

Eosinophilic Pleural Effusion as a Manifestation of Idiopathic Hypereosinophilic Syndrome[☆]



Derrame pleural eosinofílico secundario a síndrome hipereosinofílico idiopático

To the Editor,

Eosinophilic pleural effusion (EPE) is pleural effusion (PE) in which eosinophils account for more than 10% of pleural fluid (PF) cellularity. In published series on PE, about 7.5% of the population show EPE.^{1,2} The most common overall cause of EPE is air or blood in the pleural space and the most common known etiology is malignant disease. Idiopathic hypereosinophilic syndrome (IHES) is a heterogeneous group of disorders consisting of hypereosinophilia, defined as an absolute eosinophil count $>1.5 \times 10^9/l$ or >1500 cells/ μl in 2 consecutive samples obtained 1 month apart, combined with eosinophil-induced organ damage, when other causes of hypereosinophilia have been excluded.

We report the case of a 25-year-old man with bilateral PE (Fig. 1). Complete blood count showed eosinophil concentrations of $3500/mm^3$, representing 26% of total white blood cells. He had shown similar results on previous tests. PF initially had a turbid appearance, with characteristics of exudate, glucose levels of 64 mg/dl and adenosine deaminase levels of 27.7 U/l. PF cytology showed no malignant cells and acutely predominant inflammatory cells, with abundant eosinophils (50%–60% of total cell count). PF culture was negative for bacteria, mycobacteria and fungi. Echocardiography showed neither pericardial effusion nor infiltrative cardiomyopathy. Tests were performed to rule out secondary causes of hypereosinophilia, all of which were negative, and after comprehensive hematological studies, the diagnosis was IHES. Response to prednisone 1 mg/kg was favorable. The eosinophilia abated initially in peripheral blood and subsequently, after 7 months, in PE. Eosinophil levels in normal PF are low (less than 1%). High levels can be due to multiple causes. The most common causes of EPE are malignant (around 30%–34%),^{1,2} parapneumonic, and tuberculous effusion. Other possible etiologies include asbestos, drug toxicities, parasitological infections, and Churg-Strauss syndrome, or less commonly, viral infections and pulmonary embolism.³ Very little information is available on EPE due to IHES or causes other than those mentioned, as can be surmised from the few cases reported to date.⁴ The particular interest in this patient is that the disease started only as relapsing EPE. In one of the published cases, the patient had hepatosplenomegaly and ascites combined with bilateral PE, and unlike our patient, the

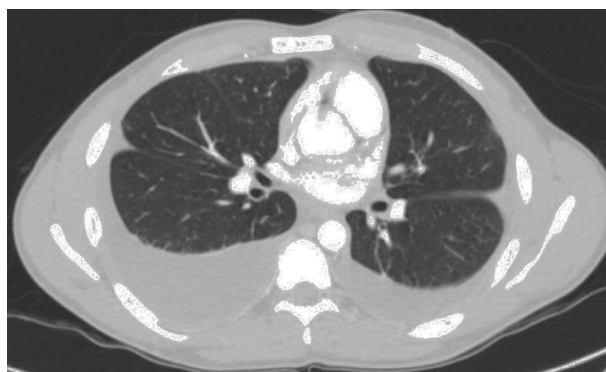


Fig. 1. Chest computed tomography showing bilateral pleural effusion with lack of pulmonary involvement.

outcome was death. In addition to bilateral EPE, the other patient presented skin lesions which were found to be due to vasculitis.⁵

When EPE with no apparent etiology is observed, IHES must be considered. Eosinophilia in peripheral blood in the absence of other manifestations should alert to the presence of this entity. Close collaboration with the hematologist is necessary to confirm the etiology of this finding.

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Lady Windermere Syndrome: Does It Occur Only in Women?[☆]



Síndrome de Lady Windermere. ¿Una entidad exclusiva de mujeres?

To the Editor,

Recent years have seen an increase in developed countries in the incidence of infections caused by environmental

mycobacteria, particularly *Mycobacterium avium* complex (MAC) infection in immunocompetent patients; the primary radiological signs of this entity are fibrocavitary disease or nodules and bronchiectasis, predominantly in the middle lobe and lingula.¹ This manifestation, known as Lady Windermere syndrome (LWS), has been described mostly in women over 50 years of age, while episodes in men are anecdotal.²

We report the case of a 64-year-old man, health worker, with no significant personal history or known toxic habits, who reported a 3-year history of non-productive cough, with no particular diurnal pattern, dysphonia and excess mucosity in the oropharyngeal cavity, and additionally, in the last year, asthenia and recurrent episodes of low-grade fever. Forced spirometry results were as follows:

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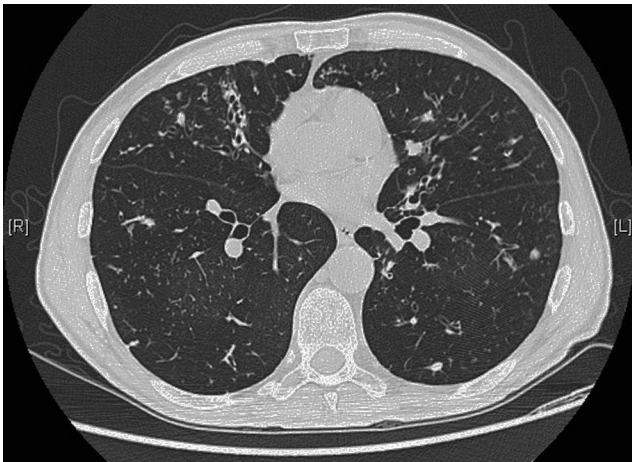


Fig. 1. Bronchiectasis in the middle lobe and lingula associated with mucus plugs and distal centrilobular nodes.

FEV1/FVC 0.46; FEV1 1.91 L (60%); FVC 4.14 L (101%). Bronchodilator challenge and skin prick tests with allergens were negative. Computed tomography of paranasal sinuses, sweat chloride test, IgG, IgM, IgA, IgE and alpha-1 antitrypsin levels were all within normal ranges. A human immunodeficiency virus test was negative. A barium swallow study of gastroesophageal transit revealed gastric reflux. Chest CT showed bronchiectasis in the middle lobe and lingula associated with mucus plugs and distal centrilobular nodes (Fig. 1). Macroscopic results from fiberoptic bronchoscopy sampling were normal, while *M. avium complex* was isolated from bronchoalveolar lavage. The patient met diagnostic criteria for environmental mycobacteria published by the American Thoracic Society (2007)³ and was diagnosed as having MAC infection consistent with LWS.

In immunocompetent patients, MAC infection can present in 2 ways: (1) fibrocavitary lesions in the upper lobes, often observed in middle-aged men and often associated with a history of smoking and/or alcohol abuse and previous lung disease and (2) nodular lesions less than 5 mm in size generally associated with bronchiectasis. These lesions occur predominantly in the middle lobe and

lingula and are more common in women. This entity is known as LWS.⁴ This syndrome was first described in 1992 by Reich and Johnson in a series of 6 older, immunocompetent women with no significant history of smoking or previous respiratory disease who developed MAC infection. They postulated that voluntary suppression of the cough reflex predisposed to the syndrome by preventing secretion drainage. Moreover, other factors, such as leptin or adiponectin levels and menopause⁵ might also play a role in the pathogenesis of this infection. Our patient had chronic cough associated with gastroesophageal reflux, which he tried to suppress during his work that involved contact with the public, and this may have promoted the development of MAC infection. Despite the patient being a man, the clinical, microbiological and radiological criteria led to a diagnosis of LWS.

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