

Melioidosis and tuberculosis: dual pathogens in a neck abscess

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Abstract

Introduction: Melioidosis is an infectious disease caused by a saprophytic bacterium, *Burkholderia pseudomallei*. It is endemic to Southeast Asia and Northern Australia. It may manifest as a pulmonary lesion, osteomyelitis, abscesses in soft tissue and various organs, or as septicaemia.

Case report: We report a case of a 40-year-old, diabetic man who presented with a neck lump resulting from super-infection of a tuberculosis cavity with *B pseudomallei*. The patient was successfully managed by drainage along with meticulous excision of the capsule and prolonged antibiotic and anti-tubercular treatment.

Discussion: Melioidosis may be confused diagnostically with tuberculosis, as both diseases are endemic in the same regions. Our patient was unfortunate to suffer from both endemic diseases simultaneously, perhaps representing the first such case in the world literature.

Conclusion: Increased awareness of melioidosis is important as, although the organism is easy to culture, it may be dismissed as a contaminant.

Key words: *Burkholderia Pseudomallei*; Neck; Abscess; Tuberculosis

Introduction

Burkholderia pseudomallei is distributed in the soil and water of the tropics, especially in a geographical belt extending from South India to Southeast Asia, Thailand and Northern Australia.¹ A regular inhabitant of rice paddy fields, this bacterium is transmitted via direct contact, inhalation, inoculation or ingestion.² The incubation period is not known. The single most important risk factor for developing severe melioidosis is diabetes mellitus.^{1–3} Other risk factors include thalassaemia, kidney disease and occupations at high risk of exposure (e.g. rice paddy farming). Clinically, melioidosis may present with local or systemic symptoms. Mortality rates for systemic disease are very high, but local infection is usually mild, often causing delay in seeking medical attention. Relapsed disease is more frequent with poor compliance, development of antibiotic resistance and immunosuppression.⁴ Diagnostic confusion with tuberculosis can occur, as both diseases are endemic in the same regions.

Case report

A 40-year-old, diabetic farmer visited our out-patient clinic with a three-week history of a painless, left-sided neck swelling and a foreign body sensation in his throat. He had never received regular diabetic medication. He gave no complaint of hoarseness or dysphagia. He stressed that he was not a smoker and denied alcohol abuse. He could not, however, rule out trauma sustained while working in paddy fields.

On examination, a smooth, well defined, 5 × 3 cm mass of firm to hard consistency and not fixed to underlying muscle or bone was found on the left side of the patient's neck in the region of the level III lymph nodes. Wary of malignancy,

we proceeded to full ENT investigation, which revealed a suspicious-looking bulge in the lateral wall of the patient's left pyriform fossa, with no other irregularity or obvious mass.

Computed tomography (CT) of the neck revealed a hypo-dense lesion with surrounding enhancement on contrast, extending from the level of the epiglottis to the C5 vertebral level, involving the strap muscles and causing effacement of the pyriform sinus on the left. Direct, laryngoscope-guided biopsies were taken from the specific area, and the histopathology report ruled out the presence of any malignant cells. Apart from aberrant blood glucose levels, the patient's haematological and biochemical parameters were within normal limits. When the fine needle aspiration cytology report confirmed the absence of any malignant tissue in the neck swelling, it was decided to perform an excision biopsy of the mass.

In the course of surgery, a well encapsulated lesion containing 30 ml of creamy, white pus was found. The pus was carefully drained and the capsule meticulously excised from its attachments to skin and underlying muscle. The area was explored and any further lesions were excluded.

The fibrotic capsule was sent for high power histopathological examination, and a diagnosis of tuberculous abscess was returned. Pus culture revealed the growth of *Burkholderia pseudomallei*, resistant to aminoglycosides. Significantly, acid-fast bacilli were not detected using standard staining and culture methods.

The patient was given antibiotic cover with intravenous cefuroxime, and his blood glucose levels were kept under control with human insulin. The post-operative period was uneventful. The patient was commenced on oral amoxicillin-clavulanate for the maintenance phase. Anti-tubercular therapy was commenced simultaneously.

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FIG. 1

Axial computed tomography neck scan showing a well encapsulated mass.

Discussion

Melioidosis is caused by the Gram-negative, environmental, saprophytic bacterium *Burkholderia pseudomallei*. It closely resembles the causative agent of glanders, and thus was named 'pseudoglanders' for a period.³⁻⁵ The condition was discovered in 1911 in Rangoon by Major Alfred Whitmore of the Indian Medical Service along with C S Krishnasami, and became known as Whitmore's disease.²

Serological studies in endemic areas have shown that sub-clinical infection is common.² There are many presentations, both acute and chronic. Many infections are initially sub-clinical but may result in latency and delayed manifestation, even



FIG. 2

Sagittal computed tomography neck scan, showing the neck mass.



FIG. 3

The patient's neck swelling.

after several decades. Clinical signs and symptoms include septicaemia, cavitating pneumonia, bone and soft tissue infections, disseminated abscesses, mycotic aneurysms, lymphadenitis, and childhood parotitis.^{2,5} Most patients have an underlying predisposition to infection, especially diabetes, renal disease, alcoholism or thalassaemia; however, in the largest Indian case series, 50 per cent of patients had no traditional risk factors.⁶

The diagnosis of melioidosis is usually established by culture from sterile sites. Laboratory misidentification is not uncommon, and in fact occurred in the current case because the diagnosis was not considered.⁵ Polymerase chain reaction is an emerging diagnostic tool although not yet extensively validated.⁷ A definite history of contact with soil may not be elicited as melioidosis can be dormant for many years before becoming acute. A serological test for melioidosis has been developed (using indirect haemagglutination).⁵ Imaging of the abdomen using CT or ultrasound is recommended routinely, as abscesses may not be clinically apparent and may coexist with disease elsewhere.

The treatment of melioidosis is divided into two stages: an intravenous, high intensity stage and an oral, maintenance stage to prevent recurrence.^{1,3,5} Following surgical drainage of the abscesses, the intravenous, intensive phase is

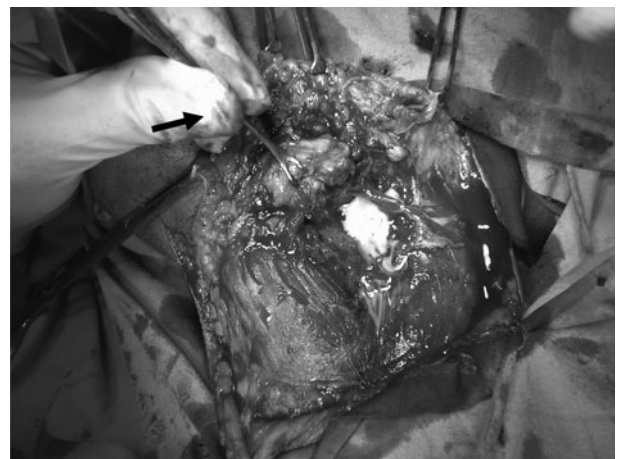


FIG. 4

Intra-operative photograph showing the abscess cavity containing pus.

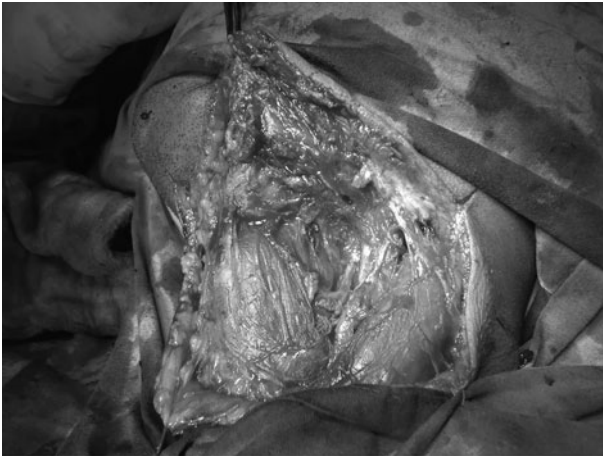


FIG. 5

Intra-operative photograph showing dissection of the fibrous capsule of the abscess.

commenced. Intravenous ceftazidime is the current drug of choice for treatment of acute melioidosis.^{2,3,5} Meropenem, imipenem and cefoperazone-sulbactam are also active. Intravenous amoxicillin-clavulanate may be used if none of the above four drugs is available, but it produces inferior outcomes.⁵ Intravenous antibiotics are given for a minimum of 10 to 14 days, and are not usually stopped until the patient's temperature has returned to normal for more than 48 hours. Following treatment of the acute disease, it is recommended that eradication treatment with co-trimoxazole and doxycycline be used for 12 to 20 weeks to reduce the rate of recurrence.^{1,5} Co-amoxiclav is an alternative for those patients who are unable to take co-trimoxazole and doxycycline (e.g. pregnant women and children under the age of 12 years).

Without access to appropriate antibiotics, the septicaemic form of melioidosis has a mortality rate that exceeds 90 per cent.^{2,5} With appropriate antibiotics, the mortality rate is about 10 per cent for uncomplicated cases but up to 80 per cent for cases with bacteraemia or severe sepsis. Relapse occurs in 10 to 20 per cent of patients. While molecular studies have established that the majority of recurrences are due to the original infecting strain, a significant proportion of recurrences (perhaps up to a quarter) in endemic areas may be due to reinfection, particularly after two years.³ Risk factors include severity of disease (patients

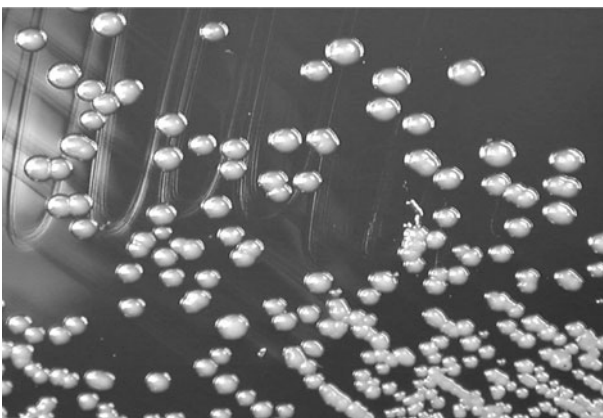


FIG. 6

Burkholderia pseudomallei on culture media.

with positive blood cultures or multifocal disease have a higher risk of relapse), choice of antibiotic for eradication therapy, poor compliance with eradication therapy and eradication therapy lasting less than eight weeks.²

In endemic areas, people (rice-paddy farmers in particular) are best advised to avoid contact with soil, mud and contaminated surface water where possible. The effectiveness of measures to reduce exposure to the causative organism has not been established. A vaccine is not yet available.⁸

- **This paper describes a case of invasion of a tuberculous cavity by *Burkholderia pseudomallei*, inoculated via vegetative trauma**
- **Most patients infected with *B pseudomallei* have an underlying predisposition to infection, especially diabetes, renal disease or any immunocompromised state**
- **Increased awareness of melioidosis is important as, although the organism is easy to culture, it may be dismissed as a contaminant**
- **Intensive treatment with intravenous ceftazidime may make the difference between life and death in patients infected with this rare but potent pathogen**

Our patient was unfortunate to suffer from two endemic diseases simultaneously, perhaps representing the first such case in the world literature. In this patient, melioidosis could have been due to super-infection of a tuberculous cavity following vegetative trauma, which would have been an ordinary event in the course of his work in the paddy fields.

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Dr V Shenoy takes responsibility for the integrity of the content of the paper.

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