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Case Report

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A case of central nervous system melioidosis

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Abstract

Melioidosis is a disease of humans and animals which is caused by motile Gram-negative bacilli, *Burkholderia pseudomallei.* Clinical presentation of melioidosis can vary according to the site of the infection and it can mimic various other diseases. Central nervous system (CNS) melioidosis is rare but has a high mortality rate. A high index of clinical suspicion, early diagnosis, and prompt antibiotic treatment are important for a better outcome. We present a case of central nervous system melioidosis which, presented as multiple macroscopic cerebral abscesses.

Keywords: *Melioidosis, Burkholderia pseudomallei, Central nervous system*

INTRODUCTION

Melioidosis is one of the emerging infections in Sri Lanka[1]. It is caused by *Burkholderia pseudomallei* which is an environment saprophyte mainly found in soil and surface water. *Burkholderia pseudomallei* is a Gram-negative, oxidase-positive, motile aerobic bacillus that resembles members of the genus *Pseudomonas*. Humans acquire the infection through percutaneous inoculation, inhalation, or ingestion of soil, water, or dust containing the organism [2]. Risk factors for the infections are diabetes mellitus, heavy alcohol consumption, renal disease, and liver disease. Melioidosis is known as "the great mimicker" as its clinical features can mimic many other diseases. The clinical spectrum of the disease varies from localized infection at the site of entry with no systemic manifestations to severe sepsis and death [2]. Pneumonia is the most common clinical manifestation. CNS infection is rare and accounts for 1.5%- 5% of cases. Encephalomyelitis and brain abscess are the major types of CNS disease [3,4]. Scientific data on rare presentations of melioidosis is sparse in the context of Sri Lanka. Here we report a rare case of CNS melioidosis with multiple macroscopic cerebral abscesses.

CASE REPORT

A 43-year-old manual labourer who is a heavy alcoholic was admitted to Teaching Hospital Batticaloa with a three-day history of fever, headache, double vision, vomiting, difficulty in speaking and walking, and gradually impaired consciousness. He had no history of seizures. On examination, he was confused, disoriented and his GCS was 11/15(Eye-3, Verbal-3,Motor-5). He had bilateral partial ptosis with bilateral complete ophthalmoplegia. His upper and lower limb

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examination revealed reduced muscle tone, exaggerated deep tendon reflexes with abnormal plantar reflex. Muscle power was difficult to assess on admission as the patient was confused and not obeying commands. Other systemic examination revealed no abnormalities. He was haemodynamically stable with a pulse rate of 80 bpm and blood pressure of 110/70 mm/Hg on admission. He had neutrophil leukocytosis and C-reactive protein level of 165 mg/dL. He had normal liver and renal function tests with high creatine kinase (CK) level (534 U/L). His cerebro-spinal fluid (CSF) full report revealed high CSF protein levels (130 mg/dL) with no other abnormalities. No organisms were isolated from the CSF culture. Ultra sound scan showed mild hepatosplenomegaly.

He was transferred to District General Hospital-Polonnaruwa for neuro-imaging. Non contrast CT scan, done at DGH Polonnaruwa was normal. He was started on intravenous ceftriaxone and acyclovir and transferred to National Hospital, Kandy for further imaging and management. On admission to National Hospital, Kandy his GCS was 4/15(Eye-1, Verbal-1, Motor-2) and he was electively intubated and transferred to the medical intensive care unit.

On the second day of admission to National Hospital-Kandy, his blood culture became positive for Gram-negative bacilli after 18 hours of incubation. With the direct smear findings of the blood culture, he was started on intravenous meropenem and intravenous ceftriaxone was omitted. VITEK advanced identification system identified the organism as *Burkholderia pseudomallei* and the isolate was sensitive to ceftazidime, meropenem and co-trimoxazole and was resistant to gentamicin and colistin. The isolate was sent to the Faculty of Medicine, University of Colombo for further identification studies and the isolate was confirmed as *Burkholderia pseudomallei*. A magnetic resonance scan of the brain revealed multiple ring-enhancing lesions in both cerebral hemispheres (Figure1,2). Burr hole aspiration of the cerebral lesions was done and pus was sent for culture. The same organism was isolated from the pus culture. Diagnosis of central nervous system melioidosis was made and intravenous meropenem was continued and oral co-trimoxazole was added as the intensive phase of melioidosis treatment. Throughout the antibiotic treatment his WBC/DC, renal functions and liver functions were closely monitored. The patient was transferred to Teaching Hospital Batticoloa, after three weeks of antibiotic treatment. It was planned to continue intravenous meropenem and oral co-trimoxazole for a total of eight weeks as the intensive phase and oral co-trimoxazole monotherapy for six months as the eradication phase. Unfortunately, patient passed away at Teaching Hospital-Batticoloa.

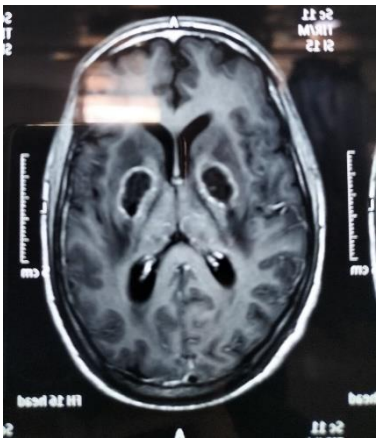

Figure 1

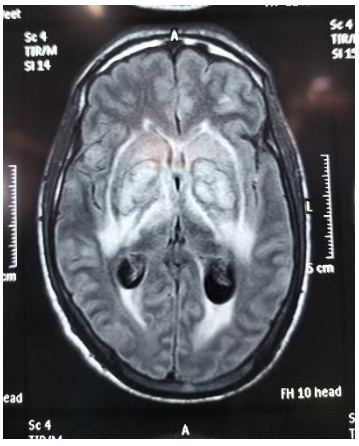

Figure 2

Figure1 and Figure 2- MRI images showing ring-enhancing lesions

DISCUSSION

Early diagnosis and prompt antibiotic treatment are important in the management of melioidosis including CNS melioidosis for better outcome. This is a case of CNS melioidosis with multiple macroscopic abscesses [2,5]. A high index of clinical suspicion is required as the presentation of melioidosis is not specific. Culture isolation of *Burkholderia pseudomallei* from clinical specimens remains the mainstay of the diagnosis. The serological diagnosis of melioidosis is difficult as high background positivity in endemic areas and false-negative results in the early disease [2,5]. As B. pseudomallei has broad intrinsic antibiotic resistance, prolong therapy is needed for the cure. An intravenous intensive phase with ceftazidime or meropenem or imipenem followed by an oral eradication phase with co-trimoxazole is recommended [6]. Duration of intensive and eradication phase depends on the site of the infection. Central nervous system melioidosis needs eight weeks of intensive phase and six months eradication phase as we have planned in our patient although we were unable to complete due to the patients demise.

In conclusions, physicians dealing with patients presenting from areas of high disease burden for melioidosis need to be aware of rare clinical presentations of melioidosis including CNS manifestations.

Author declaration

Acknowledgement:

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Author contribution:

Dr S. R Paththamperuma- Wrote the case report, drafted and edited the manuscript

Dr K. A Samarasena- Contributed to writing the case report and editing the manuscript

Dr M Kothalawla- Analysed and edited the manuscript

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Ethical Clearance:

As the patient was unconscious throughout the hospital stay, we were unable to obtain consent from the patient. Due to the COVID 19 situation in the country, none of the relatives came to visit the patient throughout the hospital stay and we were unable to contact them.

Conflict of interests:

The authors declared that there are no conflicts of interest.

REFERENCES

1. Corea EM, de Silva AD, Thevanesam V. Melioidosis in Sri Lanka. Trop Med Infect Dis. 2018;3: 22. doi:10.3390/tropicalmed3010022
2. Wiersinga WJ, Virk HS, Torres AG, Currie BJ, Peacock SJ, Dance DAB, et al. Melioidosis. Nat Rev Dis Primers. 2018;4: 17107. doi:10.1038/nrdp.2017.107
3. Owen W, Smith S, Kuruvath S, Anderson D, Hanson J. Melioidosis of the central nervous system; A potentially lethal impersonator. IDCases. 2021;23: e01015. doi:10.1016/j.idcr.2020.e01015
4. Madi D, Rai SPV, Vidyalakshmi K, Chowta KN. Neurological melioidosis presenting as intracranial abscess. Indian Journal of Pathology and Microbiology. 2016;59: 417. doi:10.4103/0377-4929.188118
5. Chakravorty A, Heath C. Melioidosis: An updated review. Aust J Gen Pract. 2019;48: 327–332. doi:10.31128/AJGP-04-18-4558
6. Sullivan RP, Marshall CS, Anstey NM, Ward L, Currie BJ. 2020 Review and revision of the 2015 Darwin melioidosis treatment guideline; paradigm drift not shift. PLoS Negl Trop Dis. 2020;14: e0008659. doi:10.1371/journal.pntd.0008659
7. Samaraweera DS. Melioidosis (Whitmore's Disease). 2017;44: 4.
8. Prasad GL. Cranial Melioidosis Presenting as Osteomyelitis and/or Extra-Axial Abscess: Literature

Review. World Neurosurgery. 2020;134: 67–75. doi:10.1016/j.wneu.2019.10.0589.

9. Wongwandee M, Linasmita P. Central nervous system melioidosis: A systematic review of individual participant data of case reports and case series. PLoS Negl Trop Dis. 2019;13: e0007320. doi:10.1371/journal.pntd.0007320

10. Melioidosis. In: Australian Journal of General Practice [Internet]. [cited 16 Aug 2021]. Available: https://www1.racgp.org.au/ajgp/2019/may/melioidosis-an-updated-review