

MELIOIDOSIS PRESENTING WITH ISOLATED SPLENIC ABSCESES: A CASE REPORT

Chun-Yu Lin,¹ Tun-Chieh Chen,^{1,2} Po-Liang Lu,^{1,2} Wei-Ru Lin,¹ and Yen-Hsu Chen^{1,2}

¹Division of Infectious Disease, Department of Internal Medicine, Kaohsiung Medical University Hospital, and ²Graduate Institute of Medicine, College of Medicine, Kaohsiung Medical University, Kaohsiung, Taiwan.

Splenic abscesses caused by *Burkholderia pseudomallei* are rarely reported in Taiwan. Here we report a middle-aged man who presented with fever, chills, and general malaise for several days. Abdominal echo revealed isolated splenic abscesses and he received antibiotics treatment according to the initial blood culture result, *Serratia marcescens*. However, fever did not subside. Then he was referred to our hospital and meropenem was prescribed. Fever subsided 5 days after the beginning of meropenem administration. Repeated fine-needle aspiration of splenic abscesses drained out the pus, which was cultured as *B. pseudomallei*. He was finally diagnosed as a case of melioidosis based on microbiological evidence. Physicians must take melioidosis into consideration when splenic abscesses are encountered clinically.

Key Words: *Burkholderia pseudomallei*, melioidosis, splenic abscess
(*Kaohsiung J Med Sci* 2007;23:417–21)

Splenic abscesses are relatively uncommon, but untreated splenic abscess has a high mortality rate [1]. Autopsy series have placed the incidence of splenic abscesses at 0.2–0.7% of all intra-abdominal abscesses [2,3]. The bacterial pathogens may be polymicrobial, monomicrobial, or sterile. In Taiwan, the most common pathogen is *Klebsiella pneumoniae*, followed by *Escherichia coli* and *Salmonella* species [4].

Melioidosis is an infectious disease caused by *Burkholderia pseudomallei*, a Gram-negative bacillus with bipolar staining. It is often described as having a “safety pin” appearance [5]. Melioidosis is regarded as endemic to Southeast Asia and Northern Australia, corresponding approximately to the tropical latitudes between 20°N and 20°S. Melioidosis is known for its propensity to cause abscesses, and may cause liver

abscesses with or without concomitant splenic abscesses [6].

In Thailand, *B. pseudomallei* is an important pathogen causing splenic abscess [7]. However, this organism has been rarely reported to cause splenic abscess in Taiwan. Here, we report a case of melioidosis presenting with isolated splenic abscesses without concomitant liver abscess.

CASE PRESENTATION

A 54-year-old male building security guard, who lived and worked in Ling-Ya district, Kaohsiung city and had never traveled overseas, presented with intermittent fever and shaking chills associated with poor appetite and malaise since April 2, 2005. He had diabetes mellitus diagnosed 3 years earlier and hypertension diagnosed 2 years earlier. He had received regular medical treatment, and had received anti-tuberculosis agents for 6 months for pulmonary tuberculosis 2 years earlier.

He visited a regional hospital on April 6, 2005, due to persistent fever and chills. At the emergency

Received: December 13, 2006 Accepted: January 11, 2007
Address correspondence and reprint requests to: Dr Yen-Hsu Chen, Division of Infectious Disease, Department of Internal Medicine, Kaohsiung Medical University Hospital, 100 Tzyou 1st Road, Kaohsiung 807, Taiwan.
E-mail: d810070@cc.kmu.edu.tw

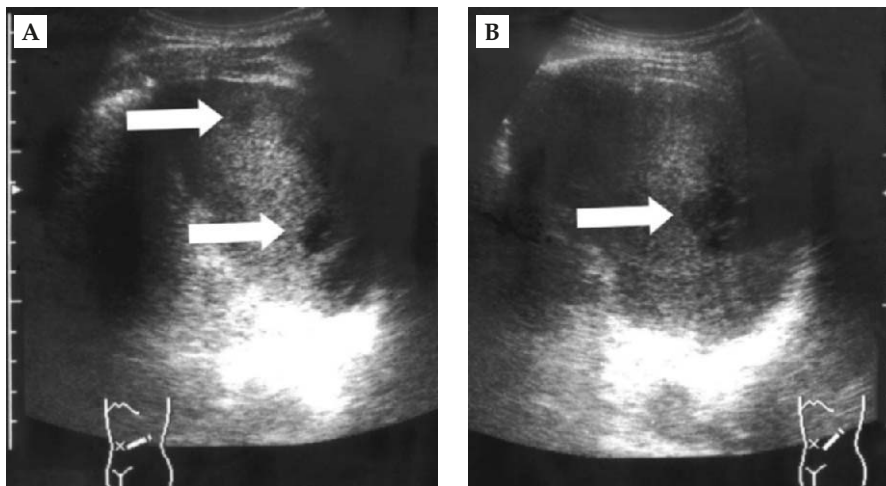


Figure 1. (A,B) Multiple splenic abscesses of various diameters (arrows).

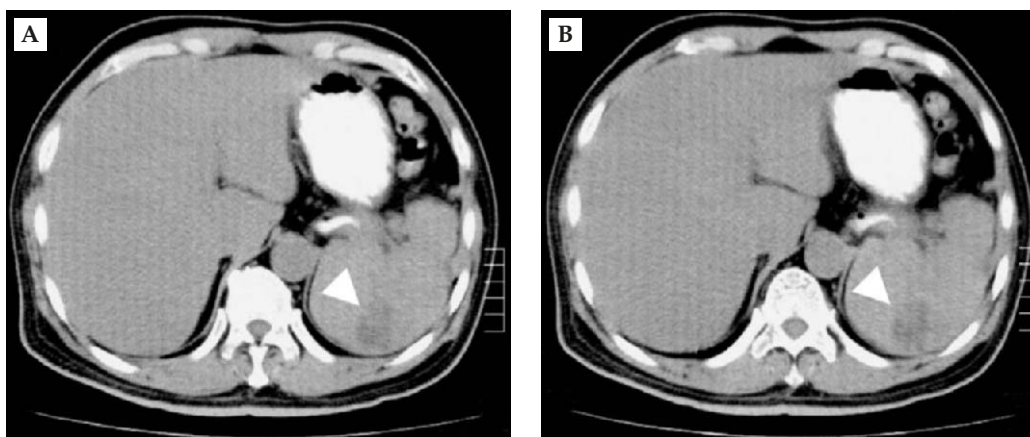


Figure 2. (A,B) Multiple splenic abscesses (arrowheads) without space-occupying lesion over liver parenchyma.

department, his tympanic membrane temperature was 39.2°C. The physical examination was grossly normal. Laboratory examination showed leukocytosis with a white blood cell (WBC) count of $11.2 \times 10^9/L$ with differential count of segment, 78%; band, 2%; lymphocytes, 20%. Hemoglobin was 13.6 g/dL and platelet count was $237 \times 10^9/L$. The other laboratory results showed abnormal liver function test: aspartate transaminase (AST), 69 U/L; alanine transaminase (ALT), 75 U/L; high C-reactive protein (CRP) concentration, 183 $\mu\text{g}/\text{mL}$; blood urea nitrogen (BUN), 21 mg/dL; and serum creatinine, 1.55 mg/dL. Chest radiograph on arrival at the regional hospital was grossly normal.

After blood sampling for bacterial culture, he received antimicrobial agents including intravenous cephalexin and gentamicin. Both abdominal sonography and computed tomography (CT) scan revealed multiple splenic abscesses of various diameters, but

no space-occupying lesion over liver parenchyma (Figures 1 and 2). Echo-guided fine-needle aspiration of splenic abscesses was done on April 9, 2005 and 5 mL viscous yellowish pus was obtained. Gram's stain of pus showed Gram-negative bacilli. Blood culture (BACTEK-9240) reported *Serratia marcescens* on April 11, 2005. The susceptibility test showed resistance to cefazolin and gentamicin but sensitivity to ceftazidime. The antimicrobial agents were shifted to ceftazidime and isepamicin, but the fever did not subside.

He was transferred to our hospital on April 21, 2005. On arrival at the emergency department, his tympanic membrane temperature was 38.2°C. The laboratory data showed WBC count, $11.03 \times 10^9/L$; hemoglobin level, 10.7 g/dL; platelet count, $374 \times 10^9/L$; AST, 40 U/L; ALT, 49 U/L; CRP, 74.2 $\mu\text{g}/\text{mL}$; BUN, 20 mg/dL; and serum creatinine, 1.3 mg/dL. Two grams of meropenem were given every 8 hours

because of the possibility of polymicrobial infection. Fever subsided 5 days after the beginning of meropenem administration.

Following abdominal sonography on the 15th day after initiation of meropenem treatment, non-resolution of splenic abscesses was noted and fine-needle aspiration was performed again. The pus culture yielded *B. pseudomallei*. An extended antimicrobial therapy course was planned according to previous therapeutic experience [5] but the patient refused. The patient was followed up 3 months later by telephone and neither fever nor malaise developed. We suggested that he come back to our outpatient department and receive further imaging study; however, he refused. Hence, there is no follow-up sonography or CT scan of the spleen available.

DISCUSSION

Splenic abscess is a rare condition of intra-abdominal abscesses, with a reported frequency in autopsy series between 0.2% and 0.7% [2,3]. In Taiwan, the common causative pathogens of splenic abscesses are *K. pneumoniae*, *E. coli*, *Salmonella* species, *Pseudomonas* species, *Proteus* species, *Staphylococcus* species, *Streptococcus* species, *Enterococcus* species, *Propionibacterium acnes*, *Bacteroides fragilis*, *Mycobacterium tuberculosis*, and fungi [4,8]. These pathogens may cause solitary splenic abscess, multiple splenic abscesses with or without extrasplenic abscess. Lee et al [6] had once reported *B. pseudomallei* as causing splenic abscess associated with concomitant liver abscess in Taiwan. However, *B. pseudomallei* has never been reported as causing isolated splenic abscess (without extrasplenic abscess) in Taiwan.

A series in Thailand reported 60 patients with splenic abscess and the most common pathogen was *B. pseudomallei*. Furthermore, it disclosed that multiple splenic abscesses were more commonly found in the melioidosis group than in the non-melioidosis group [7]. Another series in Singapore reported 28 patients with splenic abscess and the causative organisms were *Staphylococcus aureus*, mycobacteria, *Streptococcus* species, fungi, and *B. pseudomallei* [9]. The risk factors of splenic abscesses in these series included diabetes mellitus, leukemia, human immunodeficiency virus infection, intravenous drug abuse, and steroid therapy exposure.

Melioidosis is common in Southeast Asia. Recently, it has also been increasingly recognized in Taiwan [10,11]. The pathogen, *B. pseudomallei*, is an aerobic Gram-negative bacillus distributed in moist soil and water in endemic areas. It is the leading cause of community-acquired pneumonia, liver and splenic abscesses, and sepsis in northeastern Thailand [12]. The clinical manifestation of melioidosis is diversified. Although it is characterized by granulomatous disease, its clinical presentation ranges from subclinical infection to severe and fatal disease. The most frequently involved sites include lung, blood stream, liver, spleen and so on. Melioidosis should be considered as a possibility when abscesses are encountered at unusual sites in an endemic area [13].

We report on a middle-aged man suffering from splenic abscesses who was finally diagnosed as a case of melioidosis based on microbiological evidence in Taiwan. The initial blood culture reported *S. marcescens*, which was resistant to ampicillin, cefazolin, cefmetazole, ceftriaxone, gentamicin, amikacin, co-trimoxazole, and levofloxacin, and only sensitive to ampicillin/sulbactam, ceftazidime, and imipenem. The clinical presentation did not improve 9 days after the administration of ceftazidime. This may have been due to the slow response of melioidosis to ceftazidime (median time to abatement of fever about 9 days) [14]. His fever did not subside after the first percutaneous splenic abscess aspiration. This finding suggested that appropriate antimicrobial therapy plays an important role in the management of splenic abscesses. On the other hand, fever subsided after the second splenic abscess aspiration. It also suggested that defervescence may be partially attributed to percutaneous splenic abscess aspiration. In addition to antimicrobial agent therapy, no conclusive recommendations about percutaneous splenic aspiration or splenectomy were made because there is a lack of well-designed studies demonstrating the superiority of any therapy over others [4].

Several beta-lactams, such as meropenem, reduce the mortality of melioidosis, and long courses of co-trimoxazole-containing regimens are needed to prevent relapse [15]. This patient received meropenem as intensive-phase therapy and showed clinical improvement. However, he did not receive a long course of co-trimoxazole-containing regimens as an eradication-phase therapy. Hence, relapse may occur in the future.

In the laboratory setting, *B. pseudomallei* may be misidentified as *Klebsiella* species, *Pseudomonas aeruginosa*,

Enterococcus faecalis, *B. cepacia*, or *S. marcescens* [5]. This may explain the initial blood culture report (at the regional hospital) of *S. marcescens*, which should not reveal aminoglycoside resistance in a community-acquired infection. That means, *B. pseudomallei* will reveal resistance to aminoglycoside but *S. marcescens* will not. On the other hand, the susceptibility test of co-trimoxazole for the isolate showed resistance at the regional hospital. This was a problem related to disk diffusion methods. Minimal inhibitory concentration test was recommended for susceptibility test of co-trimoxazole and demonstrated a much lower rate of resistance (3–10%) [5].

B. pseudomallei is present in soil and surface water in the endemic region. The modes of acquisition are inhalation, inoculation, and rarely ingestion from a contaminated environment [16]. The patient might have contracted this pathogen from previous environmental exposure. According to the clinical course of this patient, no definite route of transmission (portal of entry) could be traced, since the patient resided near the possible endemic region as previously reported by Chen et al [17]. The risk factors for this patient to get melioidosis included diabetes mellitus and tuberculosis. Other major risk factors of melioidosis included soil/water exposure history, chronic renal disease, excessive alcohol consumption, and steroid use [5]. Although infective endocarditis is the most common condition predisposing a patient to splenic abscess [18], there was no evidence supporting this diagnosis for our patient.

Melioidosis presented with hepatosplenic abscesses may reveal a characteristic “honeycomb” appearance (multiseptate multiloculated lesions) on CT scans. For our patient, this type of image appearance was not found. However, such a finding should prompt physicians to include *B. pseudomallei* infection in the differential diagnoses of liver and splenic abscesses and consider initiation of empirical therapy for melioidosis in high-risk patients from areas of endemicity [19].

In conclusion, splenic abscess is a rare clinical condition, which is rarely caused by *B. pseudomallei* in Taiwan. We report this case of isolated splenic abscesses caused by *B. pseudomallei* as a reminder that melioidosis should be included in the differential diagnosis of patients with isolated splenic abscess, especially for those with predisposing factors and presenting with multiple splenic abscesses in the endemic region of Taiwan.

REFERENCES

1. Nelken N, Ignatius J, Skinner M, et al. Changing clinical spectrum of splenic abscess: a multicenter study and review of the literature. *Am J Surg* 1987;154:27–34.
2. Sarr MG, Zuidema GD. Splenic abscess: presentation, diagnosis, and treatment. *Surgery* 1982;92:480–5.
3. Chulay JD, Lankerani MR. Splenic abscess: report of 10 cases and review of the literature. *Am J Med* 1976; 61:513–22.
4. Lee CH, Leu HS, Hu TH, et al. Splenic abscess in southern Taiwan. *J Microbiol Immunol Infect* 2004;37: 39–44.
5. Cheng AC, Currie BJ. Melioidosis: epidemiology, pathophysiology, and management. *Clin Microbiol Rev* 2005;18:383–416.
6. Lee YL, Lee SJ, Tsai HC, et al. Pyogenic liver abscess caused by *Burkholderia pseudomallei* in Taiwan. *J Formos Med Assoc* 2006;105:689–93.
7. Sangchan A, Mootsikapun P, Mairiang P. Splenic abscess: clinical features, microbiologic finding, treatment and outcome. *J Med Assoc Thai* 2003;86:436–41.
8. Chang KC, Chuah SK, Changchien CS, et al. Clinical characteristics and prognostic factors of splenic abscess: a review of 67 cases in a single medical center of Taiwan. *World J Gastroenterol* 2006;12:460–4.
9. Joazlina ZY, Wastie ML, Ariffin N. Computed tomography of focal splenic lesions in patients presenting with fever. *Singapore Med J* 2006;47:37–41.
10. Lu PL, Tseng SH. Fatal septicemic melioidosis in a young military person possibly co-infected with *Leptospira interrogans* and *Orientia tsutsugamushi*. *Kaohsiung J Med Sci* 2005;21:173–8.
11. Ben RJ, Tsai YY, Chen JC, et al. Non-septicemic *Burkholderia pseudomallei* liver abscess in a young man. *J Microbiol Immunol Infect* 2004;37:254–7.
12. Thummakul T, Wilde H, Tantawichien T. Melioidosis, an environmental and occupational hazard in Thailand. *Mil Med* 1999;164:658–62.
13. Mathew S, Perakath B, Mathew G, et al. Surgical presentation of melioidosis in India. *Natl Med J India* 1999;12:59–61.
14. White NJ. Melioidosis. *Lancet* 2003;361:1715–22.
15. Dance DA. Melioidosis. *Curr Opin Infect Dis* 2002;15: 127–32.
16. Currie BJ. Melioidosis: an important cause of pneumonia in residents of and travellers returned from endemic regions. *Eur Respir J* 2003;22:542–50.
17. Chen WT, Chen YS, Chye SM, et al. Seroprevalence of melioidosis in diabetic patients in Taiwan. *J Microbiol Immunol Infect* 2005;38:267–70.
18. Johnson JD, Raff MJ, Barnwell PA, et al. Splenic abscess complicating infectious endocarditis. *Arch Intern Med* 1983;143:906–12.
19. Apisamthanarak A, Apisamthanarak P, Mundy LM. Computed tomography characteristics of *Burkholderia pseudomallei* liver abscess. *Clin Infect Dis* 2006;42:989–93.

以單純脾臟膿瘍作為臨床表現的 類鼻疽個案

林俊祐¹ 陳惇杰^{1,2} 盧柏樑^{1,2} 林蔚如¹ 陳彥旭^{1,2}

¹高雄醫學大學附設醫院 內科部感染內科

²高雄醫學大學 醫學院醫學研究所

在台灣地區，脾臟膿瘍鮮少被報導由 *Burkholderia pseudomallei* 造成。在這裡我們報導一位中年男性，呈現連續數天之發燒、寒顫及全身倦怠。腹部超音波檢查發現單純脾臟膿瘍。起初的血液培養結果為 *Serratia marcescens*，這位病患也據此接受了 ceftazidime 的治療。然而，發燒並未緩解。此後，這位患者被轉診到本院。我們投予 meropenem 五天後發燒緩解。後續經脾臟膿瘍抽吸出的膿液培養出 *Burkholderia pseudomallei*，因此我們根據微生物學的證據，診斷這位患者是一個類鼻疽的個案。在台灣，當臨床醫師發現脾臟膿瘍時，必須考慮類鼻疽的可能性。

關鍵詞： *Burkholderia pseudomallei*，類鼻疽，脾臟膿瘍

(高雄醫誌 2007;23:417-21)

收文日期：95 年 12 月 13 日

接受刊載：96 年 1 月 11 日

通訊作者：陳彥旭醫師

高雄醫學大學附設醫院內科部感染內科

高雄市807三民區自由一路100號