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Pulmonary inflammatory pseudotumor due to *Coxiella burnetii*. Case report and literature review

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Abstract

A 58-year-old man was admitted because of respiratory failure, episodic fever with chilling, cough, malaise, fatigue, myalgia and weight loss lasting for at least one month. Chest x-rays and CT scan of the chest showed bilateral pulmonary consolidations in upper lobes, the left lower lobe, and mediastinal lymphadenopathy. Bronchoscopy with cytology was unremarkable. A needle CT-guided lung biopsy documented an inflammatory pseudotumor, lymphoplasmacytic type. Serology showed high titer antibodies to phase II *Coxiella burnetii* infection. Therapy with doxycycline and hydroxychloroquine for three months led to a complete resolution of symptoms and radiological findings, and a marked decrease in titers to Q fever.

Keywords: Pulmonary pseudotumor; Infection; *Coxiella burnetii*

1. Introduction

Q fever is a zoonotic disease caused by *Coxiella burnetii* that may have both acute and chronic manifestations [1] and [2]. The incubation period can last up to 2–4 weeks after exposure, and the infection is known for its self-limiting character [2]. Indeed, following exposure to *C. burnetii,* a non-immune person develops a primary infection that is asymptomatic in 60% of cases [3] and [4], and most patients spontaneously recover [5]. In humans, Q fever is usually acquired by inhalation of aerosols contaminated with *C. burnetii* [1]. In the lung, alveolar macrophages and other mononuclear phagocytes seem to be the primary target cells of this obligate intracellular Gram-negative bacterium [6]. Symptomatic infection seems more likely in adult men [7]. Overt acute Q fever infection presents with myalgia, headache, anorexia, asthenia and a high fever [1] and [3]. Hepatitis, with limited or no symptoms, may also occur [1] and [3]. Chronic Q fever, instead, refers to the persistence of the pathogen after primary infection, both after symptomatic and asymptomatic primary infection [8] and [9]. If one month post acute Q fever the serum PCR is still positive, it is already possible to diagnose chronic Q fever [10]. Endocarditis is a frequent hallmark of the chronic form of the disease [1]. Furthermore, also vascular infections (both infected aneurysms and vascular prosthesis) occur frequently [8]. Pulmonary involvement is possible in both the acute and chronic forms [1]. In most patients with Q fever pneumonia, chest X-rays usually reveal nonspecific infiltrates [11] and [12]. Inflammatory pseudotumor of the lung during the course of *C. burnetii* pneumonia, instead, is a rarely reported finding in literature [1] and [2]. Janigan and Marrie have described the first case in 1983 [13], and Lipton and Fong studied the second case in 1987 [14]. Both cases were observed in the acute phase of the disease. We are now

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reporting a third patient with pulmonary inflammatory pseudotumor due to *C. burnetii*, observed in the chronic phase of the disease, that resolved completely after treatment with doxycycline and hydroxychloroquine.

2. Case report

A 58-year-old man, construction worker, with history of smoking and prolonged exposure to sheep and goat herds, was admitted to our Pneumology Clinic for respiratory failure, chills, episodic profuse sweating, cough, malaise, joint pain, anorexia, weight loss and a high fever, one week after surgery for complicated epigastric hernia. In one-month period before admission, the occurrence of unexplained recurrent febrile episodes was noted. Examination showed that temperature was 38.8 °C, respiration rate 20, O_2 saturation 92% (room air), blood pressure 130/80 mm Hg, and pulse rate 80. Decreased breath sounds, rales, and decreased tactile and vocal fremitus were noted over both the upper lobes and the left lower lobe. Moreover, at precordial examination, a mid-systolic 2/6 murmur, indicative of mitral regurgitation, was heard on the mitral area. Transthoracic echocardiogram showed no significant heart valve disease. The patient refused to perform transesophageal echocardiography. Chest-x–ray films and Computed Tomography scan of the chest revealed multiple, solid masses with irregular margins and air bronchograms, involving both the upper lobes and the left lower lobe, without evidence of cavitation. A mediastinal lymphadenopathy, above and below the carina, was also evident [Fig. 1]. Bronchoscopy with cytology was unremarkable.

Hemoglobin level was 7.8 g per 100 ml, with reduced serum iron, the white-cell count was 11.00/mm^3, with 78.7% polymorphonuclear leukocytes, 8.3% monocytes, and 10.8% lymphocytes. The erythrocyte sedimentation rate was 120 mm/h, and platelet count 324.000. Liver function tests results were normal.

Cultures of sputum and bronchial aspirate, the Quantiferon test in whole blood, the search for pneumococcus and Legionella urinary antigen tests, and serological studies for pneumotropic infective agents, including Chlamydia and Mycoplasma, were negative. Bronchoalveolar lavage fluid showed increased number of neutrophils (95%). Treatment for possible bacterial infection with parenteral amikacin, one gram daily, and piperacillin/tazobactam, 4.5 g, three times daily, for 10 days, was instituted; however, no improvement occurred. Four days after completion of antibiotic treatment, a needle Computed Tomography-guided lung biopsy was performed to establish a definite diagnosis.

Histological preparations showed replacement of the pulmonary parenchyma by numerous plasmacells, histiocytes and

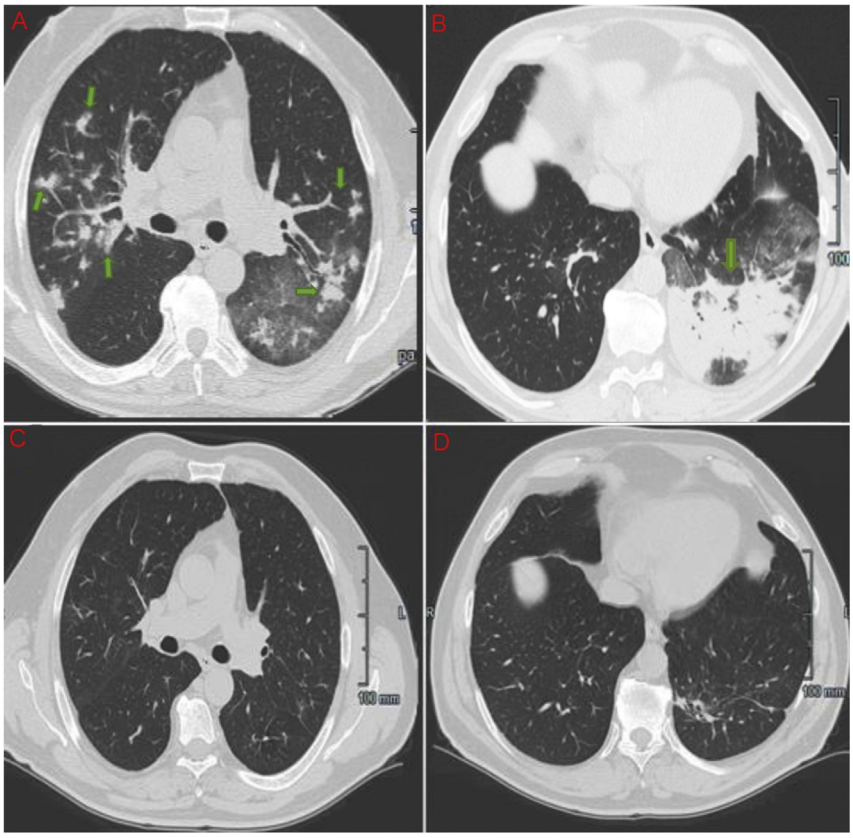

Fig. 1. Computed Tomography Scan of the lungs. Multiple, solid masses with irregular margins, mediastinal lymphadenopathy, air bronchograms and ground-glass opacity, involving the upper lobes and the left lower lobe are evident. (A, B) (arrows) Follow-up Computed Tomography scan shows complete resolution of the lesions after three months of doxycycline treatment. (C, D).