

## 흉선 절제술 이후 기질화폐렴의 자연소실

한림대학교 의과대학 호흡기내과학교실

김지현 · 최은정 · 송영진 · 박상면 · 신태림 · 최정희 · 신미경

### A Case of Spontaneous Remission Organizing Pneumonia after Resection of Thymoma

Ji Hyun Kim, Eun Jeong Choi, Young Jin Song, Sang Myeon Park, Tae Rim Shin, Jeong Hee Choi, and Mi Kung Shin

*Division of Pulmonary, Department of Internal Medicine, Hallym University College of Medicine, Seoul, Korea*

Thymoma is a rare neoplasm associated with a number of autoimmune disorders and tumors. Organizing pneumonia is a reaction of the lung to various injuries. A feature of organizing pneumonia is plugs of granulation tissue within the lumens of small airways, or alveoli. Causes of organizing pneumonia include various disorders and conditions; however, thymoma with organizing pneumonia located in both lungs is very rare. We report a case of resolving organizing pneumonia after thymoma resection. Our case is the first to demonstrate that resolving organizing pneumonia can occur after thymoma resection, as has been speculated by many researchers. (Korean J Med 2013;85:92-95)

**Keywords:** Organizing pneumonia; Thymoma

#### INTRODUCTION

Thymoma is the most common neoplasm of the thymus gland. Recent reports have shown incidence estimates of 0.15 per 100,000 person-years in the United States [1]. Thymoma accounts for 20% of mediastinal tumors and is the most common tumor of the anterior mediastinum, accounting for approximately 50% of the anterior mediastinal tumors in adults [2]. Organizing pneumonia (OP) is a disease resulting

from pulmonary reaction to noxious factors and is defined histologically. Regions of OP often accompany lung cancer and are not the predominant clinical, radiological, or pathological features [3]. Thymoma with OP located in both lungs is very rare. Moreover, disappearance of OP after resection of thymoma has not been reported previously. Having searched the literature, no similar co-existence of thymoma and OP was noted. We report a 55-year-old male with spontaneous resolution of OP following surgery for thymoma.

Received: 2012. 7. 6

Revised: 2012. 8. 21

Accepted: 2013. 2. 28

Correspondence to Sangmyeon Park, M.D.

Division of Pulmonary, Department of Internal Medicine, Kangnam Sacred Heart Hospital, Hallym University, 1 Singil-ro, Yeongdeungpo-gu, Seoul 150-950, Korea

Tel: +82-2-829-5318, Fax: +82-2-846-4669, E-mail: thoraxmd@hallym.or.kr

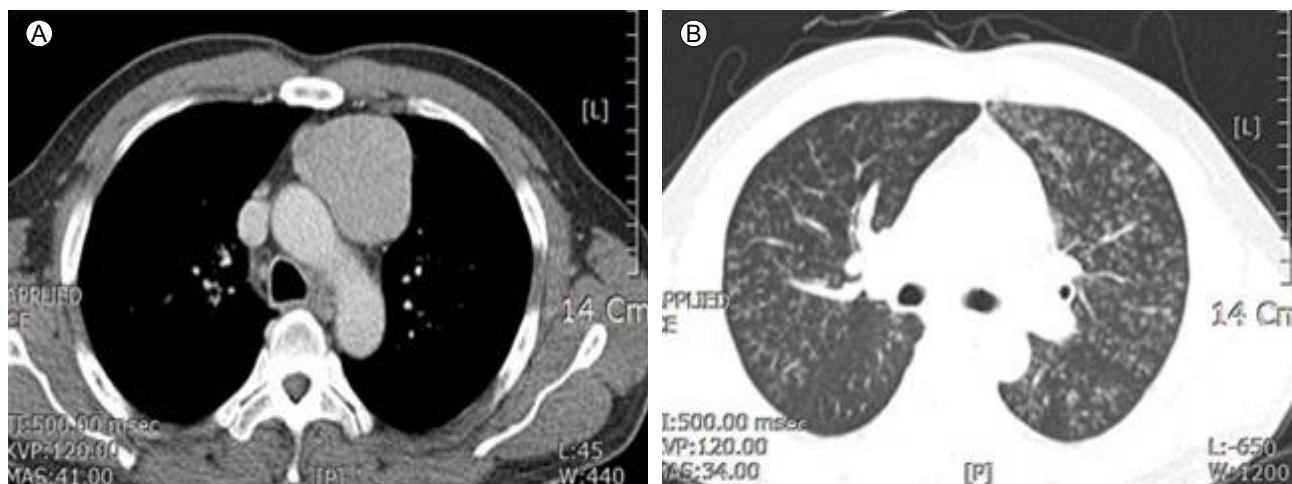
Copyright © 2013 The Korean Association of Internal Medicine

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0/>) which permits unrestricted noncommercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

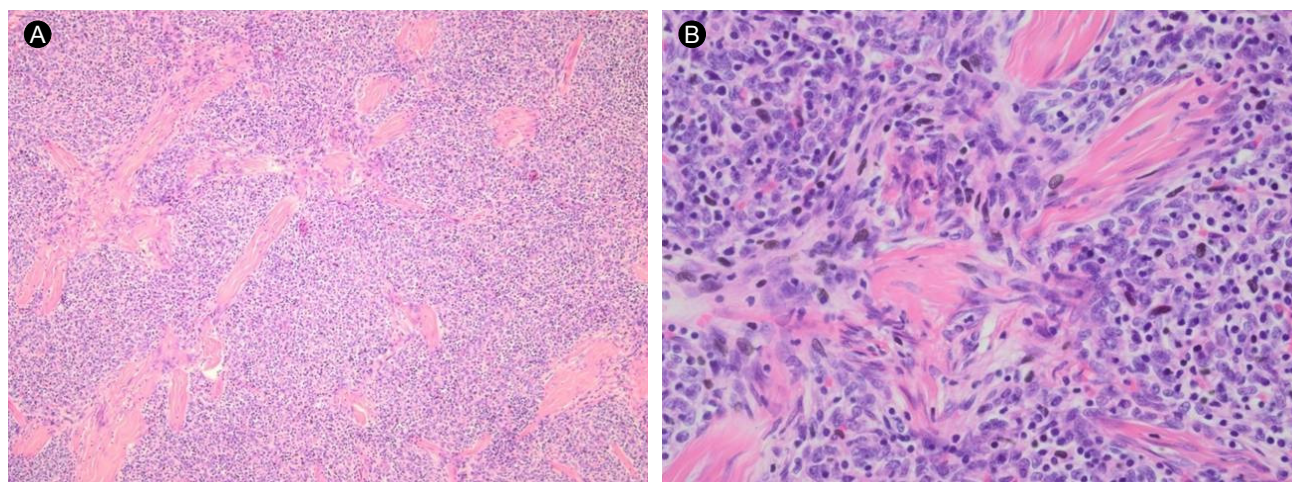
## CASE REPORT

A 55-year-old current smoker (15 pack-years) was admitted to the Kangnam Hospital of Hallym University on July 16, 2009 with diagnosis of an anterior mediastinal mass. His medical history revealed that he was a hepatitis B virus carrier. Three weeks before admission, the patient complained of dry cough and shortness of breath. On physical examination, the patient was in generally good health, except for dyspnea on exertion. Auscultation revealed crackling sounds in the bases of both lungs. On admission, a chest X-ray revealed an anterior

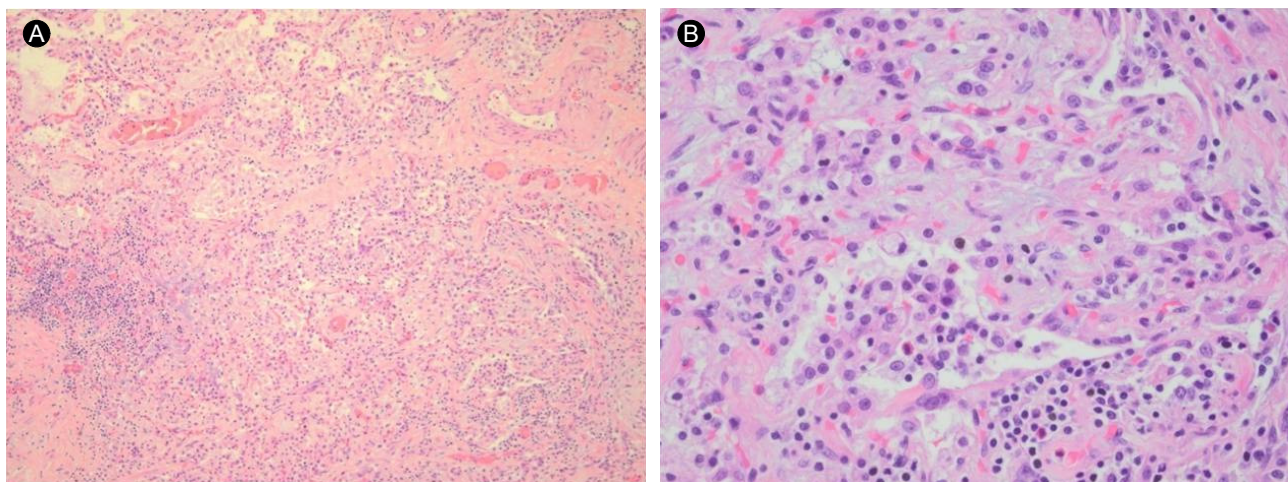
mediastinal mass and reticulonodular densities were noted in both lungs. Forced vital capacity (FVC) and forced expiratory volume in one second (FEV1) were 1.52 L (38% of predicted value) and 0.90 L (30% of predicted value), respectively. FEV1/FVC was 59%. A pulmonary function test revealed a mixed obstructive and restrictive pattern. A chest computed tomography (CT) scan demonstrated a 6.5 × 5.7 cm, well-demarcated and enhanced anterior mediastinum mass, and diffuse centrilobular nodules with tree-in-bud appearance were noted in both lungs (Fig. 1). Repeated sputum cultures for mycobacteria and other pathogens were negative. Acetyl-



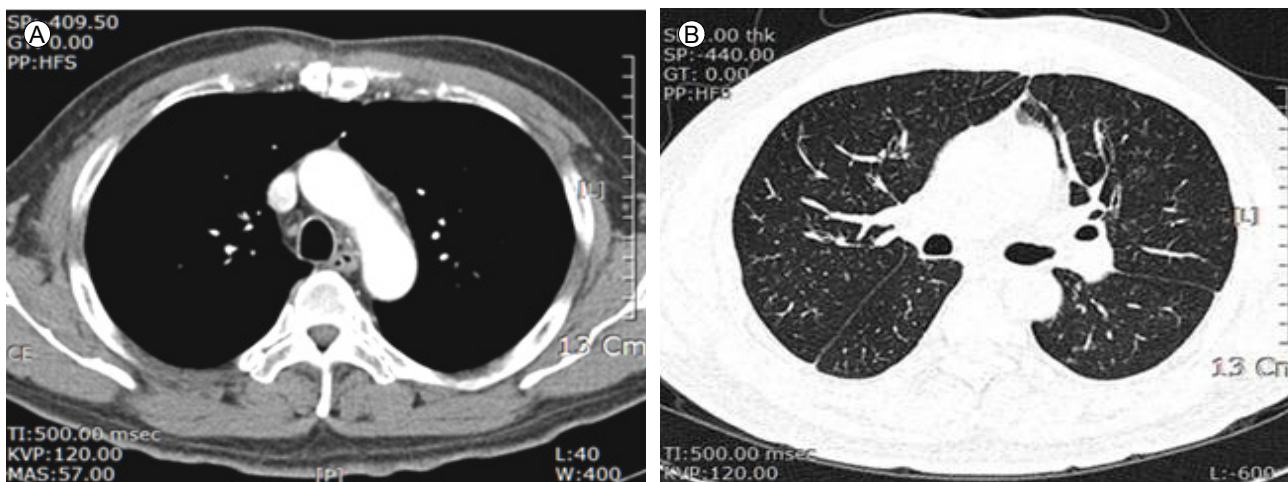
**Figure 1.** Preoperative chest, contrast-enhanced CT shows a large mass in the left anterior mediastinum (A) and multiple, ill-defined, diffuse nodular densities scattered throughout all lung fields (B).



**Figure 2.** Microscopic findings of thymoma. (A) Type AB (mixed) thymoma composed of a mixture of neoplastic epithelial cells and non-neoplastic lymphocytes (H&E, × 100). (B) Type AB (mixed) thymoma showing foci having the features of type A (spindle cell) thymoma admixed with foci rich in lymphocytes (H&E, × 400).



**Figure 3.** Microscopic findings of a lung specimen. (A) Organizing pneumonia showing a branching tongue of fibroblastic proliferation and alveolar macrophage accumulation (H&E, × 100). (B) Organizing pneumonia showing aggregates of fibroblasts in the ground substance and macrophage accumulation (H&E, × 400).



**Figure 4.** Postoperative chest, contrast-enhanced CT shows absence of thymoma (A) and disappearance of the previous multiple, ill-defined, diffuse nodular densities (B).

choline receptor (AChR)-binding antibodies were normal (0.2 nmol/L; normal, < 0.30 nmol/L), and there was no association with myasthenia gravis.

The patient underwent radical surgical procedures (extended total thymectomy and left upper lung wedge resection). Histological examination showed WHO Thymoma classification type AB (mixed) thymoma (Fig. 2). A lung-wedge resection specimen (left upper lobe) contained fragments of alveolar parenchyma with inflammation and intra-alveolar purulence consistent with OP (Fig. 3). Neoplastic cells were not found. The postoperative period was uneventful, and follow-up films

showed that the previous reticulonodular infiltrations were markedly absorbed. After 1 month, a follow-up chest CT scan showed remarkable improvement in the previous centrilobular nodules in both lungs (Fig. 4). The patient remains asymptomatic to-date, with no recurrence of thymoma.

## DISCUSSION

Thymoma is associated with various autoimmune disorders and secondary malignancy. Such autoimmune disorders may include myasthenia gravis, polymyositis, lupus erythematosus,

rheumatoid arthritis, thyroiditis, Sjogren's syndrome, and hypogammaglobulinemia. There have also been occurrences of thymoma in patients with non-Hodgkin's lymphoma and soft tissue sarcoma [4]. However, thymoma with organizing pneumonia (OP) located in both lungs is very rare.

OP is characterized by the accumulation of organized exudates within the alveoli, protruding into the small bronchioli and alveolar ducts in the form of polyps [1]. Cryptogenic organizing pneumonia (COP) is determined when no definitive cause or characteristic clinical context is found. Many diseases and conditions may trigger secondary OP. In secondary OP, causes such as infection, especially *Mycoplasma pneumoniae*, *Chlamydomphila pneumoniae*, and viruses, can be identified, or it might be the consequence of exposure to toxic substances, such as drugs or radiotherapy. Secondary OP has been observed in autoimmune and neoplastic disorders [5]. In several studies, it was found that there is no radiological, clinical, or histopathological difference between secondary and primary OP; however, generally, secondary OP has a worse prognosis. Its clinical manifestation comprises fever, dry or productive cough, dyspnea of varying severity, chest pain, sweats, and weakness. The most common radiographic abnormalities in OP are bilateral, patchy airspace opacities. Bilateral, migratory parenchymal infiltrates with air bronchogram is the most common radiological sign of OP, but also centrilobular nodules 3-5 mm in diameter confined to one lung or disseminated, bilateral, reticulonodular forms can be found. Nodular patterns of OP are represented as well-defined acinar or subtle, poorly defined nodular patterns [6]. Diffuse nodular patterns mimic miliary tuberculosis [7]. Several recent reports have noted that OP can develop from neoplastic disorders. OP may accompany lung cancer as a result of the direct influence of the tumor on the surrounding lung parenchyma or as a consequence of bronchial obstruction [8].

The mainstay treatment for thymoma is surgery with the goal of achieving a complete surgical resection as the main determinant of survival. OP associated with thymoma is rare, and there has not been a case report of it. The present case represents a very unusual radiographic manifestation of OP, with thymoma and a diffuse nodular pattern that mimics miliary tuberculosis. The standard treatment for OP is corticosteroids, but in some cases treated this way, spontaneous

regression was observed. This has already been described in other, smaller case series or case reports. Some patients that were not treated with corticosteroids nevertheless showed improvement [9]. Another study showed that this phenomenon occurred most often in patients with focal lesions that were surgically removed. It is possible that surgery cures the disease in such cases [10].

**중심 단어:** 기질화폐렴; 흉선종

## REFERENCES

1. Engels EA, Pfeiffer RM. Malignant thymoma in the United States: demographic patterns in incidence and associations with subsequent malignancies. *Int J Cancer* 2003;105:546-551.
2. Cameron RB, Loehrer PJ, Thomas CR Jr. Neoplasms of the mediastinum. In: DeVita VT Jr, Hellman S, Rosenberg SA, eds. *Cancer: Principles and Practice of Oncology*. 6th ed. Philadelphia: Lippincott-Raven Publishers, 2000:1019-1036.
3. American Thoracic Society; European Respiratory Society. American Thoracic Society/European Respiratory Society International Multidisciplinary Consensus Classification of the Idiopathic Interstitial Pneumonias: this joint statement of the American Thoracic Society (ATS), and the European Respiratory Society (ERS) was adopted by the ATS board of directors, June 2001 and by the ERS Executive Committee, June 2001. *Am J Respir Crit Care Med* 2002;165:277-304.
4. Pan CC, Chen PC, Wang LS, Chi KH, Chiang H. Thymoma is associated with an increased risk of second malignancy. *Cancer* 2001;92:2406-2411.
5. Cordier JF. Cryptogenic organising pneumonia. *Eur Respir J* 2006;28:422-446.
6. Oikonomou A, Hansell DM. Organizing pneumonia: the many morphological faces. *Eur Radiol* 2002;12:1486-1496.
7. Fruchter O, Solomonov A, Guralnik L, Naroditsky I, Yigla M. An unusual radiographic manifestation of bronchiolitis obliterans organizing pneumonia. *J Thorac Imaging* 2007; 22:263-264.
8. Radzikowska E, Nowicka U, Wiatr E, et al. Organising pneumonia and lung cancer-case report and review of the literature. *Pneumonol Alergol Pol* 2007;75:394-397.
9. Oymak FS, Demirbaş HM, Mavili E, et al. Bronchiolitis obliterans organizing pneumonia: clinical and roentgenological features in 26 cases. *Respiration* 2005;72:254-262.
10. Sveinsson OA, Isaksson HJ, Sigvaldason A, Yngvason F, Aspelund T, Gudmundsson G. Clinical features in secondary and cryptogenic organising pneumonia. *Int J Tuberc Lung Dis* 2007;11:689-694.