

## Unmasking Parotid Melioidosis: A Clinical Enigma That Defies Occam's Razor

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### ABSTRACT

Melioidosis aka Whitmore disease is a rare emerging infection of gram-negative bacterium *Burkholderia pseudomallei* is an endemic in South-east Asia but is mostly under-diagnosed and under-reported in the Indian subcontinent. We report a case series of cervical melioidosis in patients with comorbidities. This study was undertaken to analyze the epidemiological risk factors, clinical manifestations and outcomes of melioidosis in India in a tertiary health care center during the course of 2 years (2021 – 2023).

### 1. INTRODUCTION

Melioidosis, also known as Whitmore's disease, is an emerging but often under-recognized infectious disease caused by the gram-negative bacillus *Burkholderia pseudomallei*. First described by Alfred Whitmore and Krishnaswami in Burma (now Myanmar) in 1912 as a "Glanders-like" illness, melioidosis has since been identified as a significant but underestimated health concern in tropical and subtropical regions. The causative organism is a facultative intracellular, motile, aerobic, non-spore-forming, and non-fermenting bacillus that exists as a saprophyte in moist soil, stagnant water, and rice paddies. [1,2]

*B. pseudomallei* infection occurs via percutaneous inoculation, inhalation, or ingestion of contaminated soil or water. The disease is considered endemic in parts of South and Southeast Asia, Northern Australia, Southern China, and Hong Kong, with sporadic but increasing reports from India, where it is likely under-diagnosed due to limited awareness and diagnostic infrastructure. [2,3]

Melioidosis carries a high mortality rate, estimated at up to 40%, especially in patients with underlying conditions such as diabetes mellitus, chronic kidney disease, chronic liver disease, thalassemia, malignancies, alcoholism, and non-HIV-related immunosuppression. [4] Clinically, it is known for its diverse manifestations, most commonly presenting as pneumonia, cutaneous abscesses, genitourinary infections, bacteremia, and occasionally pericarditis or neurological involvement. Importantly, *B. pseudomallei* has been designated a Tier 1 Select Agent by the U.S. Centers for Disease Control and Prevention (CDC) due to its potential use as a bioterrorism agent. [5]

This case series highlights the clinical profiles, risk factors, diagnostic work-up, management, and outcomes of cervical melioidosis, particularly involving the parotid region, in patients with underlying comorbidities treated at a tertiary care hospital in India over a two-year period (2021–2023). It aims to increase clinical suspicion and encourage early diagnosis of this rare but serious infection in endemic settings.

**CASE REPORT 1:** A 37-year-old male with a six-year history of poorly controlled Type 2 Diabetes Mellitus presented with left-sided facial swelling and pain persisting for 20 days. The swelling was insidious in onset and gradually progressive, accompanied by moderate-grade fever with chills and rigors, odynophagia, and reduced salivation.

On examination there was a diffuse swelling over the left parotid region measuring 10 x 8 cm with the upper border from the level of the anterior surface of the left side of the mandible to 3 cm above the clavicle. Horizontally 3 cm from the midline of the left anterior triangle to posterior border of the trapezius. Redness, Tenderness, local rise in temperature, induration present. Swelling was firm in consistency with well-defined border, fluctuation present. Transillumination negative. Cardiopulmonary system and Central nervous system are within normal range. Laboratory findings were Hb 11.8 gm/Dl,

TLC  $12.98 \times 10^3$  cells/mm<sup>3</sup>, platelets  $10^3$  cells/mm<sup>3</sup>, HBA1c 12.1% g/dl, RA factor – 9.5 IU/ml with renal and liver function tests with normal range.

Ultra-sound of the local parts showed a bulky parotid gland with diffuse altered echotexture and vascularity. Large thickened collection inferior end and closely abutting the parotid gland measuring 3.3 x 2.8 x 2.4 cm with normal appearing neck vessels.

CT shows ill-defined necrotic peripheral enhancing collections in the left side involving the sternocleidomastoid muscle with extension 5.7 x 4x 6.1 cm anteriorly abutting the submandibular gland and superficial lobe of left parotid gland, sternocleidomastoid posteriorly, medially abutting the carotid vessels and internal Jugular Vein, laterally extending up to the deep fascia and superiorly up to the mastoid tip with adjacent fat stranding.

Incision and drainage was done and mucopurulent discharge was sent for culture and sensitivity which was suggestive of *Burkholderia pseudomallei*.

Histopathology report was suggestive of chronic inflammatory lesion consistent with abscess. Patient was started on Inj. Cefotaxime 2g for 21 days. Complete resolution of swelling was documented at the end of hospital stay. On serial follow up basis, patient showed no evidence of recurrence.



Figure 1 : gram-stain of *b pseudomallei* isolate vb976100, phase contrast microscopy with a 100 x oil immersion objective

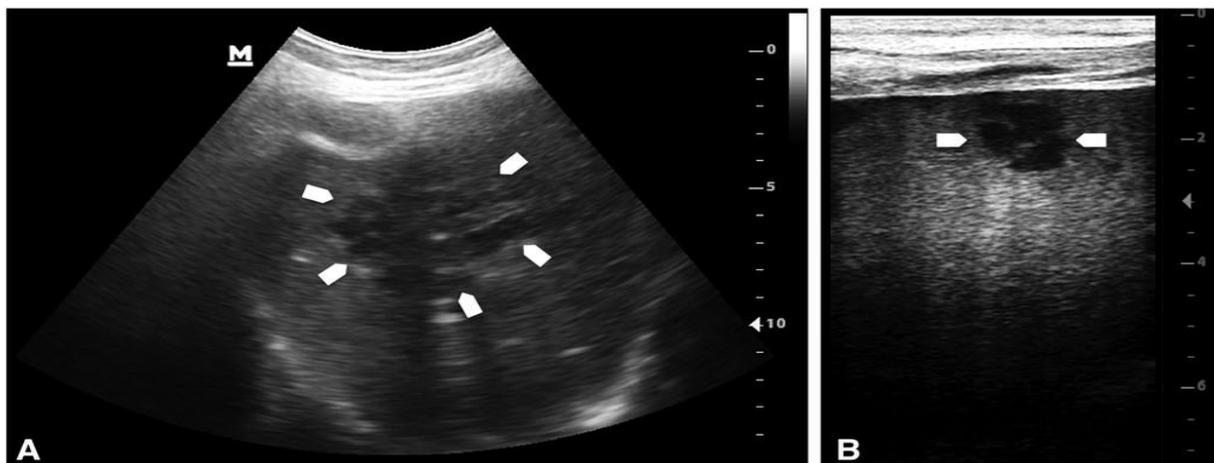


Figure 2 : Hypoechoic lesions (arrows) due to melioidosis abscesses in the (A) cervical lymph node and (B) spleen. The hypoechoic lesions in the cervical lymph node and spleen were frequently septated and/or localized in clusters which may suggest “honeycomb” or “necklace” appearance.

## CASE REPORT 2

A 40 year male with uncontrolled diabetes came with swelling over the right side of the cheek since 15 days with associated throbbing type of pain, intermittent on and off fever with chills and rigors with odynophagia and weight loss. Previous history of typhoid 5 months back.

On examination a solitary 12 x 10 cm diffuse swelling over the right side of the parotid and lower jaw was noted. Local rise in temperature, tenderness, with a smooth surface and firm in consistency with induration and fluctuation and transillumination negative. Levels Ib, II, III lymph nodes felt- tender and mobile. Carotid pulsations felt. Laboratory values showed HbA1c- 8.7 gm/dL, Hb 16.5 gm/dl , TC  $12 \times 10^3$  cells/mm<sup>3</sup>, platelets  $527 \times 10^3$  cells/mm<sup>3</sup>, s. Sodium 127 mmol, renal and liver function with normal limits.

USG suggestive of ill-defined hypoechoic collections in intramuscular and subcutaneous plane of the upper aspect of the anterior triangle extending to surface measuring 1.1 x 0.4 x 0.9 cm (1cc) with lymph nodes in the Ib, II, III, IV, Va/b.

Incision and drainage was pus sent for culture and sensitivity with suggestive of *Burkholderia pseudomallei*. Inj Ceftazidime 2gm was started for 21 days. Complete resolution of swelling was documented at the end of hospital stay. On serial follow up basis, patient showed no evidence of recurrence.



**Figure 3 : clinical photo Post incision and drainage of the parotid abscess**

### **CASE REPORT 3:**

A young 32 year old woman with uncontrolled Type 2 diabetes mellitus and hypothyroidism presented with a right sided neck abscess evolving over 5 days. On clinical examination, fever (39 C) with surrounding erythema and right sided diffuse swelling over the parotid gland with tenderness and local rise in temperature and with surrounding induration. Laboratory findings included HbA1c 7.4% , elevated leucocytic counts and CRP. Culture sensitivity done .

Abscess was drained intraoperatively and cultures showed *Burkholderia pseudomallei* , Initially intravenous ceftazidime was administered for 2 weeks and was sent home with prophylactic oral antibiotics to prevent relapses. Patient responded well to therapy, wound healing was satisfactory and follow up visits were uneventful.

### **CASE REPORT 4:**

A 35 year old male with Type 2 diabetes mellitus undergoing haemodialysis since 3 years (previously a hospital staff) presented with a progressive swelling, fever and pain over the right pre-auricular region since 20 days.

He gives history of difficulty opening his mouth. On examination, a cystic, warm fluctuant swelling measuring 4x3 cm noted over the right parotid region, with tenderness and surrounding induration. Laboratory investigations showed Hb- 13.5 gm/dl , TLC –  $16.4 \times 10^9/L$  and raised ESR- 55 mm/hr and RBS- 240 and S. creatinine- 4.9. NCCT- revealed an irregular hypodense lesion of 4.5x 4.4x 2.5 cm in the right parotid region. Intraoperatively 7 ml of seropurulent fluid was aspirated and sent for cultures.

Cultures were positive for *B. pseudomallei*. Organism was sensitive to co-trimoxazole, meropenem doxycycline. Inj. Meropenem was started for 2 weeks. Complete healing of the lesion was noted after 3 months and follow up showed no further findings.

### **Case report 5:**

A 49 year old woman presented with a preauricular swelling over the left side with recurrent episodes of continuous fever and weight loss since 10 days. No cough with expectoration. No systemic symptoms. The lesion was noted over the left sided parotid region, soft to firm swelling measuring 2x3 cm, tender with thick serosanguinous discharge. Patient was febrile with BP 110/70 mmHg, tachycardia 110 beats/ min. Laboratory investigations showed Hb 8.9 gm/dl, elevated leucocytic count

and raised ESR of 80mm/ hr. discharging pus was sent for Gram stain and was suggestive Burkholderia pseudomallei sensitive to ceftazidime, co-trimoxazole, meropenem, doxycycline, and was started on Inj. Ceftazidime for 3 weeks followed by oral co-trimoxazole for 3 weeks. Oral iron therapy was started concurrently. Patient was discharged after 2 weeks. And follow up was uneventful.

## 2. DISCUSSION

Melioidosis, caused by the gram-negative bacillus *Burkholderia pseudomallei*, is an important yet frequently under-recognized infectious disease, particularly prevalent in tropical and subtropical regions such as Southeast Asia, Northern Australia, and increasingly reported across the Indian subcontinent. This facultative intracellular pathogen thrives in soil and stagnant water, making agricultural workers and individuals with frequent environmental exposure particularly vulnerable.

Epidemiologically, *B. pseudomallei* accounts for approximately 20% of community-acquired bacteremia in endemic areas, often associated with a high case fatality rate. Clinical manifestations are heterogeneous and range from acute pneumonia and septicemia to deep-seated abscess formation in organs such as the spleen, prostate, submandibular glands, cervical lymph nodes, and soft tissues. In severe cases, the disease may progress rapidly to septic shock and multi-organ dysfunction syndrome (MODS). The variability in presentation, coupled with its ability to mimic other common infections—especially tuberculosis and deep pyogenic infections—poses a significant diagnostic challenge. [6]

Predisposing risk factors that increase susceptibility to melioidosis include poorly controlled diabetes mellitus, chronic kidney disease, chronic obstructive pulmonary disease, long-term corticosteroid or immunosuppressive therapy, alcoholism, and recent travel to or residence in endemic areas. Among these, diabetes mellitus is the most frequently reported risk factor, contributing to immune dysfunction that facilitates intracellular bacterial proliferation.

From a microbiological standpoint, the organism often grows in culture as a *Pseudomonas*-like species, exhibiting resistance to aminoglycosides (e.g., gentamicin, tobramycin, streptomycin), polymyxins (colistin, polymyxin B), and first- and second-generation cephalosporins, as well as penicillin and ampicillin. This intrinsic multidrug resistance profile often leads to misidentification and delays in appropriate treatment. In fact, isolates that appear as non-lactose fermenting, oxidase-positive, motile, gram-negative rods but are resistant to standard antibiotics should raise suspicion for *B. pseudomallei*, especially in patients with compatible clinical features and risk factors.

Definitive diagnosis relies on microbial culture and biochemical identification, although newer molecular diagnostic tools have greatly improved specificity and turnaround time. These include polymerase chain reaction (PCR)-based assays, real-time multiplex PCR, and Matrix-Assisted Laser Desorption/Ionization-Time of Flight (MALDI-TOF) Mass Spectrometry, which facilitate rapid and accurate species-level identification. [7,8] Additionally, capsular polysaccharide antigen detection using lateral flow immunoassay (LFI) and monoclonal antibodies has shown high diagnostic yield, with reported sensitivity of 98.7% and specificity of 97.2%, offering a promising point-of-care (POC) diagnostic modality in low-resource settings.

In terms of treatment, *B. pseudomallei* exhibits broad-spectrum antimicrobial resistance, but remains susceptible to certain higher-generation agents. Meropenem is currently considered the first-line agent during the intensive phase due to its low minimum inhibitory concentration (MIC), effective tissue penetration, and tolerability. Imipenem and Ceftazidime are also effective alternatives. Following initial intravenous therapy (usually 2–4 weeks), the eradication phase involves oral antibiotics such as co-trimoxazole (trimethoprim-sulfamethoxazole), often combined with doxycycline, administered for 12–20 weeks to prevent relapse. [9]

The clinical mimicry, misidentification of isolates, and lack of awareness regarding melioidosis contribute to delayed diagnosis and inappropriate empirical therapy, which significantly worsen outcomes. This underscores the importance of maintaining high clinical suspicion, especially in endemic areas or in patients with compatible risk factors who present with non-resolving abscesses, unusual sepsis, or *Pseudomonas*-like isolates with unusual resistance profiles.

To address these gaps, initiatives such as the Melioidosis Registry—a web-based open-access epidemiological network—have been established to consolidate data on the burden, geographical distribution, morbidity, and mortality associated with melioidosis globally. These collaborative databases not only assist in understanding regional patterns of disease but also promote better diagnostic protocols, treatment algorithms, and public health strategies for prevention and early detection. Such studies are particularly valuable in resource-limited settings, where early recognition and appropriate treatment can significantly improve survival rates.

	Case-1	Case-2	Case - 3	Case - 4	Case – 5
Age	35 y	40 y	32 y	35 y	49 y
Sex	Male	Male	Female	Male	Female

<b>Comorbidities</b>	Diabetes mellitus	Diabetes mellitus	DM, Hypothyroidism	DM, Chronic renal failure-on mHD	
<b>Laboratory findings</b>	HbA1c- 12.1 TLC- 12.98x10 <sup>9</sup> RA factor- 9.51	HbA1c- 8.7 % TLC- elevated ESR- elevated S.sodium- 127gm	HbA1c- 7.4% TLC- elevated CRP/ESR- elevated	RBS- 230 gm/dL TLC- elevated CRP/ESR- elevated S. Cr- 4.9	HbA1c- 8.9% TLC- elevated ESR- elevated
<b>Antibiotic</b>	Inj Ceftazidime 2gm iv x 2 weeks	Inj Ceftazidime 2gm iv x 2 weeks	Inj Ceftazidime 2gm iv x 2 weeks	Inj. Meropenem 1gm iv x 2 weeks With oral Co-trimoxazole	Inj Ceftazidime 2gm iv x 2 weeks
<b>Recovery</b>	Excellent	Excellent	Good	Good	Excellent

### 3. CONCLUSION

Melioidosis is a rare entity. Clinical suspicion is very important to diagnose melioidosis, as varied presentation ranging from pneumonia, meningoencephalitis to superficial and deep abscess of the cervical lymph nodes, septicaemia and MODS are documented. Strict intensive antibiotic treatment with Meropenem or Ceftazidime for appropriate duration followed by eradication therapy for 6 months is warranted. Primary prevention of diabetes and glycaemic control also plays a major role in eradicating this illness. UV irradiation and chlorination of the ground water reduces the *Burkholderia pseudomallei* to undetectable levels. Prevention-

A: Clinical training/Sensitization of microbiologist

B: Lateral flow immunoassay, bacterial culture, molecular tests

C: Intensive treatment and eradication therapy

#### Consent:

Written informed consents was obtained from the patients for publications of this case and accompanying images.

#### Footnote:

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