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A rare finding of *Burkholderia pseudomallei* isolate from neck abscess, 4 years after the last report in Makassar: a case report

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ABSTRACT

Introduction: *Burkholderia pseudomallei* is the causative agent of melioidosis, known as Whitmore's disease, a tropical disease in humans. The bacteria can infect humans or animals, mostly found in contaminated water and soil. It spreads by direct contact with contaminated sources, inhalation or ingestion of contaminated dust or water droplets, or contact with soil through skin abrasions. Melioidosis is highly endemic in Southeast Asia and Northern Australia but rare in Makassar.

Case description: A 44-year-old woman shows a red mass on the right neck, reddish, palpable, $\pm 6 \times 5 \times 4$ mm in size, and is soft in consistency and has a cystic impression. Direct gram showed purulent mixed with blood. 24 hours after incubation under aerobic conditions, grew small colonies, smooth, cream in color, non-lactose fermenter on Mac Conkey agar. Indirect gram shows a small, gram-negative rod (safety pins appearance). The colonies were positive in the catalase and oxidase test, with citrate and urease test was negative. *Burkholderia pseudomallei* was identified by Vitek2 with 97% Probability. It was susceptible to doxycycline, ceftazidime, and Trimethoprim/Sulfamethoxazole.

Conclusion: This is a rare finding in a clinical isolate found in Makassar. A newly emerged case, four years after the last report in Makassar. This is still a challenge to public health problems in Makassar, must be reported consistently and never lost to follow up to get proper treatment.

Keywords: *Burkholderia pseudomallei*, melioidosis, neck abscess, whitmore's disease

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INTRODUCTION

Burkholderia pseudomallei is a small, gram-negative, oxidase-positive, motile, aerobic bacillus with occasional polar flagella. On staining, a bipolar "safety pin" pattern is seen.¹ The bacteria can infect humans or animals, mostly found in contaminated water and soil. It is spread by direct contact with the contaminated source, inhalation or ingestion of contaminated dust or water droplets, or contact contaminated soil through skin abrasions. This bacteria is a saprophyte, opportunistic, and causative agent of melioidosis disease, known as Whitmore's disease, a tropical disease in humans.²⁻⁴

Melioidosis is highly endemic in Southeast Asia and Northern Australia. In Indonesia, it was first diagnosed in Cikade,

on Java Island, 1929.⁵ From then to 1960, a few additional cases reported by Bezemer and Sudibyo reported in Jakarta, Bogor, and Surabaya.⁶⁻⁹ Recent culture-confirmed cases between 2012 and 2017 (n = 42), 15 (36%) cases were children (age <15 years) and 27 (64%) were adults (age >15 years). The overall mortality was 43% (18/42), and 57% (327/548) of healthcare workers had never heard of melioidosis. There are no additional new cases or research of melioidosis in Makassar after four years from the last report in Makassar. We recently described a confirmed case of melioidosis, a rare finding in the clinical isolate of neck abscess in Makassar.

CASE DESCRIPTION

A 44-year-old woman suddenly shows

a red mass on the right neck, reddish, palpable, $\pm 6 \times 5 \times 4$ mm in size, soft in consistency, cystic impression, and has had pain and fever chills since two months ago red mass initially found on right calf, then on cheek. Since then, she has limited her diet and lost 9,5 kilograms of her weight. She started to consume ampicillin and amoxicillin, but nothing improved, so she went to the hospital. The surgeon recommends surgery because the mass was in the lymph nodes. A 44-year-old woman shows a red mass on the right neck, reddish, palpable, $\pm 6 \times 5 \times 4$ mm in size, soft in consistency, cystic impression. Direct gram showed purulent mixed with blood. 24 hours after incubation under aerobic conditions, grew small colonies, smooth, cream in color, non-lactose fermenter on Mac Conkey agar. Indirect gram shows

small, gram-negative rods (safety pins appearance). The colonies were positive in the catalase and oxidase test, with citrate and urease test was negative. *Burkholderia pseudomallei* was identified by Vitek2 with 97% Probability. It was susceptible to doxycycline, ceftazidime, and Trimethoprim/Sulfa.

DISCUSSION

Melioidosis is a serious disease, sometimes fatal infection, caused by the bacterium *Burkholderia pseudomallei*. Bacillus is found naturally in soil and stagnant water in Southeast Asia and Northern Australia. In Indonesia, there are 99 human cases previously report. Only 11 cases were found in Makassar during 2012-2017.¹⁰

Melioidosis is acquired by inoculation of surface tissue damaged by soil and water contaminated with *Burkholderia pseudomallei*.¹¹ Inhalation and aspiration of contaminated objects are the most important routes of infection. It was explained that in Vietnam, there were French and American soldiers who contracted the disease from inhaling dust contaminated with bacteria. The disease transmission process is said to originate from aerosols made by helicopters taking off from rice fields.^{12,13} The risk of aerosol transmission outdoors is about 10 times higher than indoor transmission of workers.¹⁴

Lee et al. conducted a retrospective analysis of 158 cases of neck abscesses and found that the most common pathogen found in positive cultures was *K. pneumoniae* (13.7%). This was followed by positive findings of *Streptococcus viridans* (12.3%), methicillin-sensitive *Staphylococcus aureus* (MSSA) (11.0%), and α -hemolytic *Streptococcus* (8.2%). In addition, 9.6% of cases of infection were caused by mixed pathogens.^{15,16}

Burkholderia pseudomallei is a rare cause of neck abscess worldwide. Once infected, these pathogens may remain inactive or become active after months, years, or decades when the host's immunity declines. Another similar case was found in Malaysia in 2016. It was found that patients with very poorly controlled diabetes (HbA1c: 11.7%)

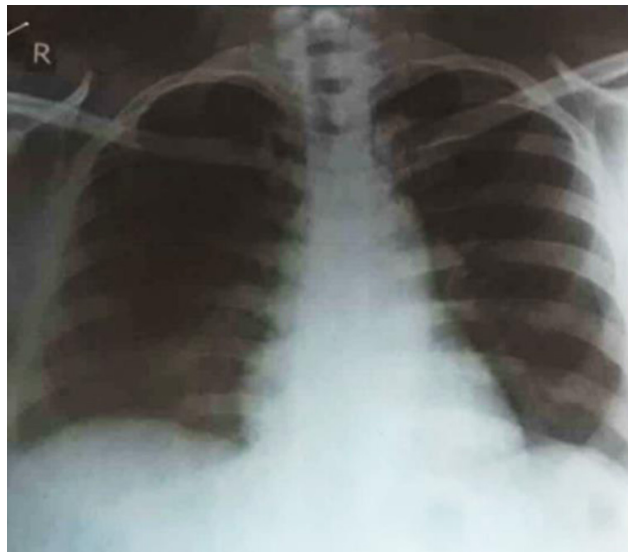


Figure 1. Chest X-ray during admission showed a bronchopneumonia on the right lobus (mild).

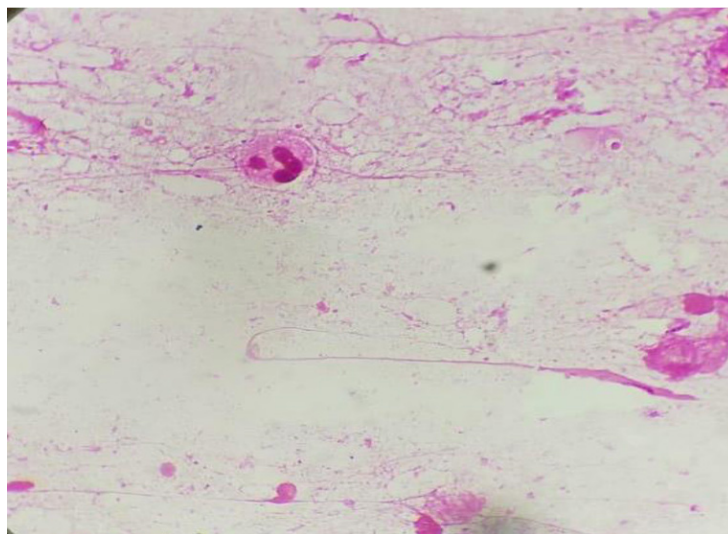


Figure 2. Leukocyte, polymorphonuclear type 2+/lpf, bipolar stained gram-negative bacilli.



Figure 3. Bipolar stained gram-negative bacilli.

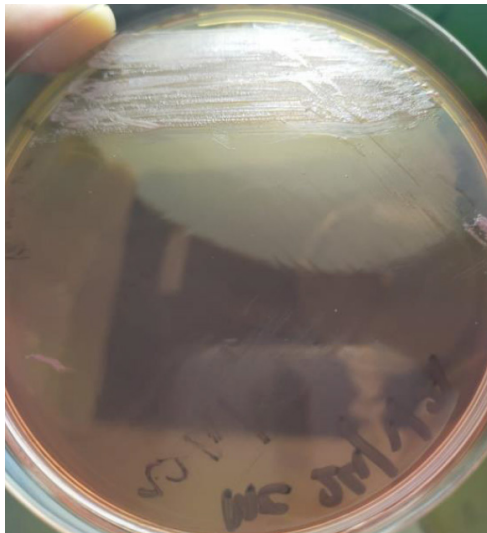


Figure 4. Small colonies, smooth, creamy in color, non fermenter, growth at 1st – 2nd quadrant on Mac Conkey agar.

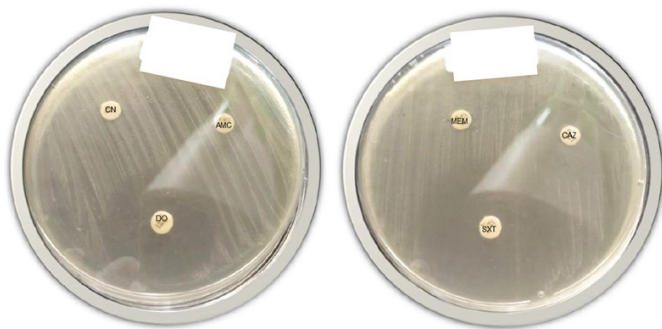


Figure 6. Antimicrobial susceptibility test of *Burkholderia pseudomallei* on MHA using Kirby-Bauer Methods. The results was susceptible to doxycycline (DO), ceftazidime (CAZ) and Trimethoprim/Sulfa (SXT), but resistant to gentamicin (CN), amoxicillin-clavulanate (AMC), and meropenem (MEM).

were predisposed to infection with the pathogen.^{15,17,18}

Isolation of *Burkholderia pseudomallei* can be easily done by taking samples from rice fields, calm or stagnant water, and moist soil that dominates in the tropics. Despite the suspected percutaneous inoculation, this patient previously had a red mass found initially on the right calf, then on the cheek two months ago, before developing a mass in his right neck. This case is similar to other studies, where they found organisms could be easily isolated

from environmental niches. It is believed that this habitat is the main reservoir from which susceptible hosts acquire infection.¹¹

Diseases closely related to melioidosis are pneumonia. After the pathogen has successfully entered from the skin that is not intact, the pathogen will inoculate and then spread hematogenous to the lungs or spread from the upper respiratory tract.¹⁴ Acute melioidotic pneumonia has a spectrum from fulminant septic shock (up to 90% mortality) to mild undifferentiated

pneumonia, which can be acute or subacute, with low mortality.¹⁹ As many as 10% of cases of secondary pneumonia occur after patients develop an untreated primary infection.

Clinical Manifestation

Several organ systems are affected by melioidosis, including the lungs and many organs. Clinical manifestations in the early phase are the finding of bilateral abscesses.²⁰ However, an infection can also be asymptomatic or only appear as local

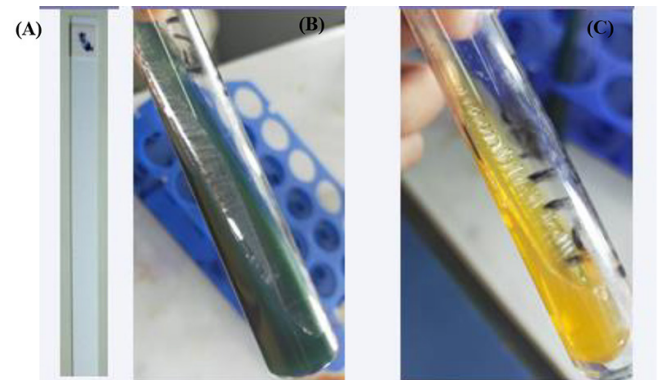


Figure 5. Biochemical reaction. (A) Positive oxidase reaction, (B) negative citrate and (C) negative urease.

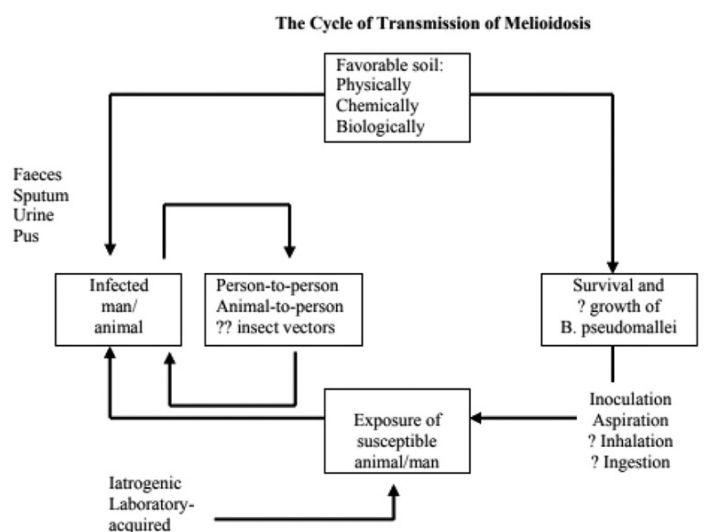


Figure 7. Animals and humans acquire infection from the environment. The organism is excreted in sputum, pus, urine and faeces. New environments may become contaminated. Rarely, human-to-human, animal-to-human and iatrogenic infections may occur. Under appropriate, and as yet poorly understood circumstances, the organism proliferates, creating a new endemic focus. Melioidosis: The cycle of transmission. The organisms proliferate, creating an endemic focus.¹¹

skin ulcers without systemic disease.²¹ Bacteremia conditions are associated with an increased mortality rate, i.e., overall mortality is 60%, 44% in Thailand, 19-46% in Australia, and 39-43% in Singapore. Melioidosis can be classified based on the duration of infection, namely acute, subacute, and chronic manifestations.²⁰⁻²⁶

DIAGNOSTIC METHOD

Burkholderia pseudomallei mimics many other conditions, especially tuberculosis, hence its other name, the “great mimicker”. Diagnosis remains a challenge, as it mocks a host of other diseases, such as tuberculosis. In Indonesia, there are 37 isolates from 42 isolates (88%) using the Vitek 2 identification system. But there are five isolates (12%) that use the Microgen method to diagnose.^{10,27} Another case report published by Azwan NAB et al. in Malaysia (2020), from June 2018 – January 2019 they found a total of four patients presented with neck abscess where the pus culture and sensitivity.²⁸

Drug of choice to treat melioidosis

Drug availability is not a challenge in Indonesia. Ceftazidime and carbapenems are widely available throughout the country and may be used and reimbursed within Indonesia’s universal health system if the diagnosis can be made. According to the World Health Organization, drug availability in Indonesia is not an issue in the country’s universal health care system.

Primary Regimens.^{29,30}

1. Initial IV therapy:

- Ceftazidime 2 gm (child 50 mg/kg) IV q6h, OR
- Meropenem 1 gm (child 25 mg/kg) (double this dose for CNS disease) IV q8h
- Duration: 14 days; ≥ 4-8 weeks in severe disease/clinical deterioration/complicated pneumonia deep-seated infection, bone/joint infection, neurological disease.

2. Eradication therapy initiated following initial therapy to prevent relapse:

- TMP-SMX 6-8mg/kg po (TMP component) bid + folic acid 5 mg once daily OR

- Pregnancy: Amoxicillin-clavulanate 875/125 mg po bid
- Duration: 3-6 months with longer duration for CNS, endovascular, or bone disease

Alternative Regimen :^{29,30}

None

Antimicrobial stewardship.³¹

Longer duration of IV therapy for certain infections: septic joint, deep tissue abscess, 4 weeks; osteomyelitis, 6 weeks, CNS infection, 8 weeks.

- Darwin Melioidosis Treatment Guideline suggestion for duration of therapy for pneumonia/bacteremia:
 - Minimum of 3 weeks of IV antibiotic if bacteremic plus single lobe infiltrate or multi-lobe pneumonia without bacteremia
 - Minimum of 4 weeks of IV antibiotic if bacteremic with multi-lobe pneumonia

Indonesia is an agricultural country in which most of the population are rice farmers, and more than 10 million people have diabetes. Preventive measures are most important for people with the following conditions: diabetes, heavy alcohol consumption, kidney disease, lung disease, cancer, receiving immunosuppressive therapy, and cuts or sores on the skin. Future challenges are to prevent melioidosis by reducing exposure, such as wearing protective gear such as rubber boots and gloves during exposure to soil.³¹⁻³³ In this patient, two risk factors may be closed related to increasing the possibility this patient get *Burkholderia pseudomallei* infection, so it can prevent melioidosis by reducing exposure to the risk factors she had. This patient must never be lost to follow up so she can get proper treatment.

CONCLUSION

This is a rare finding of *Burkholderia pseudomallei* from clinical isolate found in Makassar 2021. A newly emerged case, four years after the last report in Makassar. This is still a challenge to public health problems in Makassar, must report consistently and never lost to follow up to get proper treatment.

DISCLOSURES

The authors have nothing to disclosure.

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Conflict of Interest

The author(s) have no conflict of interest with any organizations or person that could influence the objectivity during the study, interpreting the result as well as during the writing of the manuscript.

Ethical Consent

Written informed consent was obtained from the patient for publication of this case report.

Author Contribution

Ilhamjaya A. M. contributed to conceptualization, designing and writing of the manuscript, Muhammad M., Ramadhan A. R., Sjahril R., were involved in investigation and methodology. All authors agree for this final version of manuscript to be submitted to this journal.

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