Respiratory Medicine Case Reports 16 (2015) 54–56

Contents lists available at ScienceDirect

Respiratory Medicine Case Reports

journal homepage: www.elsevier.com/locate/rmcr

Case report

Pulmonary melioidosis presenting with pleural effusion: A case report and review of literature

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ARTICLE INFO

Article history:
Received 1 May 2015
Received in revised form
19 July 2015
Accepted 20 July 2015

Keywords:
Pneumonia
Pleural effusion
Burkholderia pseudomallei
Melioidosis

ABSTRACT

Melioidosis is a serious infection, which can involve multiple systems. We report a case of pulmonary melioidosis with the initial presentation mimicking a partially treated pneumonia complicated by right-sided pleural effusion. The patient is a 49-year old man who did not respond to parenteral ceftriaxone and tazobactam/piperacillin therapy. However, upon culture and sensitivity results from blood and pleural samples isolated *Burkholderia pseudomallei*; antimicrobial therapy was de-escalated to parenteral ceftazidime. Within 72 h duration, his fever subsided and other respiratory symptoms improved tremendously. This case highlights the importance of early recognition of *B. pseudomallei* in pulmonary infection in order for prompt institution of appropriate antibiotics treatment; thus reducing morbidity and mortality.

1. Introduction

Melioidosis is a common infection in the tropical countries including Malaysia. This infection is caused by environmental saprophyte-*Burkholderia pseudomallei*. It is a gram-negative bacterium commonly transmitted via direct contact with infected soil (i.e. gardening or swimming). Melioidosis can present acutely with fever and chills secondary to a localized infection or septicemia. Chronic cases are seen when abscesses developed insidiously at various tissue sites or organs. Thus treatment for persistent infection can be challenging [1]. Diabetes is the commonest concurrent condition identified; with a figure as high as 44% and 60% reported in Thailand and Malaysia respectively [2,3].

1.1. Case presentation

A 49-year-old man, non-smoker presented to our center with intermittent fever, productive cough with whitish sputum and right-sided pleuritic chest pain for one week duration. Prior to admission, he had received a course of oral antibiotic (erythromycin) for five days. Unfortunately, his symptoms did not improve. He has no past medical illness and works as a teacher. There was no recent history of contact with soil (i.e. gardening/planting), swimming in the river, traveling or jungle trekking. There were also no constitutional symptoms such as loss of appetite or loss of weight.

1.2. Examination

His temperature was 38.4 °C, heart rate was 100/min (regular rhythm), respiratory rate 20 breaths/min, blood pressure 103/59 mmHg, and his oxygen saturation was 96% on room air. Clinically, there was a moderate right-sided pleural effusion as evidence by a reduced in chest expansion, vocal resonance, tactile fremitus, stony dullness on percussion and a reduced air entry on auscultation. Bronchial breath sounds were heard at the right mid-zone. Otherwise, there were no lymphadenopathy or organomegaly detected.

1.3. Investigations

Blood test showed white blood cells of 12.0×10^9/L; predominantly neutrophils, 70.2%; fasting blood sugar of 17 mmol/L and glycated hemoglobin (HbAIC) level of 7.6%, erythrocyte sedimentation rate of 97 mm/h; C-reactive protein, 24 mg/dL; serum sodium, 129 mmol/L; potassium, 4.3 mmol/L; urea, 5.7 mmol/L; creatinine, 99umol/L and alanine aminotransferase (ALT), 72 U/L. Culture and sensitivity performed on three sets of sputum did not

Abbreviations: TMP-SMX, trimethoprim-sulfamethoxazole.

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http://dx.doi.org/10.1016/j.rmcr.2015.07.005

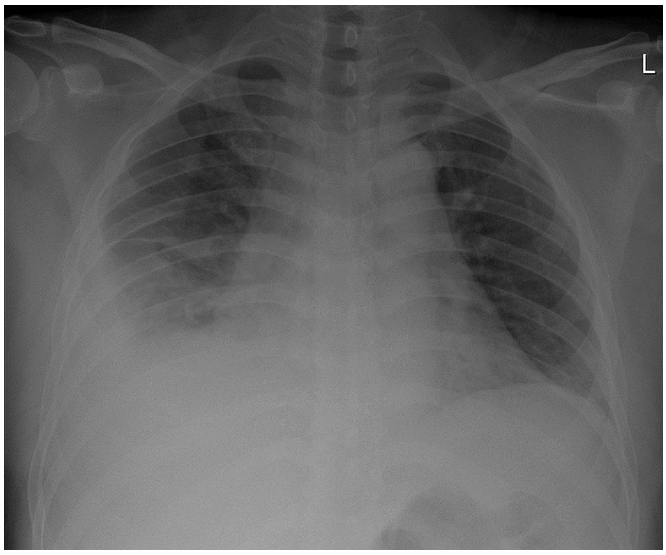

Fig. 1. Erect chest radiograph of the patient on initial presentation. Air-space heterogeneous opacities noted over the right lower zone accompany by a pleural effusion.

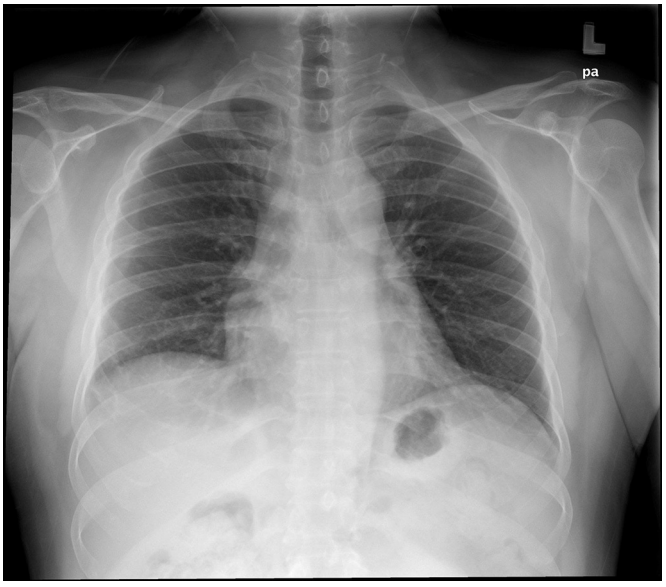

Fig. 2. Erect chest radiograph of the patient repeated after completion of treatment (1 month later).

yield any positive results. Chest radiograph (Fig. 1) showed air-space opacities at the right lower zone and blunting of the right costophrenic angle in keeping with infective process complicated with pleural effusion.

Pleurocentesis yield straw colored pleural fluid. Biochemical results of the pleural fluid were consistent with a complicated parapneumonic effusion. Pleural fluid lactate dehydrogenase (LDH), 2460 U/L; glucose, 7.6 mmol/l; and pH, 7.0. Pleural fluid cytology revealed numerous small, reactive lymphocytes with neutrophils and the presence of some red blood cells. Staining for acid-fast bacilli (AFB) was negative. Subsequently pleural drainage was carried out via an 8F pigtail catheter.

The patient was started on parenteral ceftriaxone and oral azithromycin. This was replaced with parenteral tazobactam/piperacillin in view of poor clinical response and persistent fever for more than 48 h. Subsequently, culture results of the pleural fluid and blood were reported positive for *B. pseudomallei*, which was sensitive to ceftazidime. Treatment was deescalated to parenteral ceftazidime 2 g 8hourly. Following this, the patient showed tremendous improvement and he became afebrile three days after initiation of ceftazidime. Further work-up revealed no occult abscesses detected on the abdominal sonography. On the other hand, the patient was also diagnosed to have type-2 diabetes mellitus. A good glycemic control was achieved after initiation of insulin therapy.

The patient was treated with two weeks duration of parenteral Ceftazidime. Subsequent white cell counts and C-reactive protein levels showed a reducing trend and measured 5.9×10^9/L and 5.83 mg/dL respectively upon discharge. The pigtail drainage catheter was removed on day six after drainage amount of less than 50 cc per day for two consecutive days. The total drainage was 1650 cc. Following this, the patient was given outpatient treatment with oral Doxycycline 100 mg 12 hourly and three tablets of trimethoprim/sulphamethoxazole 80/400 mg (TMP-SMX) 12 hourly with a total intended treatment duration of 20 weeks. On his review a month later, the patient was well with good glycemic control. A repeated chest radiograph (Fig. 2) showed resolution of the previous right-sided pleural effusion. He was discharged well from the respiratory outpatient clinic upon completion of his treatment.

2. Discussion

Melioidosis is endemic in Malaysia, Thailand, Singapore and Australia. A study conducted in the state of Pahang in Peninsula Malaysia demonstrated the incidence of this infection is approximately 6.1 per 100, 000 populations per year [4]. The lung is the most commonly affected organ in melioidosis infection. Patients can present with cough, fever or both as a result of pneumonia or a primary lung abscess [3]. A spectrum of pulmonary infiltrations is commonly seen; ranging from patchy to diffuse, lobar to multilobar involvement. Pulmonary melioidosis manifesting as isolated pleural effusion occurred in 12.2% of the total 162 cases reported between 1996 and 2002 in study conducted in Thailand [2]. However on the Malaysian front, How et al. mentioned that the incidence of Melioidosis reported in the state of Pahang in Peninsula Malaysia exceeded that of Thailand but unfortunately the data on the incidence of cases presented with isolated pleural effusion was lacking [4].

Melioidosis can present either acute, or runs a sub-acute course. It can mimic other diseases and may cause unnecessary delay in the initiation of appropriate antibiotics. Therefore, clinicians ought to be vigilant to consider less common etiologies of pneumonia or parapneumonic effusion if response to guideline-driven conventional intravenous antibiotics shows poor response. In this case, base on the history, clinical symptoms, pleural fluid analysis and chest radiograph findings the patient was initially treated for partially treated community-acquired pneumonia complicated with right sided parapneumonic effusion. His condition did not improve despite escalating antibiotics treatment. Clinical improvement was only observed after initiation of parenteral ceftazidime following successful isolation of *B. pseudomallei*.

A diagnosis of melioidosis requires isolation of *B. pseudomallei* from various clinical specimens. This can either be blood, tissue samples or aspirate cultures obtained from abscesses. *B. pseudomallei* is a motile (with polar flagella), aerobic gram-negative bacillus and grows readily on routine culture media. Often enough, it shows bipolar staining and is oxidase positive. Colonies of *B.*

pseudomallei vary in morphological features depending on the culture media and the type of strain isolated. They often become wrinkled in appearance after incubation for 2–3 days [5]. Alternative methods of detecting *B. pseudomallei* are using polyclonal (Pab-IFA) or monoclonal antibody (Mab-IFA). These tests can provide results as early as 24–48 h compare to blood culture. The positive predictive values for Mab-IFA and Pab-IFA are 93.9% and 72.5% respectively [6].

Important risk factors for Melioidosis are such as diabetes, renal disease, liver cirrhosis, thalassemia, alcoholism, use of immunosuppressive agents, cystic fibrosis and kava (a Hawaiian drink) consumption [7]. The antibiotic of choice is parenteral ceftazidime or a carbapenem drug (either meropenem or imipenem). It should be continued for a duration of at least 10 days follow by oral therapy with trimethoprim-sulfamethoxazole (TMP-SMX) monotherapy or combination with doxycycline for at least 12–20 weeks duration in the eradication phase with a lower risk of relapse in the later [8]. In contrast, guidelines from the northern territory of Australia suggested intensive phase with parenteral antibiotics should be given for at least 14 days duration. In the presence of severe infection, duration of treatment can be extended up to 4 weeks duration or parenteral carbapenem can be administered as an alternative drug of choice [9]. Eradication phase should be continued for a minimal period of three months duration [9].

Doxycycline has been used in combination with TMP-SMX for eradication therapy in some parts of the world. However, TMP-SMX appears to be the crucial component of this combination regimen. This was supported by the results of a multi-center, double-blind, randomized controlled trial conducted in Thailand, where there was no difference in the recurrent rates of culture-confirmed melioidosis in patients treated with either regime [8]. In this case, the patient was discharged with a combination therapy of oral TMP-SMX and doxycycline for a total duration of 20 weeks according to local treatment protocol [10]. He achieved full recovery on subsequent outpatient review.

3. Conclusion

In conclusion, melioidosis is known to cause pulmonary infection and isolated pleural effusion can be the only manifestation. In countries where melioidosis is endemic, a high index of suspicion should be considered in all high-risk individuals, who showed poor response to initial parenteral antibiotics. Prompt microbiology results are undeniably important as well to guide early commencement of appropriate antibiotic therapy as stereotypically driven empirical antibiotic used is ineffective in treating Melioidosis and can influence the outcome and recovery of the patient.

Consent

Written informed consent was obtained from our patient for the publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

SAW, FAH, and SCI were all involved in the conception of the case report, review of literature and writing the manuscript. SAW provided collection of data. SCI participated in the final revision of the manuscript and guidance. All authors read and approved the final manuscript.

Authors' information

Sopian AW is an internal medicine registrar. Faisal AH and Soo CI are both respiratory subspecialty registrars. All authors are currently practicing physicians at the Universiti Kebangsaan Malaysia (UKM) Medical Center, Kuala Lumpur, Malaysia.

Acknowledgment

We would like to thank the patient for giving us permission to publish this case report.

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