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PICTURE STORY

# Spinal melioidosis: a rare presentation

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## Abstract

Melioidosis is an emerging infection endemic to tropical countries. Neurological complications of melioidosis are rarely encountered. We report a lady with diabetes mellitus, who had an infective myelopathy secondary to melioidosis. This 40-year-old lady had a clinical picture suggestive of an acute myelopathy with a thoracic (T7) sensory level. Magnetic Resonance Imaging (MRI) revealed a long-segment myelitis with ring-enhancing lesions in the lower cord. She did not respond well to conventional immunosuppression. A blood culture revealed *Burkholderia pseudomallei* with a high titre of antibodies against the same organism. Even though she was given appropriate antibiotics, she continued to have persistent residual neurological deficits. Prompt initiation of treatment with appropriate antibiotics is vital to reduce residual neurological deficits in these patients. Therefore, the requirement of a high index of suspicion to diagnose melioidosis, as well as the significance of a simple investigation such as a blood culture in the evaluation of a myelopathy is highlighted in our case.

**KEYWORDS**

Neuromeliodosis, transverse myelitis, Sri Lanka, *Burkholderia pseudomallei*

## INTRODUCTION

Melioidosis is considered as an emerging infection which is endemic to the tropics and subtropics. It commonly involves the pulmonary and the genito-urinary systems with abscess formation and septicaemia. However numerous reports have demonstrated patients with neurological complications. Among these, spinal cord involvement is a very rare occurrence.[1]

We report a female patient with diabetes mellitus (DM) who presented with a longitudinally extensive transverse myelitis (LETM), who was found to have melioidosis in the blood culture. This case is reported, due to the rarity of this clinical entity and to the diagnostic challenge faced during managing this patient.

## CASE REPORT

A 40-year-old lady, with DM presented with a one-week history of bilateral lower limb weakness. She had a recent history of aspiration of a thigh abscess. On examination she had flaccid paraparesis with a sensory level at the 7$^{th}$ thoracic (D7) vertebral plane for pain sensation. She had neither upper limb signs nor abnormalities involving her cranial nerves. She had urinary retention which required catheterisation urgently.

With the given clinical picture suggestive of a dorsal myelopathy, magnetic resonance imaging (MRI) of her spine was arranged. The initial blood investigations did not reveal an infective clinical picture. The inflammatory markers were within normal limits C reactive protein (CRP) <6mg/dL Erythrocyte sedimentation rate (ESR) - 24 mm/1st hour. The

MRI revealed a LETM extending from the lower cervical region to the conus medullaris. There was significant cord oedema and contrast imaging revealed small ring-enhancing lesions in the region of the conus medullaris (Figure 1).

Since there was no response to the initial steroid (intravenous methylprednisolone) therapy, she was commenced on therapeutic plasma exchange (TPE). Meanwhile examination of her cerebrospinal fluid (CSF) revealed an infective picture (protein – over 300 mg/dl, very low sugar and lymphocytic pleocytosis). Her Mantoux test was negative, but she was started on anti-tuberculous therapy (ATT) empirically due to the high likelihood of tuberculous myelitis.

During the TPE she developed fever spikes and blood cultures were sent to find the infective focus. The inflammatory markers were on the rise during this period. Two of the blood cultures revealed colonies of gram-negative bacilli which were resistant to both gentamicin and colistin. (Figure 2). Gram stain revealed the characteristic safety-pin appearance of *Burkholderia pseudomallei.* Direct agglutination testing of the isolates further confirmed the presence of this organism.

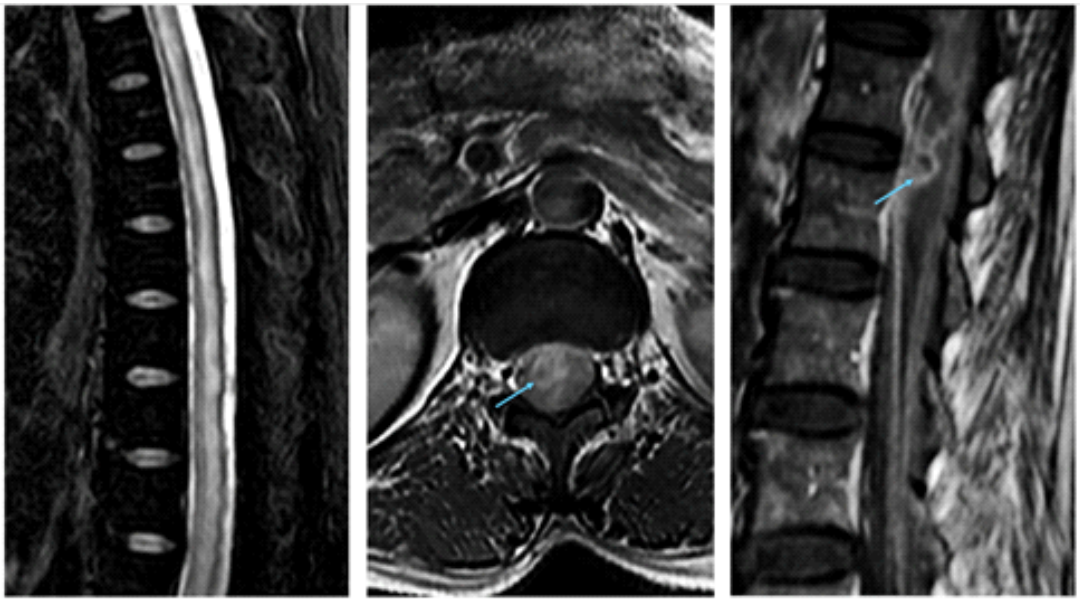


**FIGURE 1** **MRI spine - T2 sagittal image (1A) reveals a longitudinally extensive transverse myelitis. T1 contrast enhanced axial (1B) and sagittal (1C) images reveal ring enhancing lesions (arrows) in the spinal cord at L1 level.**

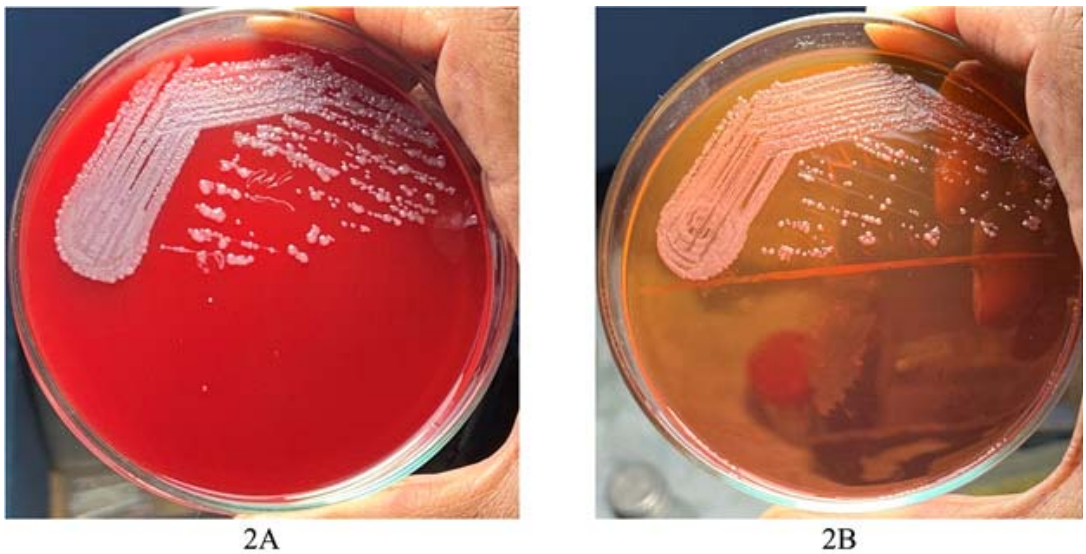


**FIGURE 2** ***Burkholderia pseudomallei*** **colonies grown on blood agar (2A) and McConkey agar (2B).**

Unfortunately, she developed acute liver injury secondary to ATT and developed early hepatic encephalopathy as well. However, following repeated blood cultures revealing evidence of infection with *Burkholderia pseudomallei*, the ATT was discontinued, and she was commenced on intravenous meropenem as for melioid-myelitis. This was continued for over 6 weeks and later combined with co-trimoxazole. With time her liver derangement settled, and she made a recovery from the encephalopathy. Her melioidosis antibodies (IgM) were highly positive (1:2560) confirming our diagnosis. Repeated brain imaging revealed resolution of the abscesses with persistence of the LETM with cord atrophy, requiring long term rehabilitation.

## DISCUSSION

Melioidosis is a bacterial infection caused by a gram-negative, aerobic rod known as *Burkholderia pseudomallei.* It is an endemic organism to the tropical regions of the world such as Sri Lanka and can cause potential life-threatening infections. The first ever reporting of melioidosis was in 1912 by Whitmore and Krishnaswami from Rangoon, where they described patients with a peculiar "pyaemic infection" caused by a gram-negative bacillus.[2] Factors which increase the risk of contracting melioidosis include DM, as in our patient, alcoholism, renal disease, immunosuppression and thalassaemia.[3] In the literature, various neurological manifestations of neuro-melioidosis have been described such as abscess formation in the brain and encephalitis.[4] There are only a few reports of myelitis secondary to melioidosis.[1,5-9] Two of these reports are from Sri Lanka.[1,9]

In 2001 Haran et al. reported a 42-year-old gentleman who developed paraplegia due to myelitis while being treated for complicated melioidosis involving the genito-urinary tract. He did not make a full recovery from his disability.[5] Saravu et al. reported a diabetic patient with disseminated bacteraemic melioidosis complicated with a subdural collection and a steroid-resistant LETM extending to the conus medullaris, as in our patient, and this patient's CSF culture had detected *B. pseudomallei*.[8] In 2021, Rudrabhatla et al. reported a patient who had LETM as a part of disseminated melioidosis infection. The patient succumbed to the illness due to severe sepsis and septic shock.[6] This highlights the importance of a high degree of suspicion to diagnose melioidosis early in order to initiate appropriate treatment and prevent disastrous consequences.

In neuro-melioidosis, the CSF usually turns out to be predominantly lymphocytic with raised protein and a normal glucose level.[4] However in our patient, as well as in the first of the two cases reported by Vithoosan et al., a significant sugar drop in the CSF was observed.[1] This led us to manage our patient as a possible case of tuberculosis initially and a similar experience was reported by Vithoosan et al. back in 2022. Unfortunately, our patient developed hepatic encephalopathy secondary to ATT prolonging and further complicating her hospital stay. The continued fever spikes led us to repeatedly send blood cultures, one of which picked up *B. pseudomallei* ultimately setting us in the right direction. Therefore, the clinical significance of a simple test such as a blood culture in the evaluation of patients with LETM goes without saying.

It is important to consider neuro-melioidosis as a differential diagnosis in patients with LETM and blood culture should play a major role in the evaluation of these patients.

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