pseudomallei. This paper reports an atypical presentation of Melioidosis, which had the submandibular lymph node as the only 'silent' focus of infection, which was misdiagnosed as an abscess.

Key words: Melioidosis; Submandibular Lump; Coastal India; Burkholderia Pseudomallei

Introduction
Melioidosis is an endemic infectious disease in South East Asia, North Australia and temperate regions bordering the equator. Recently an increasing number of cases are being reported from many parts of the globe every year, including the coastal belt of southern Karnataka, India.(1-3) The causative organism Burkholderia pseudomallei is a motile, gram-negative bacillus, transmitted by direct contact, cutaneous inoculation or inhalation.(4, 5) The disease presentation varies from asymptomatic to acute overwhelming fatal conditions.(1, 4, 5) This paper reports a case of submandibular abscess which was misdiagnosed as an abscess.

Case Report
A 39 year old patient reported to our dental outpatient clinic with a history of progressive swelling of the right lower jaw for last 2 months. There was no history of fever or weight loss. Past dental history revealed lower right quadrant tooth extraction at a local dental clinic two months back. Palpation of the lesion showed a firm movable swelling measuring 2cm x 2cm. It extended posteriorly from the left angle of mandible to anterior border of masseters anteriorly and inferiorly to the submandibular region. Temperomandibular joint on palpation was normal. The submandibular lymph nodes were palpable. Investigations showed ESR of 57 mm/hr with normal leucocyte counts. Case was provisionally diagnosed as an abscess or bacterial sialadenitis, or a dental abscess. Incision and drainage was done and 1.5ml of pus was aspirated. A prescription oral ampicillin and metronidazole were given. Patient was reported back with persistent swelling and pain on the same side after one week. Ultrasound of the neck revealed a necrotic lesion measuring 3.1cm x 1.3cm in close relation to right submandibular gland and multiple hypoechoic lesions in the upper midjugular chain bilaterally and similar solitary necrotic lesion measuring 1.6cm x 1.3cm on posterior aspect of right gland (Figure 1). This was followed by FNAC of the submandibular gland, which confirmed a suppurative lesion. Swelling was aspirated for the second time and aspiration fluid was sent for culture and sensitivity test. There were moderate pus cells and no bacteria could be seen in the direct gram stain of the smear. Culture next day showed the growth of dry, wrinkled pink coloured colonies, which was biochemically identified and API-GN (Biomeriux, France) confirmed as Burkholderia pseudomallei. It was sensitive to amoxicillin-clavulanic acid, ceftazidime, co-trimoxazole and doxycycline and resistant to gentamicin, ciprofloxacin. With the culture results, patient was referred to medical care. The CT scans of the neck showed level II and level III inflammatory lymphadenopathy with suppuration in both submandibular lymph nodes (Figure 2). Treatment with parenteral antibiotics started with Ceftazidime 2g IV 6 hourly for 14 days. Blood culture remained sterile throughout and abdomen ultrasound scan was normal. Six days after the treatment, left submandibular lymphadenopathy completely subsided and right submandibular lymph node decreased to 1.5cm x 1cm with the clinical recovery. Patient was discharged with cotrimoxazole (800/160) mg and doxycycline 100 mg BD for 18 weeks.

Discussion
A majority of uncomplicated dental infections in India get treated without the aid of
cultures as was this case. With the progressive increase of the swelling not responding to the treatment in our patient, and the significant radiological findings on the neck ultrasound, the abscess fluid was sent for microbiology, which grew Burkholderia pseudomallei. Though, Burkholderia pseudomallei is an easily cultivable organism by 48 hours in laboratory, it requires expertise to identify and recommend the treatment. The usual therapeutic option for chronic submandibular abscess might be ineffective in most occasions when the isolate is B. pseudomallei. Inappropriate and inadequate treatment may give rise to relapse as well as fatal outcome. Patient received intensive phase treatment with ceftazidime for 2 weeks and eradication phase with doxycycline and cotrimoxazole combination for 18 weeks and made a complete recovery with no fresh complaints till last follow up. This case was unique in patient not having any predisposing risk factors, and having a mild submandibular node as the only ‘silent’ focus of infection without any episodes of bacteremia or septicemia. Internal organ involvement and bacteremia is commonly associated with such cases and blood cultures and ultrasound abdomen were normal in our patient suggesting a localized disease. Environmental factors like surface water, soil, and so on have been found as the sources of transmission of melioidosis. We predict the usage of mud twigs for tooth picking by the patient as the possible route of acquisition. Also, the abscess appeared after his tooth extraction at a village dental clinic and a possibility of breach in sterile precautions cannot be ruled out. Any similar presentation should always be supported by the microbiological opinion since delay in diagnosis burdens the patient with unnecessary investigations and inadequate treatment.

**Conclusion**

This case is a paradigm for the practicing dentists to be watchful and consider melioidosis in the differential diagnosis of soft-tissue infections, especially encountered in this part of Southwestern coastal Karnataka of India. So, irrespective of the severity of presenting symptoms, prompt microbiological help is necessary to arrive at the diagnosis. With the associated invasiveness of the pathogen, a complete evaluation of foci or dissemination elsewhere in the body becomes essential.

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