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Case Report

# Cardiac and mediastinum involvement in Burkholderia thailandensis infection: A case report and literature review ☆,☆☆

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ABSTRACT

Melioidosis, an infectious disease caused by Burkholderia pseudomallei, is prevalent in Southeast Asia and Northern Australia, presenting various clinical manifestations from asymptomatic to life-threatening infections. Although primarily affecting the lungs, intra-abdominal viscera, and musculoskeletal system, melioidosis can rarely involve the heart and mediastinum, which pose significant diagnostic and therapeutic challenges. Herein, we present the case of a 53-year-old male farmer who presented with persistent fever and chest pain, progressing to pericarditis and cardiac tamponade. Imaging revealed necrotic mediastinal lymphadenopathy and an enhancing pericardium with pericardial effusion. The patient underwent emergency surgical drainage and was treated with intravenous followed by oral antibiotics. Culture confirmed Burkholderia thailandensis, a closely related but less commonly reported species. This report highlights the complexities of diagnosing and managing B. thailandensis, which can mimic aortic disease, tuberculosis, malignancies, and other inflammatory conditions, especially in endemic areas, emphasizing the need for prompt medical and surgical treatment to improve patient outcomes.



## Introduction

Melioidosis is an infectious disease caused by the gram-negative bacterium Burkholderia pseudomallei, predominantly affecting populations in Southeast Asia and Northern Australia [1]. The disease manifests in a wide range of clinical presentations, from asymptomatic infections to severe septicemia, depending on the organs involved, and can be potentially life-threatening [1,2]. The lungs are the most frequently

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affected organs, followed by the intra-abdominal viscera and the musculoskeletal system [2,3]. Although rare, melioidosis can also affect the heart and mediastinum, leading to conditions such as pericarditis and cardiac tamponade, which pose significant diagnostic and therapeutic challenges [3].

A closely related species, Burkholderia thailandensis, is less commonly associated with human infections but shares many of the virulence factors of B. pseudomallei. Infections with B. thailandensis are rare and often underreported, limiting the understanding of its clinical presentation and management [4,5].

In this case report, we describe a rare instance of melioidosis caused by B. thailandensis presenting with fever and chest pain, which progressed to pericarditis and cardiac tamponade. These symptoms closely mimicked aortic disease and infective pericarditis, complicating the diagnostic process. Our aim is to highlight the clinical features, imaging findings, diagnostic difficulties, and management strategies of this unusual presentation. Through this report, we aim to expand the current understanding of melioidosis and assist clinicians in diagnosing and managing these rare and challenging cases.

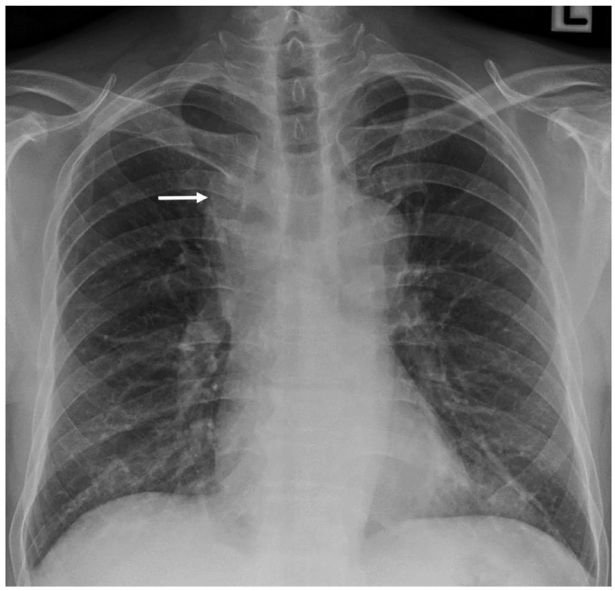

**Fig. 1 – Frontal chest radiographs demonstrated superior mediastinal widening and soft tissue opacity along the right paratracheal region (arrow).**

## Case report

A 53-year-old male farmer with no underlying diseases presented with a 1-month history of prolonged fever and pleuritic chest pain. He denied any history of tuberculous exposure. On physical examination, a high-grade fever was noted. His vital signs were as follows: blood pressure 101/47 mmHg, pulse 100/min, and temperature, 38.5°C. The physical examination showed JVP engorgement with distant heart sounds. Breath sounds were equal and clear. The ECG findings demonstrated sinus rhythm with low voltage. Laboratory investigations revealed neutrophilic leukocytosis with a total white blood cell count of $1.4551 \times 10^9$/L and neutrophils at 75.85%. Renal and liver function tests were within normal limits. His chest radiograph showed superior mediastinal widening and soft tissue opacity along the right paratracheal region, suggesting infected aortitis, mediastinal mass or another aortic disease (Fig. 1). A contrast-enhanced computed tomography angiography (CTA) of the aorta revealed a multiloculated rim-enhancing lesion with internal hypoattenuation in the right paratracheal region and an enhancing pericardium with a moderate amount of pericardial effusion (Figs. 2A and B). The thoracic and abdominal aorta appeared normal in size and contour. The lung parenchyma showed no evidence of pulmonary nodules, cavitary lesions, or consolidation. The bilateral pleural cavities were clear, and the rest of the abdominal organs were unremarkable. These findings suggest necrotic mediastinal lymph nodes with pericarditis.

The echocardiography findings revealed a thickened fibrous-like pericardium with a large circumferential hyperechoic pericardial effusion, measuring approximately 2.5 cm in maximal thickness. Ejection fraction was 62.8%. These features were accompanied by a swinging heart motion, visible right atrium (RA) and right ventricle (RV) diastolic collapse, inferior vena cava (IVC) plethora, and the presence of pulsus paradoxus—highlighted by an exaggerated mitral valve (MV) inflow velocity exceeding 25%, which are compatible with cardiac tamponade (Fig. 3).

Following the echocardiography findings indicative of cardiac tamponade, emergency surgical intervention was carried out on the patient. The operation included medial sternotomy with pericardiectomy, pericardial window creation, drainage of pericardial effusion, cardiac decortication, and debulking of the enlarged and friable right paratracheal lymph nodes. Intra-operatively, thickened pericardium was observed with approximately 200 mL of clear yellow pericardial effusion. The heart and intrapericardial vessels were covered with fibrin, alongside enlarged lymph nodes with pockets of pus in the right paratracheal region. Tissue samples and pericardial fluid were collected for microbiological and pathological examination.

Subsequent culture results from the right paratracheal lymph nodes yielded growth of Burkholderia pseudomallei/thailandensis, leading to a diagnosis of melioidosis. Intravenous ceftazidime was administered for the intensive phase of treatment. The patient responded well to the treatment, showing clinical improvement, and was discharged on oral trimethoprim/sulfamethoxazole (400/80) at a dosage of 3 tablets every 12 hours.

During a follow-up visit, the patient exhibited significant symptom improvement. A contrast-enhanced computed tomography scan of the chest revealed residual small lymph nodes with central low attenuation and resolution of pericardial effusion (Figs. 2C and D). To continue the progress, the physician decided to maintain the patient on trimethoprim/sulfamethoxazole for a total of 20 weeks as a maintenance therapy.

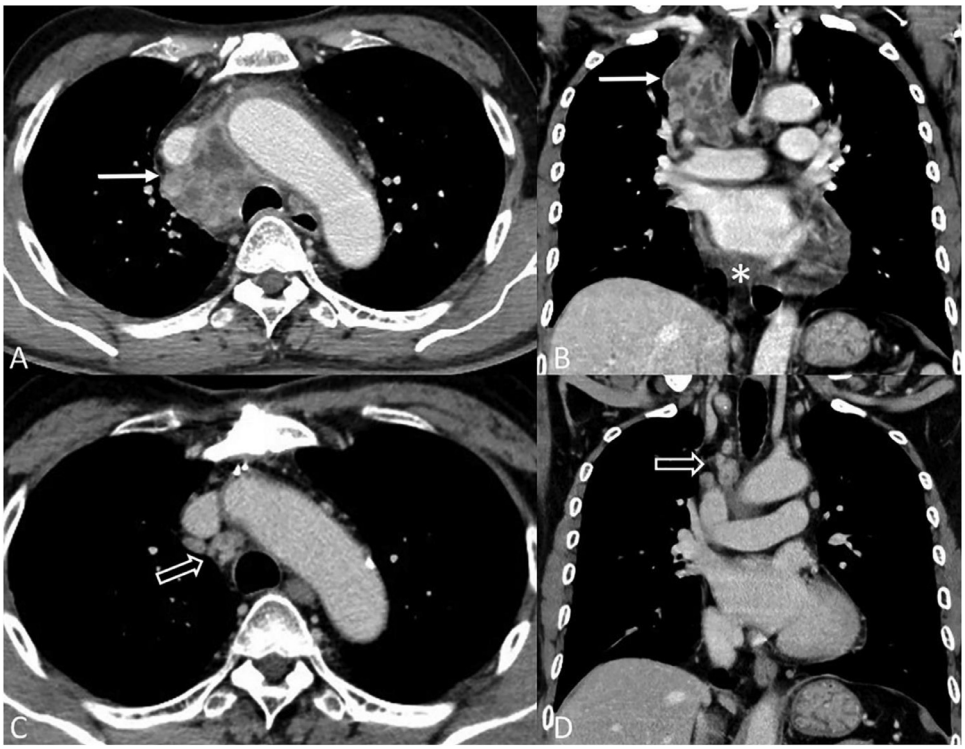


**Fig. 2 – (A and B) Axial and coronal views contrast-enhanced computed tomography angiography (CTA) of the chest show multiloculated rim-enhancing lesion with internal hypoattenuation in the right paratracheal region (arrows) and a moderate amount of pericardial effusion (asterisk). After surgical drainage and intravenous antibiotics (C and D) demonstrated few small residual lymph nodes (open arrows) and resolution of pericardial effusion.**

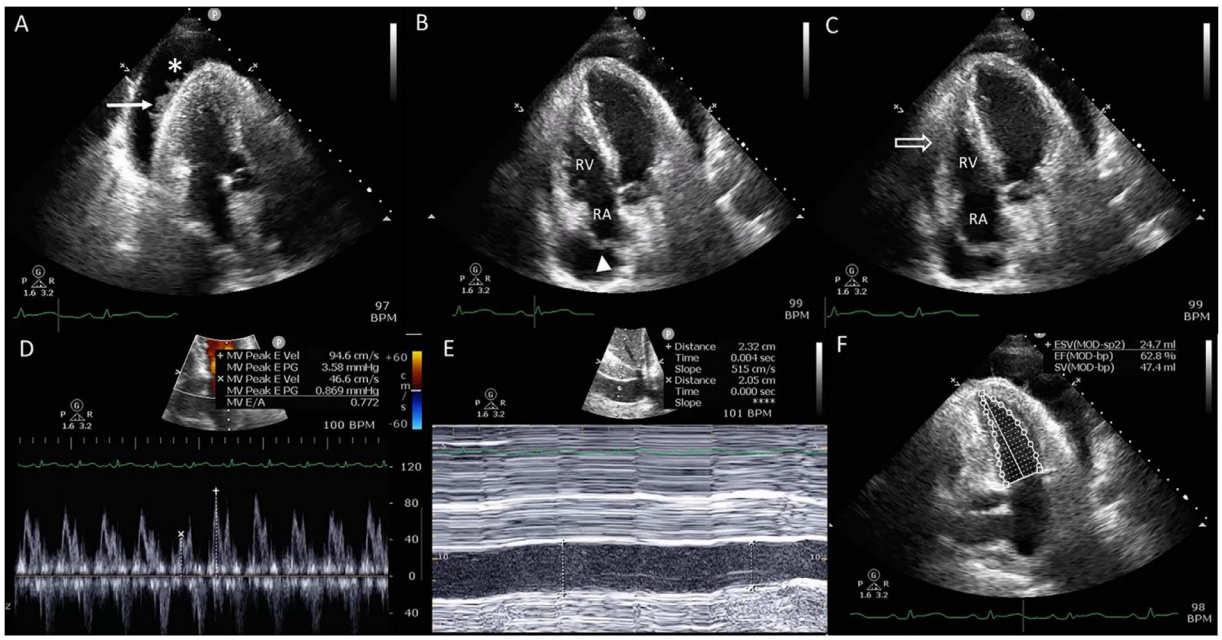


**Fig. 3 – (A) Transthoracic echocardiography demonstrated a large amount of pericardial effusion (asterisk) with frond-like hyperechoic projections from the surface of pericardium (arrow). (B and C) The collapsed RA in diastole (arrowhead) and the collapsed RV in the early diastolic phase (open arrow). (D) The mitral valve (MV) inflow velocity measurement depicted more than 25% respiratory variation in mitral inflow. (E) IVC imaged in M-mode demonstrated a lack of the normal inspiratory collapse of a dilated IVC, indicating IVC plethora. (F) Left ventricle ejection fraction assessment using Simpson's biplane method. The LVEF is about 62.8%.**

## Discussion

Burkholderia pseudomallei, the cause of melioidosis, is transmitted through inhalation, ingestion, or inoculation, commonly found in soil and surface water, especially in rice paddies [2,3]. Predisposing factors include diabetes, male gender, age over 45, alcoholism, chronic liver, lung, and kidney diseases, malignancies, hematological disorders like thalassemia, and prolonged steroid use [2,3].

Burkholderia pseudomallei includes various species. Burkholderia thailandensis, a member of the B. pseudomallei complex, is closely related to B. pseudomallei but distinguishable by its L-arabinose assimilation and genotypic analysis. Molecular and genetic analyses have further differentiated B. thailandensis from B. pseudomallei based on DNA sequences [4,5]. In 1999, Lertpatanasuwan et al. [6] reported the first case of B. thailandensis infection in a 16-year-old male in Thailand, presenting with a soft tissue abscess and septicemia following multiple organ injury and wound infection from a motorcycle accident. Since then, 6 additional cases have been reported: 3 cases in the US involved pneumonia and wound infections [5,7]; 1 case in Malaysia was diagnosed with osteoarthritis and a soft tissue abscess [8]; 1 case in China involved pneumonia [9]; and the most recent case in Thailand in 2022 presented with pneumonia and mediastinal lymphadenopathy [10]. Our case is the first report of B. thailandensis infection presenting with mediastinal and cardiac involvement.

The clinical presentation of melioidosis varies widely, ranging from asymptomatic cases to severe septicemia, depending on the organs involved. Common manifestations include pulmonary, intra-abdominal, and musculoskeletal infections [11]. Pulmonary involvement often presents as nodules or patchy opacities that can progress to cavitary lesions. Abdominal manifestations typically feature hepatic and splenic abscesses, while musculoskeletal infections result in septic arthritis, synovitis, osteomyelitis, and soft tissue infections such as cellulitis, necrotizing fasciitis, or abscess formations [11]. Isolated heart and mediastinal involvement, including mediastinal lymphadenopathy, abscesses, pericarditis, myocarditis, or endocarditis, are rare, occurring in less than 1% of melioidosis cases [12]. In 2010, the Darwin prospective melioidosis case series [3] revealed that only 1% (4 out of 540 cases) involved mediastinal melioidosis with pericarditis, and one of those cases developed acute pericardial tamponade requiring emergency thoracotomy and a pericardial window [3]. Similarly, Chung et al. [13] reported a case of melioidosis with nonsuppurative cardiac tamponade and mediastinal lymphadenopathy. Our case presented a rare instance of necrotic mediastinal lymphadenopathy, mediastinal collection, and pericarditis with cardiac tamponade, without involvement of other organs.

The differential diagnosis of melioidosis depends on the clinical scenario and radiologic manifestations. Nyanti et al. [14] conducted a systematic review on mediastinal melioidosis, finding that most cases reported symptoms of cough, fever, and malaise. In our case, the patient presented with fever and chest pain, which is unusual compared to previous literature. A chest radiograph showed mediastinal widening, initially suggesting aortic disease. However, a retrospective review revealed the widening was due to increased soft tissue opacity in the right paratracheal region, which could indicate conditions like right paratracheal lymphadenopathy, a mediastinal mass, or SVC/ascending aorta pathology. Diseases of the aortic arch and descending aorta typically involve the left side of the mediastinum, appearing as increased left mediastinal width, indistinct aortic contour, and loss of the aortopulmonary window, which was not seen in our case [15,16]. Eventually, contrast-enhanced computed tomography angiography (CTA) of the aorta identified multiloculated rim-enhancing lesions with internal low attenuation areas in the right paratracheal region, consistent with an enlarged node with internal necrosis. Additionally, an enhancing pericardium with moderate pericardial effusion, suggestive of pericarditis and cardiac tamponade, seen in the CTA and echocardiogram explained the patient's chest pain.

Necrotic mediastinal lymphadenopathy with pericarditis can be caused by various infectious, malignant, and inflammatory conditions. Tuberculous lymphadenitis and pericarditis are differential diagnoses in our case, given their prevalence in endemic areas such as Thailand, with radiographic features including multiple enlarged conglomerated or discrete nodes with low internal attenuation and peripheral enhancement [17]. Tuberculous pericarditis can manifest as simple pericardial effusion, irregular pericardial thickening with effusion, or constrictive pericarditis [18,19]. However, our patient had no history of tuberculosis exposure or constitutional symptoms typical of tuberculosis infection, reducing the likelihood of this diagnosis. Other infections such as melioidosis or fungal infections, which are endemic in Thailand, can also present with similar clinical and imaging findings. Due to the gold standard for diagnosing melioidosis is the isolation of the bacteria through culture, further investigation with gram stain, culture, or tissue pathology is essential for definitive diagnosis. In cases of nodal metastasis with pericardial involvement from malignancy, imaging typically shows irregular enhancing tissue or large conglomerated lymph nodal masses with central necrosis [20]. Irregular pericardial thickening can suggest malignant pericardial effusion [21]. However, patients with nodal metastasis usually have an underlying cancer and radiographic features of primary cancer, which were not seen in our case. Moreover, mediastinal lymphadenopathy with simple pericardial effusion without thickening, as in our case, is nonspecific for distinguishing between benign and malignant pericardial effusion. Therefore, histopathological examination of nodal tissue or pericardium remains essential for accurate diagnosis. Other inflammatory conditions such as sarcoidosis and autoimmune diseases like rheumatoid arthritis and systemic lupus erythematosus usually present with symmetrical mediastinal and hilar lymphadenopathy, less commonly causing necrosis, and pericardial effusion is rare [20]. Additionally, these conditions are often associated with systemic symptoms and signs specific to the autoimmune disease, none of which were observed in this patient.

For the treatment of mediastinal melioidosis with pericarditis and cardiac tamponade, prompt and aggressive intervention is essential for a favorable outcome. The standard approach includes an intensive phase of intravenous antibiotics like ceftazidime or meropenem, followed by a prolonged eradication phase with oral antibiotics such as trimethoprim-

sulfamethoxazole to prevent relapse [1,11]. Emergency pericardiocentesis or surgical interventions like a pericardial window and cardiac decortication may be necessary to manage cardiac tamponade [12]. Supportive care in an intensive care setting is often required. Advances in rapid diagnostic techniques and novel antibiotic regimens are emerging, aiming to enhance treatment efficacy and improve patient outcomes.

## Conclusion

This case report highlights the diagnostic and therapeutic challenges of Burkholderia thailandensis, a rare infection related to Burkholderia pseudomallei. The atypical presentation and uncommon radiological manifestations, such as isolated cardiac and mediastinal involvement, underscore the diagnostic difficulties. This report aims to raise awareness among clinicians and radiologists about the complexities of melioidosis to improve patient care and outcomes.

## Patient consent

The authors certify that they have obtained all appropriate patient consent.

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