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A Case of Constrictive Pericarditis Associated with Melioidosis in an Immunocompetent Patient Treated by Pericardiectomy

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
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Patient: Male, 38
Final Diagnosis: Constrictive pericarditis
Symptoms: Shortness of breath
Medication: —
Clinical Procedure: Pericardiocentesis • pericardiectomy
Specialty: Cardiology

Objective: Unusual clinical course

Background: Melioidosis is a rare tropical bacterial infection caused by the Gram-negative soil saprophyte, *Burkholderia pseudomallei.* Melioidosis can mimic a variety of diseases due to its varied presentation, and unless it is treated rapidly, it can be fatal. A rare case of melioidosis, with pericarditis and pericardial effusion, is described, which demonstrates the value of early diagnosis with echocardiography and pericardiocentesis.

Case Report: A 38-year-old native (Iban) East Malaysian man presented with shortness of breath and tachycardia. Transthoracic echocardiography (TTE) showed cardiac tamponade. Urgent pericardiocentesis drained a large amount of purulent pericardial fluid that grew *Burkholderia pseudomallei*. Despite appropriate dose and duration of intravenous treatment with ceftazidime followed by meropenem, the patient developed recurrent pericardial effusion and right heart failure due to constrictive pericarditis. The diagnosis of constrictive pericarditis was confirmed by computed tomography (CT) and surgical exploration. Following pericardiectomy, his symptoms resolved, but patient follow-up was recommended for possible sequelae of constrictive pericarditis.

Conclusions: After the onset of melioidosis pericarditis, the authors recommend follow-up and surveillance for possible complication of constrictive pericarditis.

MeSH Keywords: Echocardiography • Melioidosis • Pericardiectomy • Pericardiocentesis • Pericarditis, Constrictive

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Background

Melioidosis is a rare tropical bacterial infection caused by the Gram-negative soil saprophyte, *Burkholderia pseudomallei* and can mimic a variety of diseases due to its varied presentation, and unless it is treated rapidly, it can be fatal [1]. Melioidosis is a tropical infection that most commonly occurs in South East Asia, India and northern Australia [2]. Because of the availability of long-distance travel, sporadic cases of melioidosis have been reported, including a case in a Swiss traveler returning home after visiting Thailand, which is an endemic area for *Burkholderia pseudomallei* [3].

Melioidosis is an important opportunistic infection that can cause sepsis in adults with underlying conditions that impair immune function, such as diabetes, chronic renal failure, prolonged steroid use, and alcoholism [2]. Because *Burkholderia pseudomallei* is as soil saprophyte, agricultural workers may also be at risk from melioidosis [2]. The published literature has reported a significant number of melioidosis cases in Malaysia [1].

We report a rare case of melioidosis that presented with cardiac tamponade and recurrent pericardial effusion complicated by constrictive pericarditis, in an immunocompetent individual.

Case Report

A 38-year-old native (Iban) East Malaysian man, who worked in a shipyard, complained of progressive shortness of breath, loss of weight, and loss of appetite for one month. He had no predisposing comorbidities, and there was no history of diabetes mellitus, chronic renal failure, alcoholism, or malignancy.

On admission to the emergency department, he was tachypneic and tachycardic. His heart rate was 139 beats per minute, and his blood pressure was 100/70 mmHg. The jugular venous pressure was elevated, and the cardiac apex beat was diminished on palpation. The lower lung fields were dull on percussion, with bilateral reduced vocal fremitus and vocal resonance.

Laboratory investigations showed a neutrophilic leukocytosis. Renal function tests, liver function tests, and fasting blood glucose levels were normal. An electrocardiogram (ECG) showed sinus tachycardia, and low QRS voltage in the limb and chest leads. A chest radiograph showed cardiomegaly and bilateral mild pleural effusions. The transthoracic echocardiogram (TTE) showed a large pericardial effusion resulting in cardiac tamponade (Figure 1A). Echo-guided pericardiocentesis was performed urgently via an indwelling pigtail catheter, which drained approximately 1,000 ml of purulent pericardial fluid. A repeat TTE showed minimal residual pericardial effusion (Figure 1B) and a 55% left ventricular ejection fraction (LVEF).

Laboratory investigations of the pericardial fluid showed an elevated protein level, a white cell count of 80 per mm^3 with 20% lymphocytes and 80% neutrophils. Pericardial fluid microscopy showed numerous pus cells and mononuclear cells, and culture grew the Gram-negative aerobic organism, *Burkholderia pseudomallei*. The isolate was sensitive to imipenem, meropenem, ceftazidime, and co-trimoxazole. The pericardial fluid was negative for Ziehl-Neelsen stain and Mycobacterial culture. Ultrasound of the abdomen showed a normal liver, spleen, and kidneys. Blood test for human immunodeficiency virus (HIV) antibody was negative.

The patient was treated with intravenous (IV) ceftazidime 2 gm, six-hourly. However, after 17 days of IV ceftazidime, echocardiography showed re-accumulation of the pericardial effusion. A second pericardiocentesis drained approximately 900 ml of purulent pericardial fluid. Following the detection of recurrent pericardial effusion, the infectious disease physician changed the antibiotics to IV meropenem 1gm, eight-hourly, in combination with oral Bactrim (trimethoprim/sulphamethoxazole 80/400 mg) twice daily. The patient's general condition improved gradually after the second pericardiocentesis and following the change of antibiotic regime. However, the chest radiograph after two weeks of antibiotics still showed bilateral moderate pleural effusions. Therefore, a third antibiotic was added to the treatment regime, ciprofloxacin 500 mg twice daily, and the dose of IV meropenem was increased to 2 gm three times daily. Following two weeks of triple antibiotic therapy, the repeat chest radiograph showed minimal pleural effusions.

The patients was discharged from hospital after six weeks of in-hospital IV antibiotics treatment followed by one month of oral Bactrim (trimethoprim/sulphamethoxazole 80/400 mg) twice daily, doxycycline 100 mg twice daily, and ciprofloxacin 500 mg twice daily. Unfortunately, nine months later, the patient was re-admitted to hospital with right heart failure. On this admission, TTE showed moderate, loculated pericardial effusion surrounded by pleural effusions. Paradoxical cardiac septal motion ('septal bounce') that was associated with respiration was seen in the apical four-chamber view of the TTE.

A Doppler mitral inflow study showed a high trans-mitral inflow peak early filling (E-wave) and late diastolic filling (A wave) velocities, and an increased E to A ratio of 2 (Figure 1C). Doppler tissue imaging showed preserved medial mitral annular E-wave velocity (peak E-wave: 16.4 cm/sec) and loss of A-wave velocity (Figure 1D). Respiratory variations of E-wave velocities were found with each cardiac cycle. In mitral inflow, E-wave velocity was decreased by >25% during inspiration (Figure 1E), whereas, in the tricuspid inflow view, the E-wave velocity was increased with inspiration (Figure 1F). The inferior vena cava (IVC) was dilated with minimal respiratory variation. The estimated

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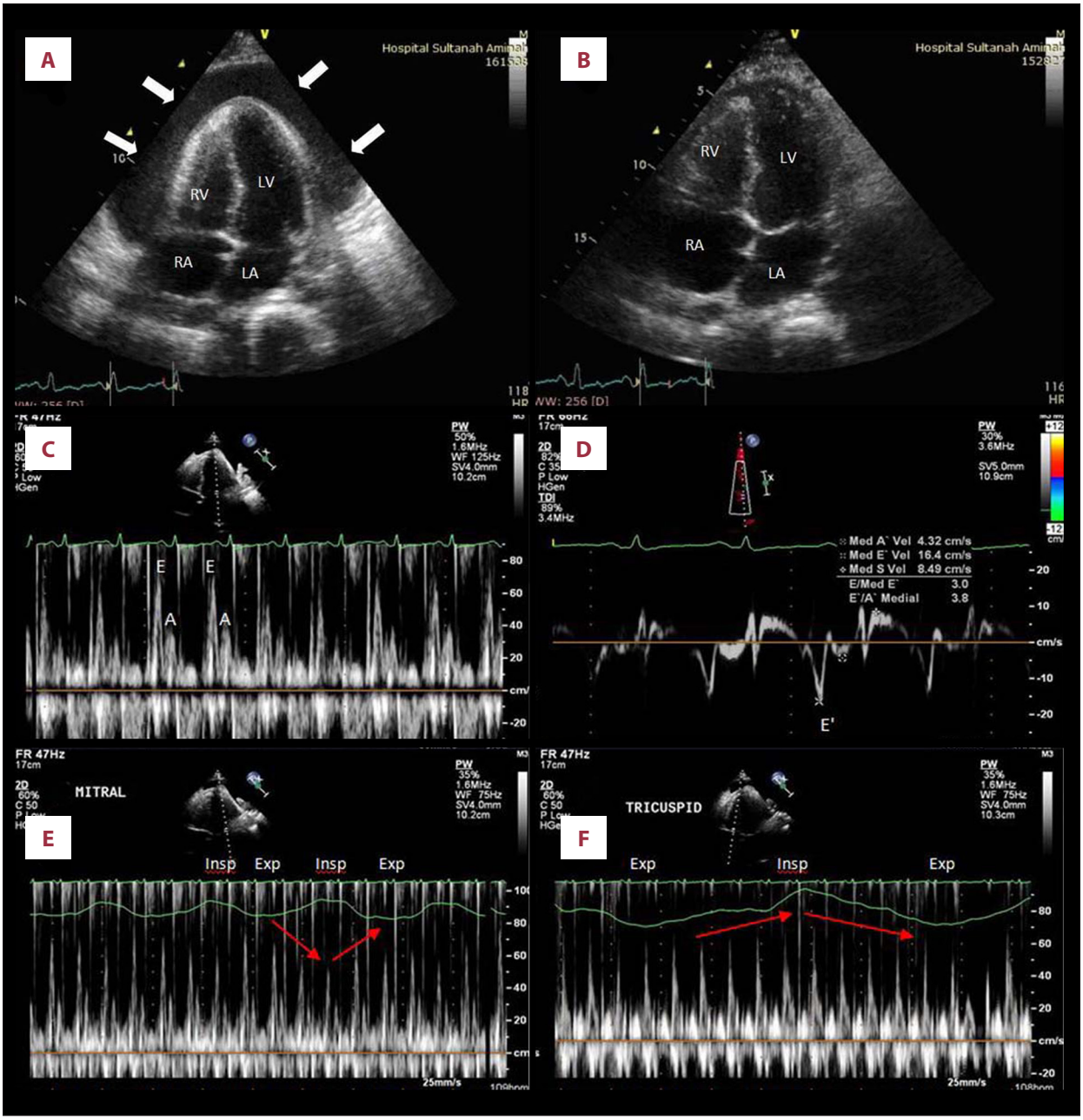

Figure 1. Transthoracic echocardiogram (TTE), apical four-chamber view. (**A**) The large pericardial effusion (white arrow) is shown. (**B**) Resolution of the pericardial effusion after pericardiocentesis. (**C**) Doppler mitral inflow study shows a high E velocity wave, shortened deceleration time, and an increased E to A ratio (>2). (**D**) Doppler tissue imaging shows preserved mitral annular E' velocity (peak E': 16.4 cm/sec and loss of A' velocity). (**E**) Increase in respiratory variation in pulse wave Doppler E velocity (red arrow). Note the inspiratory decrease and expiratory increase in E velocity (>25%) in trans-mitral flow. (**F**) Note the inspiratory increase and expiratory decrease in E velocity in trans-tricuspid flow. LV – left ventricle; LA – left atrium; RV – right ventricle; RA – right ventricle; Insp – inspiration; Exp – expiration.

pulmonary artery systolic pressure was 41 mmHg. Even though the pericardium, as viewed by TTE, was not calcified or thickened, the Doppler and tissue imaging hemodynamic findings were consistent with constrictive pericarditis.

Computed tomography (CT) of the thorax showed a thickened pericardium (8-10mm), a loculated pericardial effusion, and a predominantly left pleural effusion (Figure 2A). The clinical presentation, echo-Doppler findings, and CT evidence of a thickened pericardium were consistent with constrictive pericarditis.

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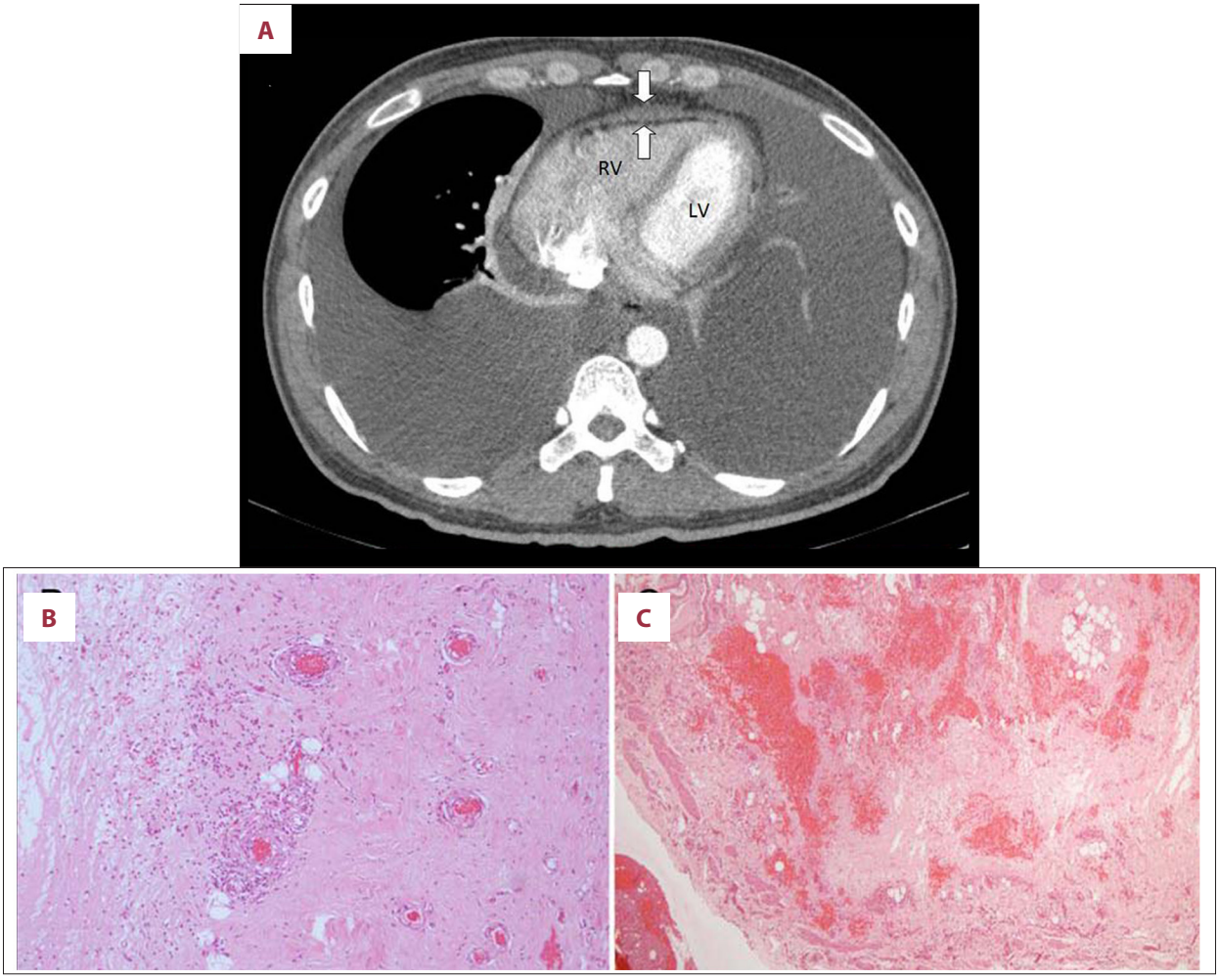

Figure 2. Thoracic computed tomography (CT) imaging. (**A**) Thoracic computed tomography (CT) image shows a thickened pericardium (arrow), loculated pericardial effusion, and a large left pleural effusion. (**B**) Photomicrograph of the histology of the inflamed pericardial fibromuscular tissue shows congested vessels. Hematoxylin and eosin (H&E) stain. Objective magnification ×10. (**C**) Pleural tissues with areas of hemorrhage, increased fibrosis, and inflamed granulations tissue formation. No granulomas are seen. Hematoxylin and eosin (H&E) stain. Objective magnification ×4. RV – right ventricle; LV – left ventricle.

The patient was referred to a cardiothoracic surgeon and underwent a successful pericardiectomy. Histologic examination and culture of pericardial biopsy (Figure 2B) showed multiple fragments of fibrous and fibrofatty tissue containing congested small-sized to medium-sized blood vessels, mild-to-moderate neutrophil infiltrates, and some lymphocytes and plasma cells. There were no vasculitic changes, no micro-organisms, and no malignancy. The pleural biopsy (Figure 2C) also showed areas of hemorrhage, increased fibrosis, and granulation tissue formation. Periodic acid–Schiff (PAS), Gomori's methenamine silver stain (GMS) and Ziehl-Neelson stains were negative. These special investigations of the pericardial and pleural biopsies ruled out tuberculosis and fungal infection as the differential diagnosis of constrictive pericarditis. Following pericardiectomy, there was no evidence of relapse of melioidosis during the follow-up period of more than two years.

Discussion

The clinical manifestations of melioidosis vary from benign localized abscesses to severe pneumonia, to acute fulminating septicemia with multiple abscesses, often leading to death [1,2]. Pericardial effusion is a rare presentation of melioidosis and has previously been reported in a series of case reports [3,4]. Constrictive pericarditis is an uncommon post-inflammatory disorder characterized by a variably thickened, fibrotic, and frequently calcified, pericardium. Common causes of constrictive pericarditis include repeated episodes of acute pericarditis, previous cardiac surgery, and radiation therapy [5].

In this case report, recurrent pericardial infection was the most plausible explanation for the development of constrictive pericarditis. However, there is limited data regarding

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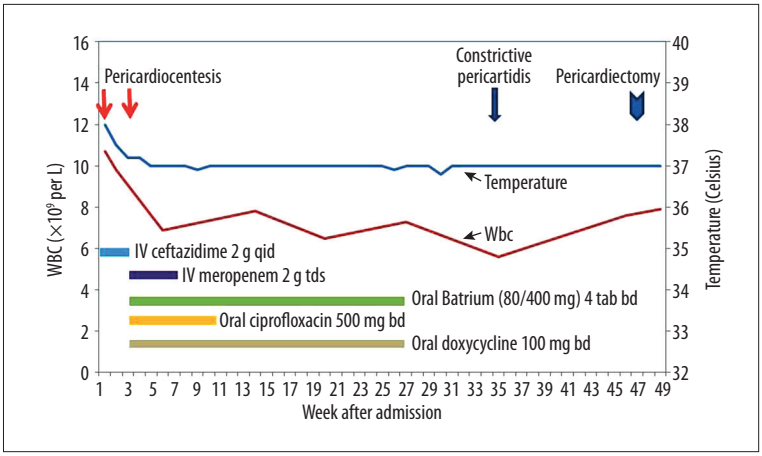

Figure 3. The clinical course of the patient. The patient developed constrictive pericarditis nine months after the first hospital admission. IV – intravenous; WBC – white blood cell.

constrictive pericarditis secondary to melioidosis. A previously published case of melioidosis pericarditis reported imaging findings and hemodynamic confirmation of constrictive pericarditis [6]. However, the clinical outcome of the patient in this previous case report was unknown, and there was no information on the pericardial histology, which is an important diagnostic test, particularly in differentiating between tuberculosis and melioidosis [6].

Because melioidosis is a great mimic of other disease processes, it may pose a diagnostic challenge if clinicians lack awareness of this infection, or have a low clinical index of suspicion. A sensitive and specific serological test can support a diagnosis of melioidosis. However, isolation of *Burkholderia pseudomallei* from the patient tissue or fluids remains the 'gold standard' for the diagnosis of melioidosis [1]. In areas where both tuberculosis and melioidosis are prevalent, clinicians will consider tuberculous pericarditis in the differential diagnosis.

Chetchotisakd et al. found that the major diagnostic method to distinguish between melioidosis pericarditis and tuberculous pericarditis was pericardial fluid culture, but pericardial histology was found to be most useful when diagnosing tuberculosis [4]. The patient lived in an area where tuberculosis and melioidosis were endemic. Therefore, pericardiocentesis for culture and pericardial biopsy are important to differentiate between melioidosis and tuberculous pericarditis because the treatments of two diseases are different. Also, melioidosis should be ruled out in patients with suspected tuberculosis while instituting presumptive anti-tuberculosis treatment in areas where both diseases are prevalent [7]. Non-septicemic melioidosis presenting as cardiac tamponade carries a mortality of between 20–60% [8]. Early detection, pericardiocentesis, and appropriate antibiotics significantly reduce the morbidity and mortality of pericardial melioidosis. However, recurrent melioidosis can occur despite the primary episode being non-bacteremic and despite adherence to appropriate antibiotic regimes [9].

In this case report, the patient survived because of early intervention and intensive treatment. In the era of modern imaging, the diagnosis of constrictive pericarditis continues to be a clinical challenge. Other pericardial diseases, such as hemopericardium or pericardial cyst [10], right heart failure due to severe tricuspid regurgitation [11], or any pathological condition that can cause a restrictive effect on the heart, may mimic constrictive pericarditis. The distinction between constrictive pericarditis and other causes of heart failure, such as restrictive cardiomyopathy, is important because pericardiectomy can be an effective treatment for constrictive pericarditis. Preserved medial mitral annular early filling, or E-wave velocity, by Doppler imaging is observed in constrictive pericarditis, which is an important point of distinction from restrictive physiology [12]. Notably, the imaging findings of constrictive pericarditis were confirmed by the surgical findings in this case. The patient's symptoms and functional status improved significantly after successful pericardiectomy. The patient worked in a shipyard, which means that exposure to muddy or stagnant water could have been the source of infection. Following hospital discharge, the patient was advised to avoid contact with contaminated soil or water and to use infection prevention precautions in future, including the wearing of a mask, gloves, boots, and a gown, whenever possible, to minimize the risk of infection exposure at work. This case highlighted the importance of serial echocardiography in the diagnostic evaluation during the clinical course of melioidosis pericarditis and the advantage of multimodality cardiovascular imaging in clinical practice. Due to the lack of clinical research on this rare complication of melioidosis, it was not possible to determine the optimal timing for follow-up clinical re-evaluation. However, based on the clinical course of the patient, as shown in Figure 3, following the onset of melioidosis pericarditis, monthly clinical follow-up of between six months and 12 months including echocardiography would be advised.

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Conclusions

Constrictive pericarditis is a debilitating and potentially fatal complication of melioidosis pericarditis. Early detection of constrictive pericarditis is important because the complication is curable by pericardiectomy. Use of multimodality cardiovascular imaging is valuable to distinguish between constrictive pericarditis and restrictive cardiomyopathy. From the findings in this case, the authors recommend that, following the onset of pericarditis due to melioidosis, monthly clinical follow-up of between six months and 12 months including echocardiography is advised to detect potential complication of constrictive pericarditis

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Conflict of interest

None.

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