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Legionella community-acquired pneumonia (CAP) presenting with spontaneous bilateral pneumothoraces

Burke A. Cunha, MD, Francisco Miled Pherez, MD, and Yelda Nouri, MD

Legionnaires' disease is a common cause of non-zoonotic atypical community-acquired pneumonia (CAP). Legionnaires' disease has varied manifestations but may be diagnosed clinically on the basis of its characteristic pattern of extra-organ involvement. In a patient with non-zoonotic CAP, the clinical and laboratory features in a patient with CAP pointing to the diagnosis of Legionnaires' disease include relative bradycardia, mental confusion/ encephalopathy, loose stools/diarrhea, abdominal pain, mild/ transient increases in serum transaminases, decreased serum phosphorous, a highly elevated C-reactive protein (CRP), elevated creatinine phosphokinase (CPK), highly elevated serum ferritin levels, or microscopic hematuria. The radiologic manifestations of Legionnaires' disease are varied and no radiographic appearance is pathopneumonic. Patchy infiltrates in Legionnaires' disease are symmetrical and rapidly progressive even on appropriate anti-Legionella antimicrobial therapy. Spontaneous unilateral pneumothorax is a rare radiographic manifestation of Legionnaires' disease. We present a case of a young male who is presenting clinical finding was that of spontaneous bilateral pneumothoraces due to Legionella CAP. We believe this is the first reported case of Legionnaires' disease presenting as spontaneous bilateral pneumothoraces. Clinicians should be aware of the protean radiological manifestations of Legionnaires' disease. In patients presenting with CAP and unilateral or bilateral spontaneous pneumothorax, clinicians should have Legionnaires' disease in the differential diagnosis. (Heart Lung® 2008;37:238–241.)

Legionnaires' disease was first described after a pneumonia outbreak among American legion members attending a convention in Philadelphia in July of 1976.[1] To date, there are approximately 4000 reports in the literature describing the many different manifestations of Legionnaires disease.[2] Legionnaires' disease is a common cause of non-zoonotic atypical community-acquired pneumonia (CAP).[3] The radiologic manifestations of Legionnaire's disease are variable, but generally, infiltrates are often rapidly progressive and there is a lag in radiologic improvement compared with clinical improvement. Legionnaires' disease may also mimic other diseases because it may produce abscesses, cavitations, and even wedge-shaped opacities in immunosuppressed patients, mimicking pulmonary infarcts and embolism.[4-7] Spontaneous pneumothorax may be associated with a variety of infectious and noninfectious disorders.[8-14] Spontaneous pneumothorax is a rare presenting feature of *Legionella* CAP.[15,16]

To the best of our knowledge, this is the first report of bilateral spontaneous pneumothorax as the presenting radiological manifestation of *Legionella* CAP.

CASE REPORT

A 20-year-old man presented to the emergency department after he was found unresponsive. His family started cardiorespiratory resuscitation (CPR). He was intubated in transit to the hospital. His chest x-ray and chest computed tomography scan revealed bilateral pneumothoraces with

From the Infectious Disease Division, Winthrop-University Hospital, Mineola, New York, and State University of New York School of Medicine, Stony Brook, New York.

Corresponding author: Burke A. Cunha, MD, Infectious Disease Division, Winthrop-University Hospital, Mineola, NY 11501.

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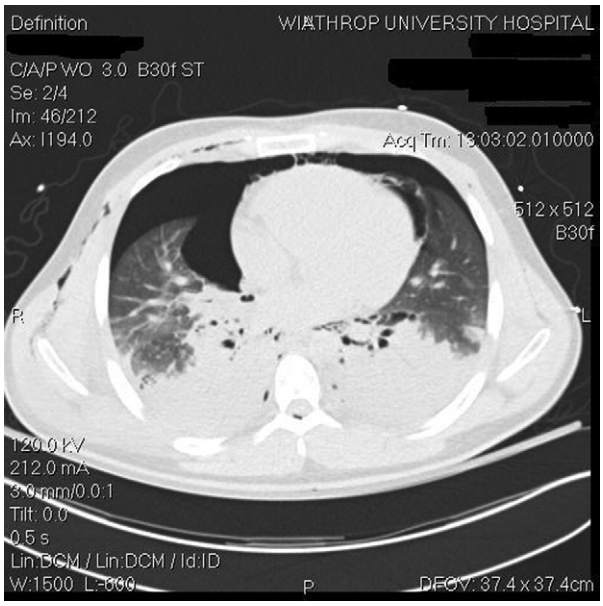

Fig 1 Chest computed tomography scan with bilateral pneumothoraces and bibasilar patchy infiltrates in a patient with *Legionella* CAP.

patchy bibasilar infiltrates (Fig 1). Chest tubes were placed bilaterally, and the patient was transferred to the intensive care unit. On admission, he was febrile (101°F) and beginning to respond to verbal stimuli. His family denied any history of trauma or drug abuse. His physical examination was unremarkable except for chest findings associated with his bilateral pneumothoraces. Admission laboratory studies revealed a hypophosphatemia of 2 mg/dL (n = 2.7-4.7 mg/dL), an elevated creatine phosphokinase of 23,485 IU/L (n = 47-422 IU/L), and elevated liver enzymes, that is, a serum glutamate oxaloacetate transaminase of 282 IU/L (n = 13-39 IU/L), a serum glutamate pyruvate transaminase of 180 IU/L (n = 4-36 IU/L), and an alkaline phosphatase of 155 IU/L (n = 25-100 IU/L). Doxycycline 200 mg was administered intravenously every 12 hours. The patient's condition improved during the next few days; he became afebrile and was extubated. Microbiological test results for typical and atypical CAP pathogens were negative, but his urine Legionella antigen test was positive. He completed 3 weeks of doxycycline therapy and had an uneventful recovery.

DISCUSSION

Pneumothorax is an accumulation of air in the pleural space and can be spontaneous or caused by a complication of trauma or medical procedures.[8,9] Spontaneous pneumothorax mostly occurs in patients with underlying lung pathology. The most common noninfectious causes of spontaneous pneumothorax are related to lung diseases (eg, emphysema). Other less common pulmonary diseases include interstitial lung disease, connective tissue diseases (Marfan's/Ehlers-Danlos syndrome), malignancy, histiocytosis X (Langerhan's cell/eosinophilic granuloma), sarcoidosis, lymphangioleiomyomatosis, and endometriosis.[8,9] The most common infectious causes of spontaneous pneumothorax are tuberculosis, necrotizing pneumonia, *Staphylococcus aureus* presenting with pneumatoceles (children), lung abscesses, *Pneumocystis (carinii) jiroveci* pneumonia, and severe acute respiratory syndrome. Septic pulmonary emboli (secondary to right-sided endocarditis) has also been reported to cause spontaneous unilateral pneumothorax. Spontaneous bilateral pneumothoraces are rare, but there has been a case report in a patient with rheumatoid lung (on steroids) infected with *Aspergillus sp*.

Unilateral spontaneous pneumothorax is a rare presenting feature of *Legionella* CAP. If there is no history or clinical findings of an underlying connective tissue disorders or lung disease, an infectious cause should be considered (Table 1).

The radiologic manifestations of Legionnaires' disease are nonspecific, ranging from interstitial infiltrates to infiltrates with consolidation, and uncommonly cavitation or pleural effusions. Although there are no pathognomonic findings of *Legionella* CAP, infiltrates characteristically are basilar and rapidly progressive.[4-7] Typically, the infiltrates of Legionnaires' CAP progress despite adequate antibiotic therapy (Table 2). This case of spontaneous bilateral pneumothoraces as the presenting manifestation of *Legionella* CAP is unique. *Legionella* CAP was suspected on the basis of nonspecific but characteristic extrapulmonary laboratory findings and the known association of pneumothorax with *Legionella*, which prompted specific diagnostic testing for *Legionella*.[17-20]

Because the patient's CPR did not result in multiple rib fractures, his pneumothoraces should not be attributed to CPR. In cases of CPR with rib fractures, pneumothorax, if present at all, is unilateral and not bilateral. Therefore, we believe this to be the first reported case of spontaneous bilateral pneumothoraces caused by *Legionella* CAP. Clinicians should be aware that spontaneous pneumothorax is a rare presenting feature of *Legionella* CAP and may be unilateral or bilateral.

Table I
Differential diagnosis of spontaneous pneumothorax

Infectious causes	Noninfectious causes
Common	Common
• TB	• Emphysema
• PCP (HIV)	• Congenital blebs
• *S. aureus* pneumatoceles (children)	• Asthma (status asthmaticus)
	• Histocytosis X (Langerhan's cell/eosinophilic granuloma)
	• Osteogenic sarcoma (metastatic to lungs)
Uncommon	Uncommon
• SARS	• Sarcoidosis
	• Interstitial fibrosis
	• Cystic fibrosis
Rare	Rare
• Septic pulmonary emboli	• Endometriosis
• Legionnaires' disease	• Lymphangioleiomiomatosis
	• Marfan's syndrome
	• Ehlers-Danlos syndrome

TB, Tuberculosis; *PCP, Pneumocystic (carinii) jiroveci* pneumonia; *HIV,* human immunodeficiency virus; *SARS,* severe acute respiratory syndrome

Table II
Radiologic manifestations of Legionaires' disease

Common	Uncommon
• Unilateral patchy lower lobe infiltrates	• Wedge-shaped opacities
• Bibasilar patchy lower lobe infiltrates	• Cavitation (more common in immunocompromised hosts)
• Infiltrates with consolidation	• Abscesses
• Rapidly progressive asymmetric infiltrates (on appropriate anti- Legionella antibiotic therapy)	• Unilateral pneumothorax
• Pleural effusion (frequency increases as disease progresses)	

REFERENCES

1. Fraser DW, Tsai TR, Orenstein W, et al. Legionnaires' disease: description of an epidemic of pneumonia. N Engl J Med 1977;297:1189-97.
2. Stout JE, Yu VL. Legionellosis. N Engl J Med 1997;4;337:682-7.
3. Cunha BA. The atypical pneumonias: clinical diagnosis and importance. Clin Microbiol Infect 2006;12(Suppl 3):12-24.
4. Tan MJ, Tan JS, Hamor RH, File TM Jr, Breiman RF. The radiologic manifestations of Legionnaire's disease. The Ohio Community-Based Pneumonia Incidence Study Group. Chest 2000;117:398-403.
5. Muder RR, Yu VL, Parry MF. The radiologic manifestations of Legionella pneumonia. Semin Respir Infect 1987;2:242-54.
6. Falco V, Fernandez de Sevilla T, Alegre J, et al. Legionella pneumophila. A cause of severe community-acquired pneumonia. Chest 1991;100:1007-11.
7. Sakai F, Tokuda H, Goto H, et al. Computed tomographic features of Legionella pneumophila pneumonia in 38 cases. J Comput Assist Tomogr 2007;31:125-31.
8. Gill MV, Cunha BA. Spontaneous pneumothorax. Infect Dis Pract 1994;18:101.
9. Sahn SA, Heffner JE. Spontaneous pneumothorax. N Engl J Med 2000;342:868-74.
10. Martinez-Vazquez C, Seijas M, Ocampo A, et al. Pneumothorax in patients infected by the human immunodeficiency virus. Ann Med Intern 2001;18:521-4.
11. Mezghani S, Abdelghani A, Njima H, et al. Tuberculous pneumothorax. Retrospective study of 23 cases in Tunisia. Rev Pneumol Clin 2006;62:13-8.
12. Sihoe AD, Wong RH, Lee AT, et al. Severe acute respiratory syndrome complicated by spontaneous pneumothorax. Chest 2004;125:2345-51.
13. Sheu CC, Hwang JJ, Tsai JR, Wang TH, Chong IW, Huang MS. Spontaneous pneumothorax as a complication of septic pul-

monary embolism in an intravenous drug user: a case report. Kaohsiung J Med Sci 2006;22:89-93.
14. Winne L, Praet M, Brusselle G, Veys E, Mielants H. Bilateral spontaneous pneumothorax in a patient with pulmonary rheumatoid nodules, secondary infected by Aspergillus. Clin Rheumatol 2007;26:1180-2.
15. Bali A, Pierry AA, Bernstein A. Spontaneous pneumothorax complicating Legionnaires' disease. Postgrad Med J 1981;57:656-7.
16. Sundkvist T, Carlsson MG. Legionnaires' disease: unusual presentation with pneumothorax. Scand J Infect Dis 1983;15:127-8.
17. Cunha BA. Clinical diagnosis of Legionnaire's disease. Semin Respir Infect 1998;13:116-27.
18. Cunha BA. Diagnostic significance of non-specific laboratory tests in infectious diseases. In: Gorbach SL, Bartlett JB, Blacklow NR, eds. Infectious diseases in medicine and surgery, 3rd edition. Philadelphia: WB Saunders Co; 2002.
19. Cunha BA. Hypophosphatemia: diagnostic significance in Legionnaires' disease. Am J Med 2006;119:e5-e6.
20. Cunha BA. Pneumonia essentials, 2nd edition. Royal Oak, MI: Physicians Press; 2008.