CASE REPORT

Spontaneous rupture of the spleen associated with Legionella pneumonia

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Spontaneous rupture of the spleen associated with *Legionella* pneumonia is a rare and life-threatening complication; only three cases have been reported to date. The authors describe a case of a 47-year-old man who presented with pneumonia and abdominal pain. He underwent a splenectomy, and was successfully treated with clarithromycin and levofloxacin.

Key Words: Legionella pneumophila; Pneumonia; Splenic rupture

Spontaneous nontraumatic rupture of the spleen is extremely rare, and a potentially life-threatening complication of infectious or neoplastic diseases (1). Although the multisystem involvement of Legionnaires' disease has been well documented, pneumonia continues to be the most common and important manifestation of infection with *Legionella* (2). Pneumonia has only occasionally been associated with spontaneous rupture of the spleen, and only a few cases are due to *Legionella* (3-5). In 1996, Domingo et al (5) offered a broad review on the topic, covering *Legionella* and other pathogens.

We report a case of a patient who presented to the Hospital of La Linea (Cadiz, Spain) with spontaneous rupture of the spleen and pneumonia caused by *Legionella pneumophila*. Cases of *Legionella*-associated spontaneous rupture of the spleen are also reviewed.

CASE PRESENTATION

A 47-year-old man with no apparent general disease presented to the emergency department with a 10 h history of dry cough, dyspnea and high fever. He also suffered from abdominal pain, which he described as being vague periumbilical pain. It was not radiated, but worsened with inspiration. He did not experience any previous chest or abdominal trauma.

The patient's medical history was unremarkable. He did not take any regular medications and had no known drug allergies. He did not report smoking or alcohol use. He did not have any recent repair of domestic plumbing; he denied any recent travel or visits to any whirlpool bath or spa facilities; had not participated in any unusual hobbies; had not come in contact with any sick people and had no risk factors for HIV infection.

The findings from his physical examination on admission were as follows: body temperature 39.1°C; blood pressure 130/80 mmHg; pulse rate 123 beats/min; and irregular, respiratory rate 30 breaths/min. He was anemic, and his chest auscultation revealed hypoventilation on the right upper lung field.

The patient's laboratory findings on admission were as follows: hemoglobin level of $8.5 \, g/L$; hematocrit level of 0.25; white blood cell count of $12,900\times10^9/L$, with 89.9% neutrophils, 4.4%

Une rupture spontanée de la rate associée à une pneumonie à Legionella

La rupture spontanée de la rate associée à une pneumonie à *Legionella* est une complication rare qui met la vie en danger. Seulement trois cas ont été déclarés jusqu'à présent. Les auteurs décrivent le cas d'un homme de 47 ans qui a consulté à cause d'une pneumonie et de maux de ventre. Il a subi une splénectomie et a été traité avec succès au moyen de clarithromycine et de lévofloxacine.

lymphocytes and 5.5% monocytes; and a platelet count of $150,000\times10^9/L$. His serum sodium level was 128.6 mmol/L; urea concentration was 28.9 mmol/L; creatinine level was 210.39 µmol/L and albumin level was 21 g/L. His C-reactive protein level was 162 mg/L. Arterial blood gas analysis on room air showed a PO_2 of 58 mmHg, PCO_2 of 24 mmHg, HCO_3^- of 17.9 mmol/L, base excess of -4.1 mmol/L, oxygen saturation of 92% and pH of 7.5. Blood glucose, total bilirubin, aspartate aminotransaminase, alanine aminotransaminase and creatine kinase levels were normal. His lactic dehydrogenase level was 186 U/L. The coagulation values were also normal.

On admission, the patient's chest x-ray showed a homogeneous infiltrate of the right upper lobe diagnosed as pneumonia. Chest computed tomography also showed alveolar condensation of the right upper lobe. Abdominal computed tomography revealed splenic intraparenchymal laceration with a subcapsular hematoma and moderate free intra-abdominal pool of blood. The patient was transferred for surgery.

Emergency laparotomy showed 1400 mL of blood and rupture of the spleen with a subcapsular hematoma. The spleen was removed without complication and was sent for pathological investigation. It was enlarged and weighed 295 g. Histological examination revealed a normal white pulpa, but a hyperplastic red pulpa was markedly congested and had an acute inflammatory reaction (splenitis).

An ordinary bacteriological culture of a respiratory specimen failed to yield any significant pathogens for the pneumonia. Spleen cultures for bacteria, mycobacteria and fungi were negative, as were blood and urine cultures. A urine test for urinary antigen to pneumococci (NOW Streptococcus pneumoniae, Binax Inc, USA) was negative. A urinary antigen test for *L pneumophila* serogroup 1 (Binax Legionella Urinary Antigen EIA, Binax Inc, USA) with concentrated urine on admission was positive. The serum level of indirect fluorescent antibody titre to *L pneumophila* serogroup 1 was negative on admission but showed a titre of 1:256 three weeks later. The serological tests for infection with Epstein-Barr virus, cytomegalovirus, HIV, mycoplasma, Coxiella burnetii and Chlamydia pneumoniae were all negative.

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TABLE 1
Clinical characteristics of patients with spontaneous rupture of the spleen associated with *Legionella* pneumonia

Interval								
Year (reference)	,	Underlying conditions	Location of pneumonia	between initial		Complications before spleen rupture	Spleen pathology	Outcome
				pneumonia and rupture, days				
					Renal failure			
1993 (4)	42/M	Moderate tobacco and alcohol use	Left basal	4	Cefotaxime + erythromycin	None	Enlarged	Survived
1996 (5)	52/M	None	Left lower lobe	5	Cefotaxime + erythromycin	None	Enlarged, splenitis	Survived
2008 (present case)	47/M	None	Right upper lobe	0	Clarithromycin + levofloxacin	None	Enlarged, splenitis	Survived

The patient was transfused with three units of packed red blood cells in the immediate postoperative period. He was treated with intravenous clarithromycin 500 mg every 12 h and intravenous levofloxacin 500 mg every 24 h for 14 days. The recovery was uneventful, and the patient was discharged in good health 17 days later.

DISCUSSION

In the MEDLINE literature search, three cases of spontaneous rupture of the spleen associated with pneumonia caused by *L pneumophila* were found. The clinical characteristics of these patients and our patient are summarized in Table 1.

In previous cases, spontaneous rupture of the spleen occurred at a mean of 6.6 days (range four to 11 days) after the beginning of pneumonia symptoms. However, in our patient, the rupture of the spleen was present at admission, although he had only a 10 h history. One patient died; this death was directly related to hemorrhagic shock (3). Complications that could have contributed to the spontaneous rupture of the spleen were liver and renal failure. Interestingly, three of the cases did not have any underlying conditions, and only one patient (4) was a moderate smoker and drinker. Legionnaires' disease, even in severe cases, may occur in previously healthy subjects without predisposing factors (6).

Legionella is a cause of both community and nosocomial pneumonia. A delay in starting appropriate therapy for Legionella pneumonia significantly increases the mortality (7). However, pneumonia caused by L pneumophila cannot be differentiated from other types of pneumonia by clinical, radiographic or laboratory findings (6,8). In our patient, we found the etiological agent due to the early recognition of Legionella pneumonia using urine antigen testing; we thus started early therapy, which may have contributed to the favourable clinical response.

The mechanism of spontaneous rupture of the spleen associated with legionellosis remains uncertain, but probably results from a combination of factors, particularly splenic distension, infection and thrombosis (1,5,9). A sudden increase in portal pressure produced by coughing, vomiting or defection may precipitate splenic congestion and rupture.

Spontaneous rupture of the spleen should be suspected in patients admitted with *Legionella* pneumonia who suddenly develop clinical symptoms consistent with hypovolemic shock and failing hemoglobin levels. Definitive diagnosis requires investigations by ultrasonography, computed tomography and/or radionuclide scanning.

Treatment of spontaneous rupture of the spleen associated with Legionella pneumonia should be laparotomy with splenectomy, which in most cases should be undertaken without delay. Our patient was treated with intravenous clarithromycin and levo-floxacin. In moderate cases, intravenous fluoroquinolone monotherapy or a new injectable macrolide should be given (10,11). In mild cases, monotherapy with oral fluoroquinolones, azithromycin or telithromycin (12) may be adequate.

CONCLUSION

Spontaneous rupture of the spleen is a rare and life-threatening complication that should be borne in mind in patients with *Legionella* pneumonia who present with abdominal pain and low or failing hemoglobin levels. When properly diagnosed and treated, the outcome of the patients is favourable.

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