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Case report

Melioidosis mycotic aneurysm: An uncommon complication of an uncommon disease

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A B S T R A C T

Melioidosis is often considered an exotic and uncommon disease in most parts of the world. However it is an endemic disease in Southeast Asia and Northern Australia with an expanding distribution. Melioidosis can involve almost any organ and can deteriorate rapidly. In this report, we describe a rapidly fatal case of a mycotic aneurysm associated with melioidosis despite aggressive antibiotic therapy. The morbidity and mortality of this uncommon complication remains high despite prompt diagnosis and treatment. Especially when treating persistent/recurrent melioidosis, the physician's caution to the development of mycotic aneurysms is imperative so that early treatment and surgical intervention may be considered.

Introduction

Melioidosis is a granulomatous infectious disease caused by the aerobic Gram-negative bacillus *Burkholderia pseudomallei* usually found in contaminated water and soil. It is considered a rare disease in most parts of the world but is endemic in Southeast Asia and Northern Australia. It often presents as community-acquired pneumonia and sepsis, but can involve almost any part of the body. In this case report, we illustrate a rare but fatal complication of this increasingly reported and important disease.

Case report

An 82 year old man presented with fever and cough for a few days. He was an ex-smoker and ex-social drinker, and had history of myocardial infarction treated by coronary angioplasty with stenting 4 years ago. The patient was a retired water pipe worker with good functional status. There was no history of diabetes, renal or lung disease.

Upon admission, chest x-ray (CXR) showed right lower zone consolidation and blood/urine biochemistry revealed hyponatremia consistent with syndrome of inappropriate antidiuretic hormone secretion (SIADH). Complete blood count (CBC) was unremarkable but inflammatory markers (erythrocyte sedimentation rate [ESR], C-reactive protein [CRP]) were elevated. The case was treated as community-acquired pneumonia and he was started on empirical intravenous amoxillcin + clavulanate and azithromycin. He improved with treatment but blood culture yielded *B. pseudomallei* (sensitive to ceftazidime). Computed tomography (CT) scan showed bilateral lower and right middle lobe consolidation; the abdomen and pelvis were unremarkable and there was no evidence of other organ involvement. He was treated with a three week course of intravenous ceftazidime and then switched to an eradication phase of oral amoxillin + clavulante and doxycycline for three more months. There was complete resolution of symptoms and serial CXRs were clear. Repeat blood culture showed clearance of *B. pseudomallei*. Reassessment CT performed in August 2014 showed largely resolved consolidative changes. The patient completed the course of oral antibiotics in September 2014. He remained well and there was no significant travel or contact history.

However he experienced fever and cough again three weeks after cessation of antibiotic therapy. CXR on presentation showed no new abnormalities, but blood tests again confirmed SIADH with raised inflammatory markers and *B. pseudomallei* septicaemia (sensitive to ceftazidime). He was restarted on ceftazidime but his symptoms and fever persisted. Ceftazidime was switched to meropenam but still without signs of clinical improvement. Serial blood cultures showed persistent *B. pseudomallei* septicaemia without change in antibiotic susceptibility. Whole body positron emission tomography–CT (PET-CT) was performed, which revealed

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a new heterogeneous hyperdense mass at the left side of the superior mediastinum associated elevated fluorodeoxyglucose (FDG) uptake, consistent with mycotic aneurysm of the aortic arch/left subclavian artery; and patchy FDG uptakes are noted within the vertebral bodies of the cervical and thoracic spine, suggestive of possible bone involvement (Fig. 1). Echocardiogram showed a structurally normal heart and no vegetations were seen. The dihydrorhodamine reduction (DHR) test revealed a low oxidative burst response.

In view of possible intravascular/bone involvement, the meropenam dose was escalated and doxycycline was added but still without clinical improvement. The patient was assessed by our cardiothoracic and vascular teams, but was deemed not suitable for surgery. Doxycycline was replaced by intravenous minocycline, and moxifloxacin was also added. However blood cultures were persistently positive for *B. pseudomallei* and the patient continued a downhill course. Serial CXR and CT (Fig. 2) showed rapid interval enlargement of aortic arch aneurysm. Despite continued antibiotic and supportive treatment, the patient complained of sudden onset of neck pain which was followed by massive haemoptysis. He succumbed on the same day, just within three weeks since his presentation of melioidosis relapse.

Discussion

Melioidosis is a granulomatous infectious disease caused by the aerobic Gram-negative bacillus *B. pseudomallei*. It is an environmental organism found in soils (classically rice paddy fields) and contaminated waters. It is endemic in Southeast Asia and Northern Australia with an expanding geographical distribution [1]. Like many soil bacteria, it is a difficult organism to kill, with an incubation period ranging from 2 days to 26 years [2] and can survive even in distilled water for up to 16 years [3]. Our patient was born in Hong Kong and had never resided in a village with rice paddy fields. It is likely that our patient was infected locally, given the increasing reports of melioidosis in our locality [1,4] as well his previous occupation as a water pipe worker. Although often presenting as community-acquired pneumonia and sepsis, melioidosis can involve almost any part of the body. In this case report, we illustrate a rare but fatal complication of this increasingly reported and important disease.

The disease can manifest as either acute or chronic forms and has a wide range of presentations. Acute disease is defined as symptoms lasting for less than two months and chronic disease is defined as symptoms persisting for longer than two months [5]. Acute melioidosis is more common and, such as in this case, around half of patients present with chest complaints and are often indistinguishable from other forms of community-acquired pneumonia. Other common presentations include skin infections, genitourinary infections, osteomyelitis/septic arthritis and internal organ abscesses (especially prostatic, splenic, kidney and liver abscesses) [6]. Acute suppurative parotitis is a characteristic manifestation of melioidosis amongst children in Thailand, and has been reported to account for around a third of all paediatric cases [7].

Melioidosis is also a rare but well documented cause of mycotic aneurysms [8–10] and is described to occur in around 1% of affected patients. In fact, *B. pseudomallei* was reported as the most common causative agent of mycotic aneurysm in North-eastern Thailand [10]. As with other causes of mycotic aneurysms, the proposed mechanism is haematogenous seeding of previously damaged arteriosclerotic vessels. Prolonged antibiotic therapy with surgical intervention is considered the most appropriate management for melioidosis-associated mycotic aneurysms. *B. pseudomallei* is intrinsically resistant to many antibiotics and current recommendations for antibiotic treatment includes initial an intravenous intensive phase of at least 10–14 days and followed by an oral eradication phase [11]. Usual first-line therapy for the intensive phase of treatment includes ceftazidime or a carbapenem.

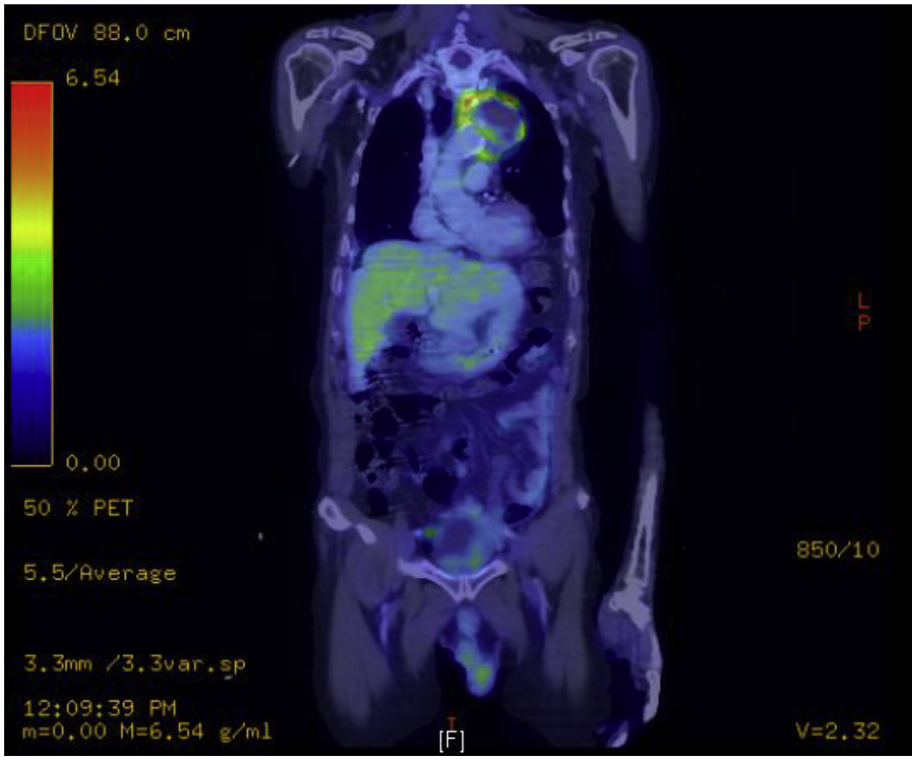

Fig. 1. PET-CT showing hypermetabolic mass at the left side of the superior mediastinum, consistent with mycotic aneurysm of the aortic arch/left subclavian artery.