

MELIOIDOSIS PRESENTING AS PAROTID ABSCESS IN AN ADULT - A CASE REPORT

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Abstract

Burkholderia pseudomallei, a facultative intracellular Gram-negative bacterium causes melioidosis, a tropical disease which is more commonly reported these days. Melioidosis can present as visceral abscess, pneumonia, localized ulcers, musculoskeletal and neurological infections, even as full blown sepsis. Melioidosis presenting as parotid abscess is commonly reported among pediatric population, however in adults it is a rare entity with only very few cases being reported. There are very limited studies that have described melioidosis with abscess in unusual sites apart from liver and splenic abscess. Here we present a case of a diabetic male with left sided parotid abscess and on microbiological evaluation found to have melioidosis which was successfully treated with combined surgical and medical treatment.

Keywords: Melioidosis, Parotid abscess, *Burkholderia pseudomallei*

Introduction

Melioidosis is a tropical disease caused by facultative intracellular Gram-negative bacillus *Burkholderia pseudomallei*. The bacterium was first isolated by Captain A Whitmore, in 1911 at Rangoon General Hospital, Burma, when treating a young boy with pneumonia.^[1] The term Melioidosis was coined by Stanton and Fletcher in 1921.^[2] In the recent years the prevalence of melioidosis is widespread in India, with many cases being reported from West Bengal, Bihar, Tamil Nadu, Orissa, Maharashtra, Kerala and Tirupura.^[3] The number of cases of melioidosis reported in India is considered to be the tip of an iceberg as several cases may not be diagnosed due to lack of awareness among the clinicians about the infection and the laboratories not capable of isolating and identifying *Burkholderia pseudomallei*.^[4] In endemic areas, the parotid gland involvement is seen in around one third of paediatric patients affected with melioidosis, but it is rare in adults with very few reported cases.

Case Report

This 53- year- old gentleman who was a newly diagnosed with uncontrolled diabetes was admitted with the complaints of fever for 25 days duration. He also had complained of swelling of the left side of the face and neck associated with difficulty in opening the mouth for 10 days duration. He was initially evaluated at his home town hospital, as was clinically diagnosed as mumps and suggested conservative management. However in the view of persistent fever, he was referred to our hospital for further management. On clinical examination, he was febrile (temperature 102 F). He also had an enlarged and

tender left parotid gland. (Figure 1) Complete blood counts showed neutrophilic leucocytosis

(Neutrophils-90%), White Blood count (WBC) was 18,500 cu.mm and Erythrocyte Sedimentation Rate (ESR) was 80 mm per hour. His random blood sugar was 452 mg/dl and HbA1c was 12.6%. Contrast Enhanced Computed Tomography (CECT) Neck showed ill-defined soft tissue swelling with enhancement and areas of necrosis within the left pterygopalatine fossa and extending to the parapharyngeal space. It measured 2.0 x 3.4 x 3cm in size (Figure 2). There was diffuse swelling of the left temporalis, pterygoid and masseter muscles with mild mass effect on the nasopharyngeal mucosa. The left parotid appeared mildly bulky with multiple intra parotid nodes. One of these in the superficial lobe measuring 1.5 cm showed areas of necrosis within and few sub-centimetre lymph nodes were noted at level II on both sides, more on the left side (Figure 3). Blood cultures were sent and he was started on intravenous ceftazidime. For glycemic control he was placed on a basal bolus insulin regimen along with oral anti-diabetic drugs.



Figure 1: Left sided Parotid swelling

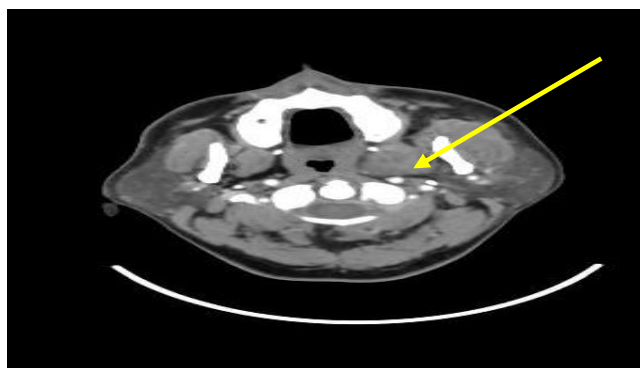


Figure 2: CECT Neck showing left pterygopalatine fossa and parapharyngeal soft tissue swelling



Figure 3: CECT Neck showing left intraparotid necrotic node

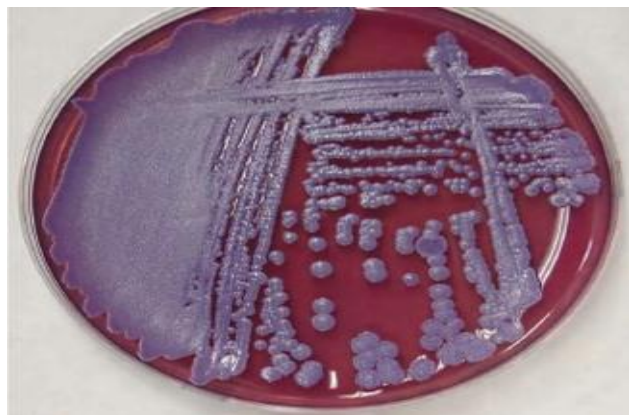


Figure 4: *Burkholderia pseudomallei* colony morphology as demonstrated on Ashdown's selective medium

Eye Nose Throat (ENT) consultation was sought in the view of source reduction of the left sided parotid abscess. Left infratemporal fossa exploration was done by transparotid approach. 20 ml of pus was drained from the abscess and it was sent for microbiological examination. Left lower parts of superficial and deep lobe were excised and deep lobe was sent for histo-pathological analysis. Both tissue culture and blood cultures grew *Burkholderia pseudomallei*, which was sensitive to ceftazidime. Histopathological examination revealed chronic

inflammation. He was given intravenous ceftazidime 6 gram per day in three divided doses for 2 weeks along with co-trimoxazole (320mg+1600mg) in two divided doses for 6 months. Patient improved significantly and got discharged in a stable condition.

Discussion

Melioidosis is caused by *Burkholderia pseudomallei*, a Gram-negative facultative intracellular saprophytic bacterium, commonly living in soil or contaminated stagnant water.^[5] It was earlier called as *Pseudomonas pseudomallei*. It is endemic of Southeast Asia and Northern Australia regions and is being increasingly reported in other tropical areas of the world such as India, China, and Brazil.^[6,7] Infection with *Burkholderia pseudomallei* is acquired through several routes, which includes subcutaneous inoculation, ingestion, and aspiration of contaminated water during near-drowning episodes and laboratory-acquired infection.^[8] Melioidosis is known as a 'remarkable imitator' since it mimics several diseases from pyogenic bacterial infection to tuberculosis, and there is no pathognomonic sign of melioidosis.^[9-11] Melioidosis manifests as inapparent infection, acute localized suppurative infection, acute pulmonary infection, acute septicemia infection or chronic suppurative infection. Risk factors for the infection include diabetes mellitus, alcoholism, renal insufficiency, chronic liver disease, malignancy, connective tissue disorders and chronic steroid use. Diabetes mellitus is the single most common risk factor which is seen in around 23%-60% of the patients.^[12] Our patient was also a diabetic, who remained undiagnosed prior to this and presented to us with very high blood sugars and alarmingly high HbA1c.

The disease can affect any organ in the body and commonly presents as a pulmonary infection and/or multiple visceral abscesses. Although melioidosis involves most organs, parotid involvement is rare especially in adults. The most common presentation is pneumonia occurring in around 30% of the cases.^[13] According to Tipre M et al study, they identified a total of 99 cases published between 1953 and June 2016, originating from India (n=85) and Bangladesh (n=14). Soft tissue abscess (37%) was the most common clinical presentation reported from India followed by pneumonia (24%) and osteomyelitis/septic arthritis (18%). Neurological melioidosis (12%) presented as pyemic lesions of the brain or meninges. 4 cases prostatic abscess in men and 4 cases of parotid abscess were also noted. All of the parotid gland abscesses occurred among females, two of whom were pediatric cases aged 3 and 12 years.^[14] It is possible that the proportion of parotid abscess in India may be due to exposure to *Burkholderia pseudomallei* through ingestion of unchlorinated drinking water. A majority of rural population in India have limited access to chlorinated

drinking water which may increase their risk of contracting melioidosis.^[15]

Complication of melioidosis parotitis includes abscess formation, spontaneous rupture into the auditory canal, facial nerve palsy, septicæmia, osteomyelitis and necrotizing fasciitis.^[16,17] Isolation of *Burkholderia pseudomallei* from clinical specimen culture remains the gold standard for diagnosis^[12] and it grows readily in routine laboratory culture media such as blood agar and MacConkey agar. The use of selective media like modified Ashdown medium (with colistin), B pseudomallei selective agar, and Burkholderia cepacia selective agar is also recommended.^[18] It is oxidase positive, and hence it is often misreported as pseudomonas species. A high index of suspicion and awareness is essential among laboratory staff regarding the colony morphology and the culture characteristics of the organism is warranted as early diagnosis is pivotal to avoid life threatening complications, as delayed diagnosis leads to septic shock, which has a case fatality rate of around 20% -50%.^[19]

High-dose intravenous ceftazidime is currently the first antibiotic of choice for the recommended treatment of acute melioidosis. Treatment generally starts with intravenous Ceftazidime 50 mg/kg three times a day for 10-14 days, followed by 3-6 months of trimethoprim /sulfamethaxazole (co-trimoxazole) 160/800 mg twice a day during eradication phase. Switching to meropenem is indicated if patient's condition worsens while receiving ceftazidime, e.g. organ failure, development of a new focus of infection during treatment or if repeat blood cultures remain positive.^[20] With appropriate surgical and medical therapy, the prognosis of this condition is good, which contrasts with the high mortality rate seen if septicæmia develops. Patient follow up is extremely important as relapse rates are high if the patient is less compliant during the eradication phase.

Conclusion

Most clinicians are not aware about the clinical presentation of melioidosis; therefore, it is often misdiagnosed or under-diagnosed. Physicians should suspect melioidosis if any febrile patient presenting with multiple abscesses with risk factors like uncontrolled diabetes, with history of travel to or living in melioidosis endemic area, especially when they do not respond to conventional antibiotics. This case is being reported in view of its rarity of presentation. This case highlights the importance of high index of suspicion of melioidosis in any fever of unknown origin or skin and soft tissue infection, especially parotid abscess. Microbiologists and laboratory technicians should be adequately trained so that they can

identify *Burkholderia* species correctly, as it is often misreported as *Pseudomonas* species. Best clinical judgment and focused microbiological investigations are very important for early diagnosis and early treatment thereby resulting in better prognosis.

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