Case Report: Six New Cases of Melioidosis in the Indian Ocean Region: Comoros, Madagascar, Reunion Island

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Abstract. Melioidosis is an emerging disease caused by the environmental Gram-negative bacillus *Burkholderia pseudomallei*. Melioidosis has been reported to be endemic mainly in northern Australia and Southeast Asia. Here, we report the first two cases of melioidosis on the Comorian island of Mayotte. We also describe four cases that occurred over a short period of time in patients who had traveled between Mayotte, Madagascar, and Reunion Island.

INTRODUCTION

Melioidosis is an emerging disease caused by the environmental Gram-negative bacillus *Burkholderia pseudomallei*. This bacterium thrives in moist soils, and melioidosis has been reported to be endemic mainly in northern Australia and Southeast Asia.^{1,2} Here, we report the first two cases of melioidosis on the Comorian island of Mayotte. We also describe four cases that occurred over a short period of time in patients who had traveled between Mayotte, Madagascar, and Reunion Island. These three islands are geographically connected and share a common history linked to French colonization.

CASE SUMMARY #1

In 2016, a 75-year-old man with a history of high blood pressure and chronic kidney failure visited the Mamoudzou hospital in Mayotte with complaints of fever. The patient did not report any recent travel. Chest imaging revealed bilateral pulmonary abscesses, which were treated with amoxicillin-clavulanate. *Burkholderia pseudomallei* was identified in blood cultures using matrix-assisted laser desorption/ionization time-of-flight mass spectrometry (MALDI-TOF MS). Treatment was changed to ertapenem in combination with trimethoprim/sulfamethoxazole for 21 days, followed by trimethoprim/sulfamethoxazole for 3 months. The patient survived.

CASE SUMMARY #2

In November 2017, a 74-year-old man presenting with asthenia and febrile dyspnea visited the Mamoudzou hospital in Mayotte 1 month after returning from a trip to Madagascar, and then the Saint-Denis hospital in Reunion Island 2 weeks later. The patient had a history of chronic kidney failure and congestive heart failure. The clinical and biological assessment conducted in the Saint-Denis university hospital revealed infectious mitral endocarditis due to *Streptococcus bovis* that required surgery. One month later, the patient was admitted to the intensive care unit for septic shock associated with pulmonary abscesses. *Burkholderia thailandensis* was identified in blood cultures using MALDI-TOF MS. Four days

later, the strain was correctly identified as *B. pseudomallei* by real-time PCR using hydrolysis probes designed for three genetic markers of the type III secretion system (*orf11*, *orf13*, and *BpSCU2*). The patient was treated with ceftazidime, but the evolution was rapidly fatal.

CASE SUMMARY #3

In March 2018, a 54-year-old woman visited the Mamoudzou hospital in Mayotte with complaints of fever. A thoraco-abdominopelvic computed tomography (CT) scan revealed multiple pulmonary abscesses. The patient did not report any travel in the past year. *Burkholderia pseudomallei* was identified in blood cultures. The patient received prolonged treatment with imipenem (1 month), followed by trimethoprim/sulfamethoxazole for 3 months, which led to her recovery.

CASE SUMMARY #4

In July 2018, a 61-year-old man from Madagascar with no medical history visited the Mamoudzou hospital in Mayotte with complaints of diarrhea and fever. *Burkholderia pseudomallei* was identified in urine samples and blood cultures. The strain was susceptible in vitro to meropenem, ceftazidime, and tetracycline and was resistant to trimethoprim/sulfamethoxazole and levofloxacin. The patient was treated with ceftazidime in combination with doxycycline for 15 days, followed by doxycycline for 6 months. The evolution was favorable.

CASE SUMMARY #5

In April 2019, a 65-year-old man from Madagascar, retired from the French Navy, visited the Saint-Denis hospital in Reunion Island 1 month after returning from a trip to Madagascar. The patient, who had a history of diabetes, malignant hemopathy, and chronic obstructive bronchitis, presented with fever, cough, weight loss (8 kg in 1 month), night sweats, asthenia, and left chest pain. A chest CT scan revealed pneumonia of the upper left lobe. The patient was initially treated with amoxicillin-clavulanate. *Burkholderia pseudomallei* was identified in a respiratory specimen, and the patient was administered a third-generation cephalosporin in combination with trimethoprim/sulfamethoxazole. Given the lack of clinical improvement, a new CT scan was performed 10 days later, revealing multiple pulmonary, splenic, and hepatic abscesses. Antibiotic treatment was changed to ceftazidime in combination

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with trimethoprim/sulfamethoxazole for 4 weeks. The evolution was favorable.

CASE SUMMARY #6

The last case was a 68-year-old man living in Madagascar with a history of smoking cessation, high blood pressure, and chronic kidney disease. The patient had recently spent 10 days on the northwestern coast of Madagascar. In December 2019, he visited the hospital in Madagascar with complaints of fever, altered general condition, weight loss (20 kg in 6 months), and recent chest pain. In January 2020, he was transferred to the cardiology department of the Saint-Pierre hospital in Reunion Island, where he was diagnosed with an old myocardial infarction. He developed acute kidney failure and a biological inflammatory syndrome with evening fever, prompting the medical team to postpone coronary angiography and to transfer him to the nephrology department. A positron emission tomography revealed pneumonia of the right upper lobe. *Burkholderia pseudomallei* was identified in blood cultures taken in February. Treatment with ceftazidime in combination with trimethoprim/sulfamethoxazole was initiated. The patient's clinical condition improved, and he was discharged home.

DISCUSSION

In this article, we report six new cases of melioidosis from the Indian Ocean region that occurred over a relatively short period of time (3 years). We also report the first two cases of melioidosis on the Comorian island of Mayotte (Cases #1 and #3).

It is difficult to study the occurrence of melioidosis in the islands of Mayotte, Madagascar, and Reunion without grouping them together because most patients traveled to two or three of these islands before being diagnosed with melioidosis. This was true for three of the seven patients in the case series that we reported earlier³ and for four of the six patients in the present series.

The first seven cases of community-acquired melioidosis that we reported for the Indian Ocean region had occurred over a period of 14 years.^{3,4} Six of these cases had come from the Mahajanga region of Madagascar,⁴ where *B. pseudomallei* was recently identified in soil samples.⁴ The last case of melioidosis had occurred in Reunion Island and was indigenous.⁵

In the present series, two cases of melioidosis in Mayotte were likely indigenous because the patients did not report any travel. It may be that Reunion Island and Mayotte are at risk of melioidosis due to their tropical climate.

It is important that physicians and biologists be informed and trained about melioidosis because MALDI-TOF MS has been shown to misidentify pathogens. This could have consequences on the evolution of the disease, as a wrong antibiotic regimen could be selected by a clinician who fails to consider the possibility that the patient has in fact contracted melioidosis.⁶ When *B. thailandensis* is identified in a patient from an endemic area with symptoms compatible with melioidosis, further testing of the isolated microorganism should be performed with conventional biochemical assays. Ideally, molecular identification procedures should be conducted, such as 16S rRNA gene sequence analysis or real-time PCR assays targeting specific genetic markers.

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