

Case Report

Unusual presentation of chronic eosinophilic pneumonia with unilateral predominance and pleural effusion: a case report

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Abstract: Chronic eosinophilic pneumonia (CEP) is a rare disorder of unknown cause that is characterized by accumulation of eosinophils and lymphocytes in the pulmonary interstitial space and alveoli and can present subacute or chronic respiratory symptoms. It usually manifests as bilateral non-segmental peripheral air space consolidations with upper lobe dominance. We report a case of CEP in a young female patient showing atypical radiologic manifestations with unilateral predominance and pleural effusion. Because of the asymmetric upper lobe predominance and subacute to chronic symptoms appeared similar to those seen in pulmonary tuberculosis and lead to delayed treatment.

Keywords: Chronic disease, lung, pulmonary eosinophilia, computed tomography

Introduction

Chronic eosinophilic pneumonia (CEP) is a rare idiopathic and potentially life-threatening condition that manifests as subacute or chronic progressive respiratory symptoms, fever, and weight loss. In histological examination, it is characterized by accumulation of eosinophils and lymphocytes in the pulmonary interstitial space and alveoli [1]. The typical chest radiography and computed tomography (CT) findings of CEP are bilateral non-segmental peripheral air space consolidations with upper lobe predominance [2, 3]. However, a few atypical cases of idiopathic CEP have been reported with findings such as ground-glass opacity (GGO), nodules, reticulation, round consolidation with reversed halo sign [4], bronchial involvement [5]. To our knowledge, there is no report of histopathologically proven CEP with unilateral predominance except the unilateral case with iatrogenic cause in breast cancer patient [6]. Here, we report a case of pathologically confirmed CEP in a young female patient showing

atypical radiologic findings with unilateral predominance and pleural effusion.

Case report

A 25-year-old female visited our outpatient clinic presenting with cough, sputum, and weight loss (2 kg during 1 month) without fever during the previous six weeks. The symptoms were aggravated one week before the visit to our hospital. She has never smoked a cigarette and does not have any allergic history such as allergic dermatitis or asthma.

On initial chest radiography, consolidation with bronchial dilatation mainly in the left upper lung zone was noted (**Figure 1A**), and we suspected that the lesion was active pulmonary tuberculosis or pneumonia. On chest CT performed the same day, multifocal, non-segmentally distributed, peribronchial, and subpleural consolidations with bronchial dilatation and mild volume loss of left upper lobe were noted. However, no cavity or nodule was noted (**Figure 1B-D**). Another focus of subpleural GGO was noted in

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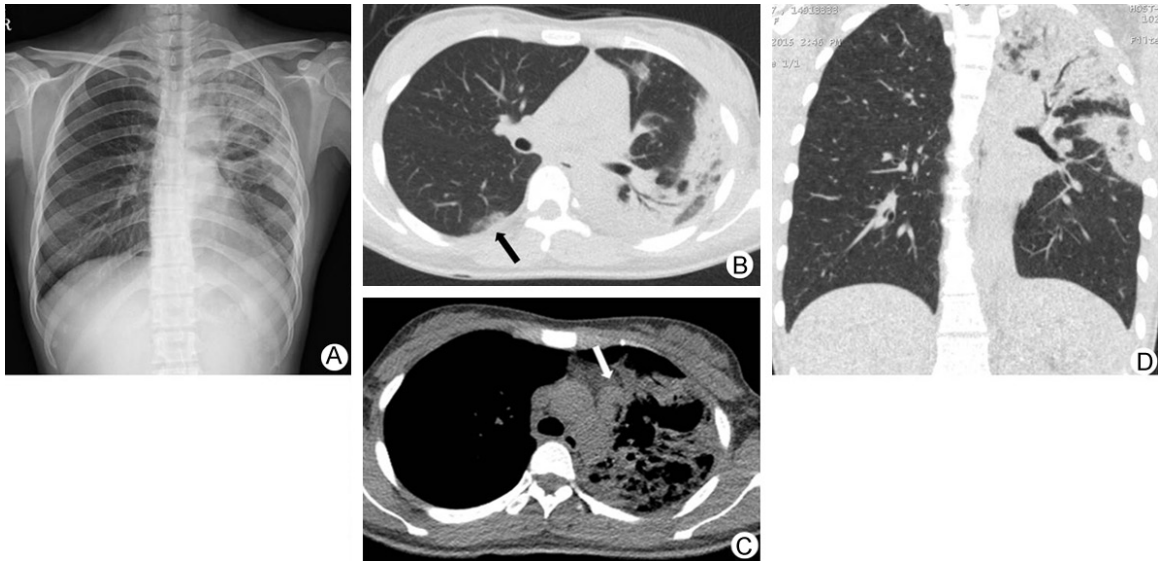


Figure 1. Chronic eosinophilic pneumonia in a 25-year-old female with sustained symptoms on conventional treatment. (A) Chest radiograph shows peribronchovascular patchy consolidations in the left upper lung zone with left costophrenic angle blunting. (B-D) Non-enhanced chest CT in axial and coronal images with lung (B, D) and mediastinal (C) settings show peribronchovascular and subpleural consolidations with bronchial dilatation in the left lung with volume loss. A small amount of pleural effusion in the left hemithorax and mediastinal lymphadenopathy (white arrow, C) also are noted. In addition, a focal foci of subpleural ground glass opacity lesion is also present at the right lower lobe (black arrow, B).

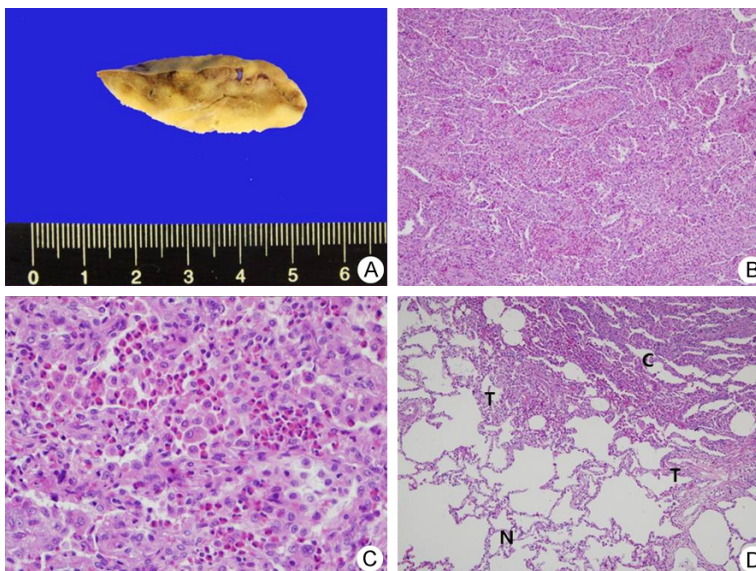


Figure 2. Chronic eosinophilic pneumonia in a 25-year-old female with sustained symptoms on conventional treatment. (A) Gross specimen obtained after left upper lobe wedge resection showing tan/brown tissue. (B, C) Light microscopy images showing dense eosinophilic infiltration of a lung parenchymal lesion with inflammatory changes and interstitial fibrosis (hematoxylin-eosin stain; original magnification 200 x B, 400 x C). (D) The transitional zone (T) between normal lung parenchyma (N) and consolidated lung (C) reveals dramatically obliterated air space by eosinophils with fibrosis (hematoxylin-eosin stain; original magnification 100 x).

the contralateral right lung (**Figure 1B**, black arrow). There was also unilateral pleural effu-

sion with mediastinal lymphadenopathy in the left hemithorax (**Figure 1C**, white arrow).

Thoracentesis performed for the pleural effusion showed exudate with normal adenosine deaminase (ADA) level (171 U/L) suggesting a reduced possibility of pulmonary tuberculosis infection. There was no other laboratory evidence of pulmonary tuberculosis, including a sputum acidfast bacilli (AFB) study. However, laboratory tests of blood showed mild leukocytosis (12,890/L), peripheral eosinophilia (34%), and elevated Ig E level (5,381 U/mL). These laboratory findings showed the possibility of eosinophilic lung disease; however, the imaging findings of unilateral predominance and ipsilateral pleural effusion without evidence of asthma

was not consistent with typical features of chronic eosinophilic lung disease.

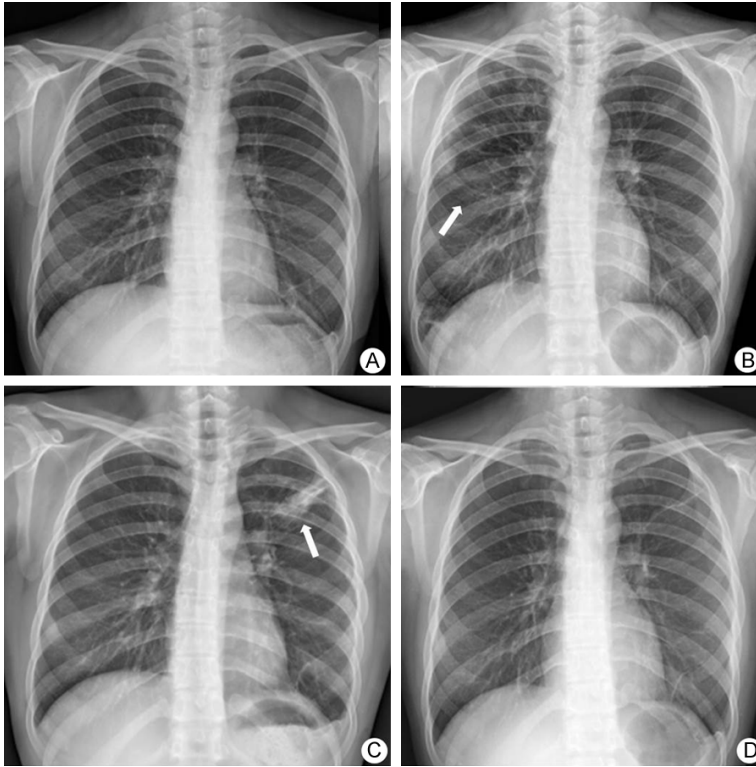


Figure 3. Chronic eosinophilic pneumonia in a 25-year-old female with sustained symptoms on conventional treatment. At the 2 months (A), 1 year (B), 2 year (C), 2 year and 2 weeks (D) follow-up after operation and steroid treatment, chest radiographs show waxing and waning patterns of the lesions (white arrows), but much improved compared with initial chest radiograph.

Therefore, we considered the lesion as atypical pneumonia such as mycoplasma or influenza as the first diagnosis rather than eosinophilic pneumonia and started treatment with clarithromycin combined with conventional antibiotics such as ceftriaxone. However, despite antibiotic medication for six days, the symptoms and laboratory findings were aggravated. Follow up chest radiography and CT also showed an increase in the extent of the unilateral lung lesions with fibrotic. Because of this atypical imaging finding and relatively rapid progressive fibrotic course in spite of conventional antibiotic therapy, we couldn't exclude the possibility of atypical pneumonia or pulmonary tuberculosis. However, iatrogenic pneumothorax was occurred during drain tube insertion and the patient underwent wedge resection of the left upper lobe by video-assisted thoracoscopic surgery (VATS) biopsy instead of bronchoalveolar lavage (BAL) for tissue confirmation. The final diagnosis was CEP showing dense eosinophilic infiltration with in-

flammatory change and fibrosis along alveoli and in interstitial spaces were detected and the transition between normal lung parenchyma to consolidation revealed obliteration of air space by inflammatory cells such as eosinophils, with dramatic fibrosis (Figure 2). After the operation, steroid therapy (methylprednisolone 125 mg IV) was started and both symptoms and radiological findings were dramatically improved (Figure 3A). At the 2-years follow-up, chest radiography showed waxing and waning lung lesions but much improved (Figure 3B-D).

Discussion

CEP is an idiopathic disorder that was first described in 1969 by CB Carrington. It usually occurs in middle-aged women and non-smokers with chronic symptoms such as chronic cough, sweat, fever, and weight loss [1]. On plain

chest radiography, CEP shows dense peripheral opacities parallel with pleura with non-segmental distribution, usually in apical or axillary levels. When the peripheral opacities extend to the basal lung, the finding is called "photographic negative pulmonary edema" [2]. On CT, the typical finding of CEP is inhomogeneous patchy consolidation, which presents a non-segmental, bilateral, peripheral distribution with upper lobe predominance [3, 7]. It can also show GGO, nodules, and streaky opacities on follow-up CT [8].

In our case, unilateral predominance of consolidation and progressive volume loss in a young female patient were noted. And unilateral pleural effusion with mediastinal lymphadenopathy were also noted. Because these image findings are not typical of CEP and mimic other infectious diseases like tuberculosis or atypical pneumonia, the proper diagnosis and treatment might be delayed. In addition, in case of patient not available with BAL,

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imaging and thoracentesis are only capable diagnostic tools, however, these atypical findings mimic other atypical infection make doctors hesitate to start steroids.

A few CEP cases that presented unilateral predominance have been reported. Gaensler et al. reported six cases that presented unilaterally distributed consolidation on plain chest radiography among 24 patients diagnosed with CEP; however, this study investigated only plain chest radiographs and some cases showed migratory consolidation from one side to the other, and volume loss was not shown in any case [2]. In contrast to these cases, our histopathologically proven case with CT showed prominent associated ipsilateral volume loss within the involved lung. In another case of unilaterally distributed consolidation in CEP with volume loss also was not confirmed histopathologically [9].

Moshimaru et al. reported the clinicopathological differences between acute eosinophilic pneumonia (AEP) and CEP [10]. They showed that fibrin deposition in alveoli accounted for the space-occupying consolidation, and the cytotoxicity of eosinophilic granules accumulated in lung parenchyma destroyed the basal lamina and subsequently led to intraluminal fibrosis [10]. We suggest that this process could explain the mediastinal shifting observed in our case and that of Andrew et al. [9]. The unilateral predominance of consolidation with progressive fibrosis in a CEP patient could account for volume loss of the affected lung and migration from the mediastinum to ipsilateral side.

In conclusion, although bilateral and non-segmental consolidation mainly in the upper lung zone is known to be a typical imaging finding of CEP, it can manifest as atypical imaging findings such as a unilateral predominant lesion with volume loss and accompanying ipsilateral pleural effusion with lymphadenopathy, mimicking pulmonary tuberculosis or atypical pneumonia.

Disclosure of conflict of interest

None.

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