Case Report

Functional and Radiological Characteristics in Chronic Untreated Tropical Pulmonary Eosinophilia

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Abstract

Long standing untreated tropical pulmonary eosinophilia (TPE) generally results in interstitial lung disease (ILD), as underlying chronic inflammation heals by fibrosis. The presentation of TPE as ILD is a rare entity. A patient was evaluated for persistent cough over two years. High resolution computed tomography of the chest was suggestive for ILD in this case. The patient was diagnosed, however, with TPE based on persistent high eosinophil count, positive antifilarial antibody and dramatic response to diethylcarbamazepine. Follow-up HRCT of the chest after 16 months of treatment showed complete resolution of ground-glass opacities. A significant improvement in spirometry was also noted. **[Indian J Chest Dis Allied Sci 2017;59:43-45]**

Key words: Tropical pulmonary eosinophilia, Interstitial lung disease, Fibrosis.

Introduction

Tropical pulmonary eosinophilia (TPE) is a hypersensitivity reaction following infestation by parasite *Wuchereria bancrofti* and *Brugia malayi*. Untreated TPE has diverse symptomatology, radiological and spirometric findings depending on the duration of the disease process.[1] In view of the persistent eosinophilia; TPE was suspected in our patient who presented as a case of pulmonary fibrosis.

Case Report

A 73-year-old male, a retired businessman, resident of Mumbai, with no history of addictions presented with symptoms of dry cough, mainly in the day time since 2 years. The patient did not have any history of atopy, gastro-esophageal reflux disease (GERD) or any co-morbid diseases. On examination, the patient was hemodynamically stable with an oxygen saturation of 98%, with post-exercise desaturation up to 93%. There was no clubbing, cervical lymphadenopathy, pallor, pedal oedema. The jugular venous pressure (JVP) was normal. On auscultation, patient had end-inspiratory fine crackles in bilateral infra-axillary and infra-scapular regions. Chest radiograph (postero-anterior view) showed bilateral lower zone reticulo-nodular opacities (Figure 1). Complete blood count (CBC) was done 8 months prior showed haemoglobin of 13.8 g/dL, total white blood cell count of 14060/mm^3, eosinophil of 38% with absolute eosinophil count (AEC) of 5413/mm^3. Serum immunoglobulin-E (IgE) was 11,658 IU/mL.

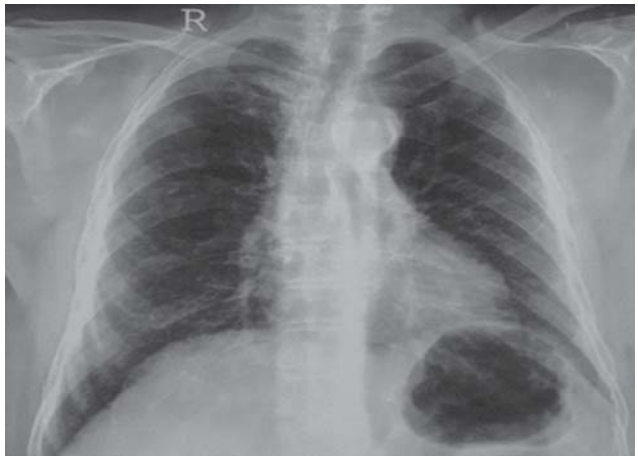

Figure 1. Chest radiograph (postero-anterior view) showing few bilateral lower zone reticulo-nodular opacities.

High resolution computed tomography (HRCT) of thorax done 6 months earlier showed bilateral sub-pleural, interlobular and intralobular interstitial thickening along with patchy ground-glass opacities in lower lobes (Figure 2). Previous chest radiographs done at one and half years back showed bilateral lower lobe opacities with mottled appearance (Figure 3), and CBC revealed haemoglobin was 14 g/dL, total white blood cell count was 20040/mm^3, an eosinophil count of 63% with AEC of 12600/mm^3. The patient also gave history of breathlessness and wheezing associated with bouts of cough 2 years earlier, that subsided within 2-3 months. The patient was treated earlier with inhaled bronchodilators, inhaled and short course oral steroids repeatedly with no complete relief of the symptoms.

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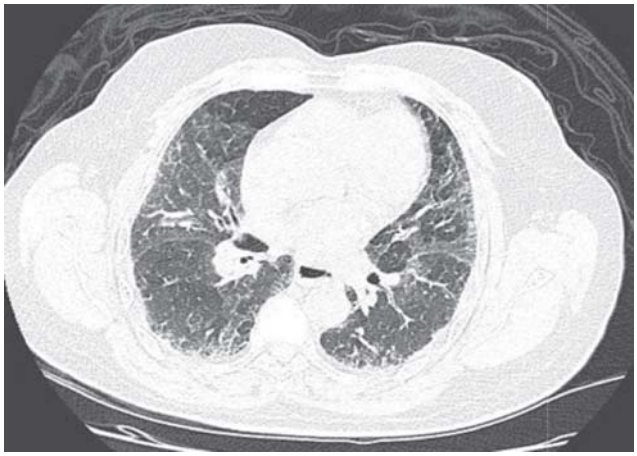

Figure 2. High-resolution computed tomography of thorax showing subpleural, interlobular and intralobular thickening with patchy ground-glass appearance.

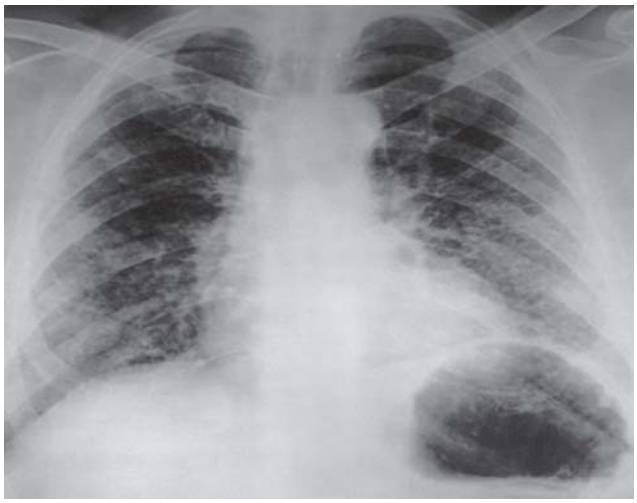

Figure 3. Chest radiograph (postero-anterior view) at the onset of symptoms showing bilateral lower zone mottled appearance.

At our institute, patient was evaluated with complete blood count (CBC) and HRCT thorax. The AEC was 5413/mm^3, serum IgE was 17,960 IU/mL, and antifilarial antibody IgG was positive. The repeat HRCT of the thorax showed no further increase of interstitial and ground-glass opacities. Spirometry showed mild restrictive defect with forced vital capacity of 2.07 litres (70.4%).

The patient was provisionally diagnosed to have tropical pulmonary eosinophilia (TPE) based on high eosinophilic count, high serum IgE levels and positive antifilarial antibody. The patient was started on oral diethylcarbamazepine (DEC) (100 mg three times a day) for 21 days along with oral prednisolone (0.6 mg/kg once a day) for 7 days. After completion of the therapy of 21 days with DEC, cough subsided completely and AEC value was 117/mm^3. However, chest radiograph showed persistent reticulonodular opacities in bilateral lower zones. On further follow-up after 6 months, the patient was asymptomatic with normal AEC. There was no further deterioration in the chest radiograph. On subsequent follow-up after 16 months, patient continued to be asymptomatic and spirometry showed an improvement in FVC by 420 mL (20.2%) compared to last spirometry at the time of diagnosis. Repeat HRCT showed near complete resolution of ground-glass opacities with residual subpleural, intralobular and interstial thickening predominantly in the lower lobes (Figure 4).

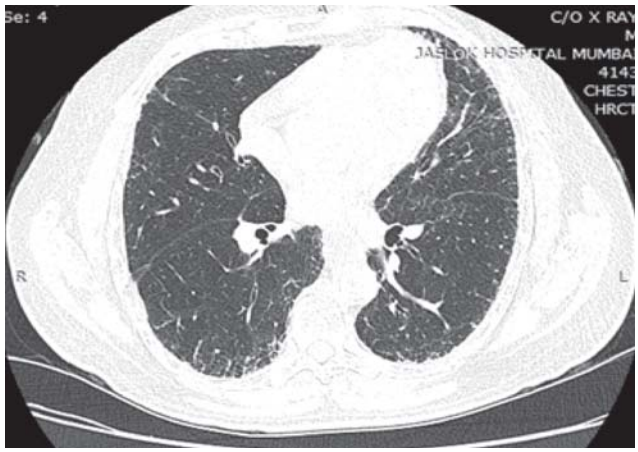

Figure 4. High-resolution computed tomography of thorax (16 months after treatment with diethylcarbamazepine) showing complete resolution of previous ground-glass opacity and residual subpleural interlobular and intralobular thickening in bilateral lower lobes.

The excellent response to DEC supported the diagnosis of TPE.

Discussion

Tropical pulmonary eosinophilia (TPE) is a manifestation of hypersensitivity reaction predominantly affecting the lungs following infestation with nematode *Wuchereria bancrofti* and *Brugia malayi*. However, less than 1% of patients infected with these parasites develop TPE.[1] TPE is endemic in areas where filariasis is endemic. It is most commonly found in regions of the Indian subcontinent, South-East Asia, South America and Africa.[2-4] In India, it is mostly found in the western coastal regions from Maharashtra to Kerala and eastern coastal regions from West Bengal to Tamil Nadu.[2] The prevalence of TPE in various settings in India has varied from 0.5% among children in Tamil Nadu to 9.9% among jail inmates in Patna.[1,5,6] Mature gravid filarial parasite living in lymphatic system releases microfilaria which gets trapped in pulmonary microcirculation. The human body triggers a graded immune response to these degenerating microfilariae, which is initially type I hypersensitivity reaction, followed by type 2 and type 4 reaction.[2,7,8] A major IgE inducing antigen (Bm2325) of the parasite *Brugia malayi*, have been observed in the patients with TPE and is found to

be the homolog of the enzyme, gamma-glutaryl transpeptidase light chain subunit.[9-11] The profound antibody response observed in TPE has been proposed to be due to molecular mimicry between the parasite gamma-glutaryl transpeptidase and the human gamma-glutaryl transpeptidase present on the surface of pulmonary epithelium.[10-12]

In untreated cases, histopathological studies have suggested different stages of inflammatory response as the duration of the disease increases. Symptomatology and spirometry corelates well to these different histopathological stages.[1,13,14]

During early stage of the disease (first 2 weeks of illness), there is histiocytic inflammation in the alveolar interstitial, peribronchial, perivascular spaces with preservation of lung architecture.[1,15] If untreated (one to three months), this is followed by eosinophilic infiltration with eosinophilic bronchopneumonia and micro abscesses in the lung. There is bronchial wall mucosal oedema and mucosal thickening due to eosinophilic infiltration.[1,15] During this period, cough, breathlessness, and wheeze are common symptoms. Symptoms are usually at night, but may occur in day time.[2] The respiratory symptoms may be accompanied with constitutional symptoms, like fever, weight loss, and malaise. At this stage, chest radiograph usually shows bilateral lower lobe mottled appearance or may be normal in 20% cases.[2] Spirometry shows obstructive pattern in 30% of cases, particularly within one month of illness.

Long standing untreated disease (6 months onwards) has chronic mixed cellular inflammation (histiocytes, epitheloid cells, lymphocytes and eosinophils) in a nodular pattern in the interstitium which heals by fibrosis leading to pulmonary fibrosis.[1,15] Spirometry shows restrictive defect with or without concomitant obstructive defect.[1] Even with treatment, chronic mild interstitial lung disease has been found to persist in these patients. However, in some patients, pulmonary fibrosis is not always progressive and the eosinophilia also progressively wanes spontaneously.[1]

Thus, it is important to understand time dependent changes in clinical, spirometric, radiological and pathological presentation of long-standing untreated TPE. A high index of suspicion is required so that this treatable disease entity is not missed. In addition, small dose of systemic steroids with DEC may also help untreated TPE with ground-glass opacities in preventing progression of pulmonary fibrosis.

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