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CASE REPORT

An adolescent with type 1 diabetes and coinfection with melioidosis and COVID-19: a case report

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Abstract

Melioidosis is a rare and potentially fatal infection, emerging in Brazil and endemic in Ceará, caused by the bacterium *Burkholderia pseudomallei*, which requires a high index of clinical suspicion for its diagnosis. It can manifest acutely, chronically or latently and, in general, in symptomatic patients there is sepsis with or without pneumonia. Diabetes mellitus is the main known risk factor. We present the case of a diabetic adolescent diagnosed with septicemic melioidosis and co-infection with COVID-19.

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INTRODUCTION

Melioidosis is an infection caused by a gram-negative, non-spore-forming bacillus first described in 1911, known as Burkholderia pseudomallei. It can be found in soil and water, predominantly in tropical and subtropical regions. It is endemic in northern Australia and parts of Southeast Asia, and affects both animals and humans[1]. Sporadic cases have been reported in Central and South America[2]. The main routes by which B. pseudomallei is acquired are inoculation through the skin, inhalation, and ingestion. Infection manifests in acute, chronic, or latent forms. Only symptomatic patients are considered to have developed melioidosis. In most cases, immunocompetent individuals develop subclinical disease only. Sepsis, with or without pneumonia, is seen in individuals with the disease[3]. The most significant risk factor for melioidosis is diabetes mellitus (DM), along with thalassemia, chronic kidney disease, chronic lung disease, alcoholism, liver cirrhosis, and immunosuppression[4]. Although individuals of all ages are affected, cases of the disease in children are less common. Incidence may be higher than recorded, due to diagnostic difficulties and underreporting[4]. Mortality is lower, the association with immunosuppression is weaker, and cases of localized infection occur more frequently in pediatric patients when compared to adults[5]. Melioidosis has been considered an emerging disease in Brazil since 2003, when the first case was diagnosed based on culture results. The Brazilian Northeast is considered an endemic zone[6]. In addition to Ceará, two other Brazilian states have confirmed cases of the disease: Mato Grosso and Alagoas[7]. In 2005, Ceará declared melioidosis a notifiable disease[8].

CASE REPORT

A 16-year-old young man with type 1 diabetes mellitus (T1DM) residing in a town in the countryside of the state of Ceará came to our service complaining of a dry cough lasting for 20 days, myalgia for 14 days, fatigue and fever. He sought care at an emergency unit in his hometown, and was referred to a tertiary pediatric hospital. He had tachydyspnea, hypoxemia, dehydration, and pulmonary auscultation with bibasilar crackles on admission. Treatment for community-acquired pneumonia was started with ceftriaxone, oxacillin, azithromycin, and oseltamivir, and therapeutic measures for diabetic ketoacidosis were introduced. The patient was intubated and referred to the pediatric intensive care unit (PICU). COVID-19 was considered in differential diagnosis, since the patient developed severe acute respiratory syndrome and the pandemic was at its peak at the time. Laboratory tests on admission showed leukopenia, thrombocytopenia, arterial blood gas levels indicative of severe metabolic acidosis, and elevated C-reactive protein (CRP), transaminase, lactate dehydrogenase, and ferritin levels. A rapid test for COVID-19 came back negative, but serology and molecular tests were positive. X-ray (Figure 1A) and chest tomography (CT) scans (Figures 1B and 1C) showed airspace nodules distributed diffusely in the lungs and consolidation in the lower portion of the lungs. The patient went into refractory shock 24 hours later and was prescribed vasoactive drugs. Multisystem inflammatory syndrome in children (MIS-C) was considered due to the severity of symptoms, and the patient was started on dexamethasone, albendazole, and piperacillin-tazobactam due to the possibility of Pseudomonas aeruginosa infection and because he was diabetic. Transthoracic echocardiogram findings suggestive of mild left ventricular dysfunction combined with laboratory tests and symptoms suggestive of MIS-C led to a prescription of intravenous human immunoglobulin. After four days of treatment, the patient still had daily fever spikes and was prescribed meropenem. During this period, he presented with pancytopenia on complete blood count and elevated CRP. A myelogram showed histiocytes with hemophagocytic activity. Vancomycin and amikacin were added, in addition to fluconazole for axillary skin lesions suggestive of fungal infection. He presented with bilateral pleural effusion requiring bilateral chest drainage due to empyema and hypertensive pneumothorax. Additional imaging exams ordered in preparation for surgery showed bilateral cavitated lesions (Figures 1D, 1E and 1F). The patient underwent a lung decortication with video-thoracoscopy and had two chest drains placed to the right. He was extubated 14 days later. The patient was given meropenem for 14 days and became afebrile after five days of treatment. Fever resumed after the medication was discontinued. The possibility of a fungal infection (coccidioidomycosis) was considered, since the patient had gone armadillo hunting two weeks prior to the onset of symptoms. He was prescribed amphotericin B. Laboratory tests worsened and he developed anemia, leukocytosis, and elevated CRP. Blood cultures were ordered and treatment with levofloxacin and amikacin was initiated. He said he had bathed in a water reservoir around the time he had gone armadillo hunting. Blood cultures showed the presence of Burkholderia pseudomallei, which caused the discontinuation of medication and the reintroduction of meropenem. He still had fever 48 hours later, and was started on trimethoprim-sulfamethoxazole (TMP-SMX). The patient complained of pain in the left hip when he tried to walk. Magnetic resonance imaging showed signs of osteomyelitis. The patient underwent left hip surgery and progressed well. He had no fever after 48 hours of treatment with TMP-SMX. He was given intravenous antibiotic therapy for eight weeks and remained on TMP-SMX for six months. The patient was followed in the outpatient clinic and had control imaging tests (chest X-ray and CT) performed for about 12 months, which showed slight scar tissue alterations (Figure 2). He is currently asymptomatic.

DISCUSSION

Melioidosis is an emerging infectious disease that is rare and difficult to diagnose. Inhalation is the main route of development of lung infection[4]. Tests are required to confirm

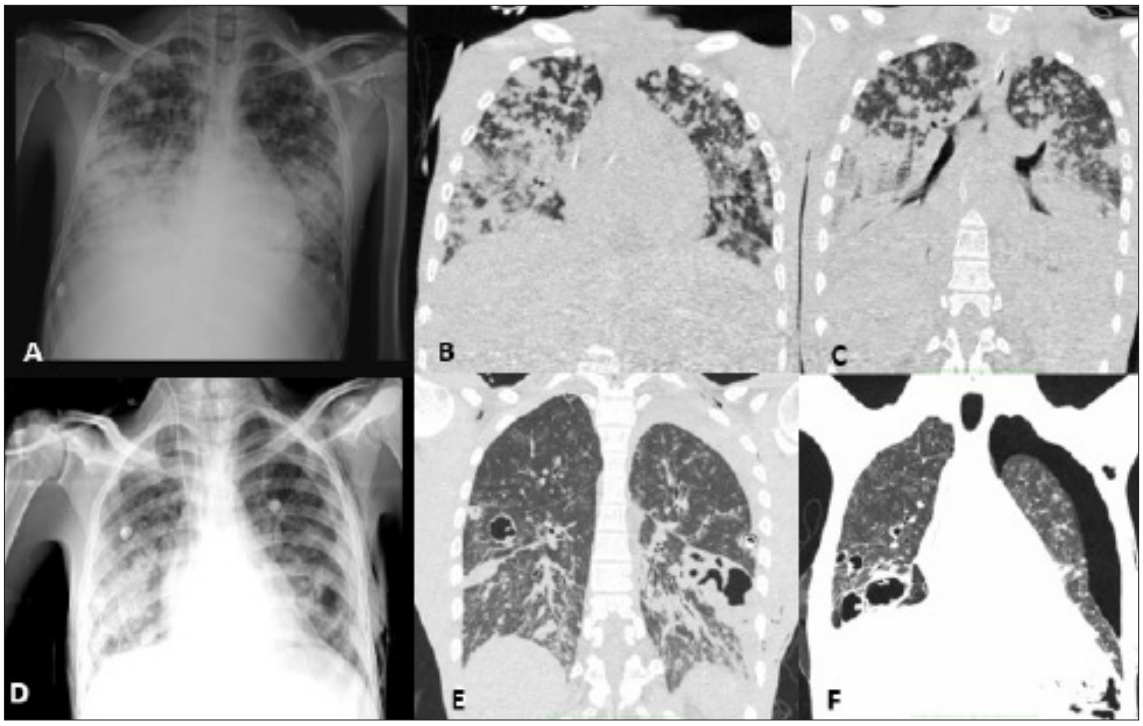


Figure 1. A. A. Anteroposterior chest radiograph: ill-defined nodules distributed diffusely throughout both lungs, with consolidations in the lower lung views. B. Coronal reconstruction with lung window in the anterior plane: multiple airspace nodules in the upper lobes, middle lobe and lingula. C. Coronal reconstruction with lung window in the posterior plane: airspace nodules in the upper lobes with consolidations in the lower lobes, very similar to the one seen in the chest x-ray image. D. Anteroposterior chest radiograph: residual consolidation in the lower third of the right lung with permeating pneumatoceles. Pneumatoceles are also present in the lower third of the left lung. Chest drain on the left with minor laminar pleural effusion and subcutaneous emphysema. E. Coronal reconstruction with lung window in the posterior plane: multiple ground-glass nodules showing a true abscess in the lower left lobe. F. Coronal reconstruction with lung window in the anterior plane: multiple diffuse ground-glass nodules showing nodular cavitation/necrosis in the middle lobe and laminar pneumothorax on the left. X-ray/computed tomography scans taken in later moments show attenuation of nodules in the airspace into small centrilobular ground-glass opacities, resolution of consolidations of the lower lobes and development of cavitated nodules.

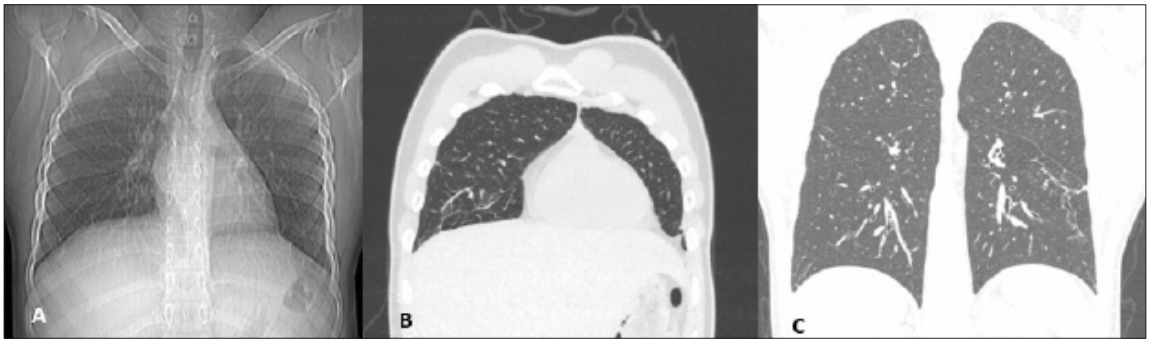


Figure 2. Anteroposterior radiograph: normal findings. B. Anterior coronal reconstruction showing scar parenchymal bands in the middle lobe. C. Posterior coronal reconstruction showing scar parenchymal bands interspersed with mild traction bronchiectasis in the lower left lobe.

diagnosis and culture is the gold standard test. Although it grows in most culture media, the bacterium causing the disease may not be recognized and can be confused with other bacteria, in addition to also being considered as a source of contamination[3]. In the case described, bacterial growth was observed in blood and urine samples from admission, but there was a delay in diagnosis because it was first identified as other bacteria (Burkholderia cepacia and Pseudomonas aeruginosa). The positive molecular test for COVID-19 masked the diagnosis, and given the severity of symptoms, the patient was treated for MIS-C. After the culture results were out, specimens were sent to the Central Public Health Laboratory (LACEN) for confirmation with a polymerase chain reaction (PCR) kit produced by VITEK, as stipulated in the protocol in effect in the state of Ceará. Melioidosis does not respond to the antimicrobials used in the treatment of community-acquired infections, and requires the use of specific drugs such as ceftazidime or meropenem. In addition, prolonged oral treatment to eradicate the bacteria is required for complete resolution[9]. A recent study that included 20 children suspected with or with a confirmed diagnosis of melioidosis reported a significant mortality rate, severe symptoms, and considerable environmental exposure. This indicates that melioidosis in children may be more severe in Brazil, differently from previous reports in the literature[8].

CONCLUSION

This report described the case of an adolescent with an infection rarely identified in Brazil, which is however endemic in the Brazilian Northeast and an emerging condition in our country. Melioidosis must be considered in the differential diagnosis of patients with severe pneumonia of apparent community origin who do not respond to the usual antimicrobials, with risk factors for the disease, and environmental exposure. Considering melioidosis is important, so that biological material is collected and sent to a certified laboratory, and treatment is initiated as early as possible, in such a way to decrease morbidity and mortality.

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