Mass-forming Eosinophilic Infiltration Involving the Central Airway: A Case Report of Endobronchial Involvement of Eosinophilic Inflammation Mimicking Bronchogenic Malignancy

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We report a very unusual case of endobronchial involvement of eosinophilic inflammation. A 58-year-old woman visited our clinic complaining of cough. A chest computed tomography scan showed a poorly enhancing mass compressing both main bronchi in the subcarinal area. Bronchoscopy also showed stenosis of the two main bronchi with irregular mucosal thickening. A bronchoscopic endobronchial mucosal biopsy revealed eosinophilic inflammation without evidence of malignancy. The subcarinal mass disappeared after systemic steroid treatment. This is the first report of mass-forming eosinophilic infiltration involving the central airway mimicking primary lung cancer. (Korean J Med 2014;87:733-737)

Keywords: Eosinophils; Subcarinal mass; Bronchial stenosis

INTRODUCTION

Eosinophilic lung diseases are a diverse group of pulmonary disorders associated with peripheral or tissue eosinophilia. They are classified as eosinophilic lung diseases of unknown cause, eosinophilic lung diseases of known cause, and eosinophilic vasculitis [1]. Eosinophilic infiltration in these diseases usually involves the lung parenchyma.

Eosinophilic infiltration in a bronchus usually indicates an allergic disease, such as bronchial asthma or eosinophilic bronchitis. The typical pattern of bronchial involvement is diffuse airway inflammation without a definite radiological abnormality.

Herein, we report a case of a subcarinal tumor with eosinophilic inflammation in a patient with a history of multiple organ
involvement (breast, oral cavity, and cervical lymph node). The initial impression based on the radiological findings was a lung malignancy, such as primary lung cancer or lymphoma. However, a bronchoscopic biopsy of the subcarinal mass revealed only eosinophilic infiltration. The mass decreased in size after steroid therapy. The patient had no peripheral eosinophilia or other allergic cause.

CASE REPORT

A 58-year-old woman presented to our clinic with a non-productive cough for the past month. The patient took regular medication for hypertension, diabetes mellitus, and dizziness. She had been diagnosed with breast cancer 2 years earlier and had undergone a partial mastectomy and 12 courses of chemotherapy with radiotherapy at another hospital. She had also been taking an aromatase inhibitor for 1 year.

She had presented with a right breast mass 1 year 8 months earlier and had undergone a breast gun biopsy. The biopsy showed extensive eosinophilic infiltration. The breast mass decreased in size after steroid therapy. She presented with a right upper gingival mass 1 year later. Small nodules in the right middle lobe of the lung and cervical lymphadenopathy were discovered incidentally on positron emission tomography-computed tomography (PET-CT). The biopsy revealed nonspecific inflammation and focal mild eosinophilic infiltration at the gingival mass, organizing pneumonia in the right middle lobe, and reactive hyperplasia with some eosinophilic and neutrophilic infiltration in level II cervical lymph nodes. No specific treatment was prescribed, and the patient was followed up regularly.

No specific findings were noted on a physical examination, including lung auscultation, except decreased breathing sounds in both lung fields. Initial laboratory studies showed a white blood cell count of $4.24 \times 10^9/L$, hematocrit of 33\%, platelet count of $312 \times 10^9/L$, and eosinophil count of 3.8%. The total IgE level was within the normal range (60.8 U/mL). A chest X-ray did not show any other pulmonary problems. However, a chest CT scan revealed a poorly enhancing conglomerated mass ($4.7 \times 3.2 \text{ cm}$) in the mid-mediastinum. This mass was located over the lower paratracheal, subcarinal, and paraesophageal areas. Endobronchial narrowing from both the main bronchi to the proximal bronchus intermedius was also noted (Fig. 1A). A PET scan re-
Figure 2. Positron emission tomography scan reveals hypermetabolism at the endobronchial mass (p-SUV, 6.1). No other significant abnormal hypermetabolic lesions suggesting malignancy were detected.

Figure 3. Histopathological examination at the abnormal mucosal surface of the tumor reveals chronic inflammation and eosinophilic infiltration (hematoxylin and eosin stain, ×40).

Figure 4. A follow-up computed tomography scan reveals that the previously noted soft tissue mass in the subcarinal area had resolved, and that endobronchial narrowing of the bronchus intermedius and bilateral main bronchi had improved (A). Follow-up bronchoscopy revealed improved bronchial obstruction with a recovered mucosa (B).

evealed hypermetabolism (p-SUV, 6.1) only at the mass without an abnormality at any other site (Fig. 2). A bronchogenic malignancy was suspected, and a bronchoscopy was performed. Irregular mucosal thickening with stenosis was detected in both main bronchi, particularly the carinal area. A 7-mm scope was passed through the left bronchus intermedius but could not be passed through the right bronchus intermedius (Fig. 1B). A bronchoscopic biopsy was performed at the abnormal mucosal surface of the tumor. Airway inflammation with eosinophilic infiltration was evident in the endobronchial mucosa. No evidence of malignancy was detected in the biopsy specimen (Fig. 3).

We performed several diagnostic studies to rule out a secon-
dary cause for the eosinophilic infiltration. We performed a bone marrow aspiration and biopsy to rule out chronic eosinophilic leukemia; we detected normocellular marrow with mild interstitial eosinophilic infiltration. Expression of platelet-derived growth factor receptor (PDGFR) was negative. She had no history of asthma or other allergic disease. She denied a history of taking medication that could have caused eosinophilia. Serological markers for a parasitic infection (Clonorchis, Paragonimus, Cysticercus, and Sparganum) were all negative.

We started intravenous administration of 62.5 mg/day methylprednisolone (-1 mg/kg) and then tapered this dosage. We repeated the chest CT and bronchoscopy after 1 month of steroid therapy. The chest CT after treatment showed that the previously noted soft tissue mass in the subcarinal area had almost resolved, and that the endobronchial narrowing of the bronchus intermedium and bilateral main bronchi had improved (Fig. 4A). A bronchoscopy after treatment showed improved bronchial obstruction with recovered mucosal thickening (Fig. 4B). The patient has been taking 10 mg/day prednisolone and is continuing regular follow-up. She did not show any symptoms at the 6-month follow-up, and her physical examination was normal.

DISCUSSION

Eosinophilic lung diseases are a heterogeneous group of pulmonary disorders characterized by an increased number of eosinophils in the airway or lung parenchyma. Their major differential points are presence of asthma, peripheral eosinophilia, bronchoalveolar lavage (BAL) fluid eosinophilia, IgE level, and radiological findings. This case showed no evidence of asthma, peripheral or BAL fluid eosinophilia, or increased IgE level. The chest CT findings also did not meet the characteristics for eosinophilic lung disease. This case was very unusual, as it did not qualify as an eosinophilic lung disease but showed extensive eosinophilic infiltration on the endobronchial lesion histology.

Diseases that cause eosinophilic infiltration, such as allergic diseases, parasite infections, and neoplasms, were ruled out. A secondary cause for the eosinophilic infiltration was not found. We performed a bone marrow aspiration and biopsy to rule out chronic eosinophilic leukemia, which could cause infiltration into various body tissues. However, we found normocellular marrow with mild interstitial eosinophilic infiltration. PDGFR expression was also negative.

In particular, the pattern of bronchial involvement was very noticeable. In general, eosinophilic infiltration in a patient with eosinophilic lung disease usually involves the lung parenchyma. Such diseases usually show nonspecific diffuse airway inflammation when the bronchus is involved without a prominent radiological abnormality. However, the eosinophilic infiltration formed an airway-obstructing mass > 4 cm, mimicking primary lung cancer. Such a case has not been reported previously. This case showed entirely different radiological and endoscopic patterns than those usually seen in patients with eosinophilic lung disease.

Few reports have been published regarding bronchial involvement by eosinophils. One such report involved two patients with Churg-Strauss syndrome who had multiple tracheobronchial nodular lesions [2]. The histopathology of these lesions revealed necrotizing bronchial inflammation with many eosinophils. The other case was a patient with chronic eosinophilic pneumonia who showed endobronchial white nodules revealing eosinophilic inflammation on a histological examination [3]. In another case report, a small polypoid lesion in the main bronchus was associated with asthma, and the histological diagnosis was an inflammatory polyp with marked infiltration of eosinophils [4]. However, our patient was unique, because she presented with a relatively large endobronchial tumor rather than a nodule or polyp without evidence of a parenchymal lesion. Additionally, she had no underlying pulmonary disease, such as Churg-Strauss syndrome, eosinophilic pneumonia, or asthma.

Eosinophils very rarely generate a mass in the bronchus. A few cases has been reported in children and adults [5,6]. These cases presented with pneumonia and atelectasis, respectively, and were confirmed to be eosinophilic granulomas after thoracotomy resection.

A mass-forming eosinophilic infiltration of the central bronchus mimicking primary lung cancer has never been reported. We could not rule out the possibility that this was the secondary
result of a specific unknown disorder. This is the first report of a mass-forming eosinophilic infiltration involving the central airway. Accordingly, we present the case along with references.

중심 단어: 호산구; 기관 분기부하 종괴; 기관지 협착

REFERENCES