

## An Endobronchial Inflammatory Myofibroblastic Tumor Treated by Modified Left One-stoma-type Carinoplasty

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Endobronchial inflammatory myofibroblastic tumor is a rare primary lung disease. A 39-year-old woman with dyspnea and a productive cough underwent complete surgical resection of a small-sized inflammatory myofibroblastic tumor that invaded the left main bronchus and the carina with lung-saving modified left one-stoma-type carinoplasty. We report this case with a review of literature.

Key words: 1. Tumor, benign  
2. Bronchial neoplasms  
3. Carina  
4. Bronchoplasty

### CASE REPORT

A 39-year-old female patient was referred to us for surgical treatment. She was a non-smoker but had undergone medical treatment for pulmonary tuberculosis for several years and was receiving ventilator care until just before her transfer from the local medical clinic. At that time, she complained of a moderate degree of dyspnea and a cough with purulent sputum despite extubation. Vital signs and an electrocardiogram were within normal limits but we could auscultate rhonchi and wheezing in the left lung field by physical examination. Routine blood tests were unremarkable with the exception of hypokalemia of 2.7 mEq/L and mild hypoxemia of PaO<sub>2</sub> 68 mmHg.

Computed tomography (CT) showed an endobronchial mass-like lesion obstructing the left main bronchus, stretching from the carina to 2 cm distal with segmental atelectasis and extraluminal invasion to subcarinal area (Fig. 1A). A bron-

choscopic finding revealed a yellowish endobronchial mass obstructing the proximal portion of the left main bronchus (Fig. 1B).

We planned segmental resection of the tumor containing left main bronchus which was followed by bronchoplasty using end to end anastomosis. The operation was accomplished through a left posterolateral thoracotomy via the 4th intercostal space. The most proximal left main bronchus was transected from carina for safe margin according to preoperative bronchoscopic finding, but unexpectedly the tumor was found to be grossly invading part of the carina extraluminally. We performed a complete tumor resection including part of the carina and the left main bronchus, followed by modified left one-stoma-type carinoplasty (Fig. 2) with tension free anastomosis by U-shaped pericardial incision below the left inferior pulmonary vein and reinforcement of the anastomotic site with the mediastinal fat pad.

In the macroscopic pathologic findings, a yellowish mass,

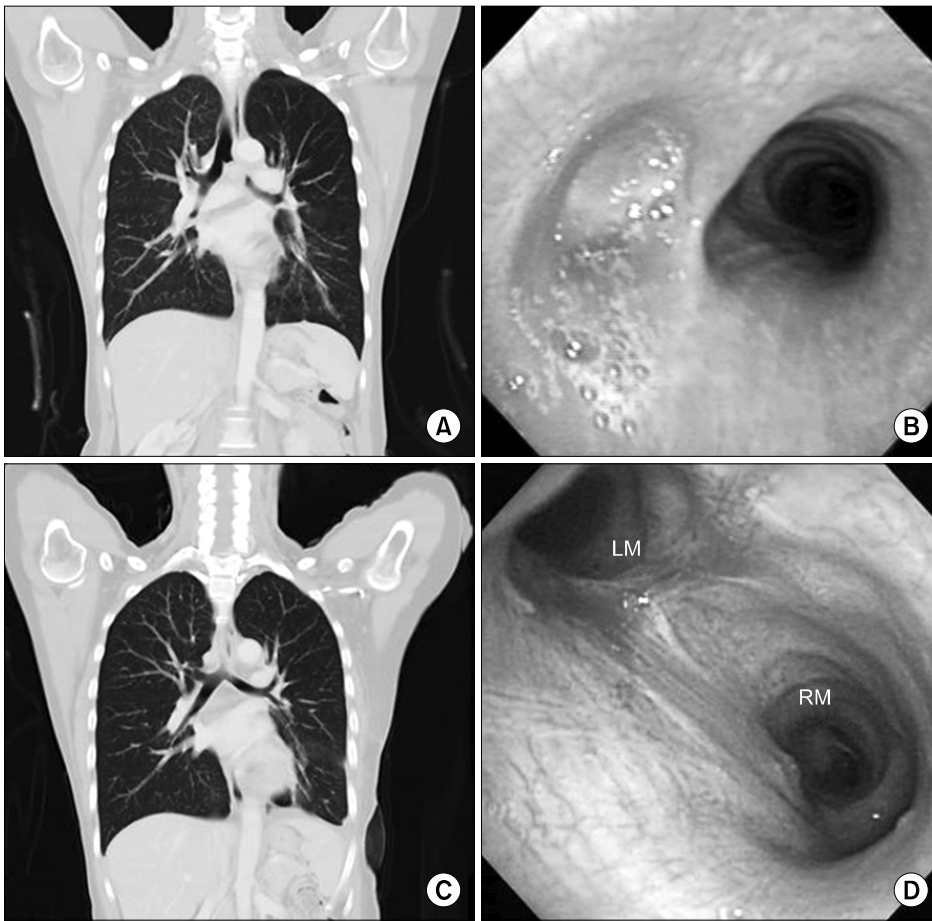
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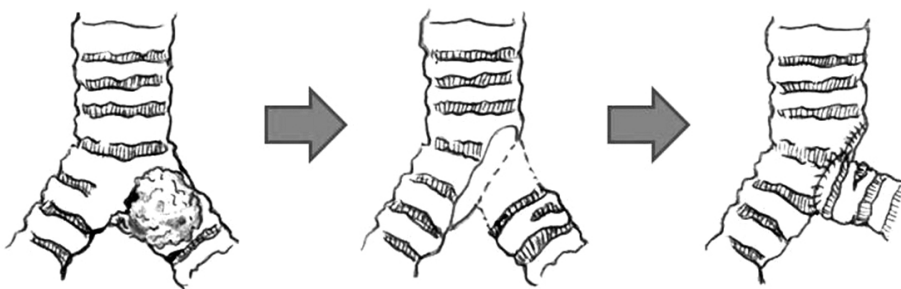
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**Fig. 1.** (A) Preoperative chest computed tomography (CT) scan shows an endobronchial mass obstructing the left main bronchus, and extraluminally invading subcarinal area. (B) Upon bronchoscopy, a endobronchial mass obstructing the left main bronchus is noted. (C) Postoperative chest CT scan shows good patency of the airway. (D) Bronchoscopy shows luminal patency at the anastomotic site without narrowing at 6 months after operation. LM, left main bronchus; RM, right main bronchus.

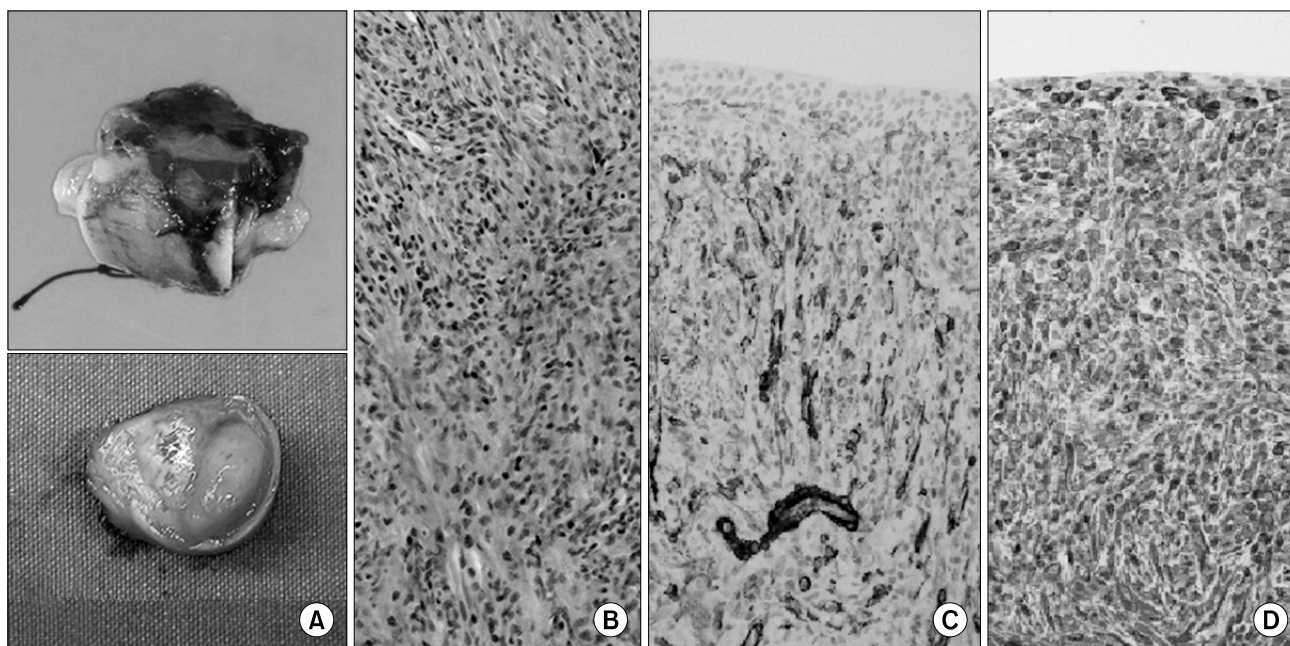


**Fig. 2.** Schema of modified left one-stoma-type carinoplasty; resection of the proximal left main bronchus and part of the carina followed by partial closure of the upper portion of stoma and end to side anastomosis between the remnant distal portion of the left main bronchus and stoma of the carina.

2.5×1×1 cm in size and almost completely obstructing the lumen of the left main bronchus with focal extraluminal invasion to the carina was noted (Fig. 3A). Microscopically, we noted a fusiform nucleus with abundant fibrous matrix, infiltration of lymphocytes and plasma cells (Fig. 3B). Tumor involved whole layer of bronchial wall and resection margins were free from tumor. A definitive pathological diagnosis of inflammatory pseudotumor was made according to im-

munohistochemical staining for actin (+), vimentin (+), S-100 (+, partial), and C-kit (-) (Fig. 3C, D).

A small amount of air leakage persisted until the 10th postoperative day but ceased spontaneously. The luminal patency was good without recurrence or stenosis through 10 months after surgery according to CT scans and bronchoscopy (Fig. 1C, D).



**Fig. 3.** (A) Gross finding of resected endobronchial mass. (B) Microscopic findings showing abundant spindle cells and inflammatory cells (H&E, ×200). (C) Immunostaining for smooth muscle actin (×400) and (D) vimentin (×400) is diffusely strongly positive on the neoplastic cells.

## DISCUSSION

Inflammatory myofibroblastic tumors are also known as plasma cell granulomas, inflammatory pseudotumors, xanthomatous pseudotumors, fibrous histiocytomas, and mast cell granulomas. The main pathologic characteristic is spindle cell proliferation with a fibroinflammatory appearance. These tumors can develop in any part of the body but are most commonly found in the thorax; however, they rarely manifest as primary lung tumors, with an incidence of only 0.04% to 1% [1,2].

The cause of inflammatory myofibroblastic tumors is not completely understood, but several theories suggest that these lesions represent past inflammatory tissue reactions [3] and possibly immune-related lesions due to recent viral infections such as Epstein-Barr virus and human herpes virus-8. However, neoplastic disease is generally accepted as the cause based on the adjacent tissue invasion, metastasis, recurrence and molecular genetic evidence [2,4].

Inflammatory myofibroblastic tumors occur preferentially at a younger age, with an average reported onset of 37.9 years,

and in men (22 in 28 cases, 81.5%). The clinical manifestation is most commonly a cough and is often accompanied by dyspnea, sputum, hemoptysis, low fever, and chest pain. An asymptomatic patient often may present with an incidental mass in radiologic images.

The most common site of origin is the intrapulmonary parenchyma, but endobronchial and bronchial lesions are relatively uncommon [5]. The histopathologic diagnosis before surgery is difficult because the lesion contains abundant inflammatory tissue [1], and differential diagnosis from other primary pulmonary tumors is also difficult because the radiologic findings are most commonly reported as a solitary well-demarcated tumor with or without calcification, cavitation or multinodular densities. In addition, when the tumor size is very large, pneumonia or atelectasis induced by the secondary mass effect is apt to be misdiagnosed [6]. Complete surgical resection is an essential therapeutic modality because incomplete tumor removal affects recurrence and prognosis [2,7].

In the operative technique, we performed resection of the proximal left main bronchus and part of the carina containing

the extraluminally invaded endobronchial mass. Because of size discrepancy between large oval-shaped proximal stoma and the distal portion of the left main bronchus, we accomplished partial repair of the upper portion of stoma using several stitches of interrupted 4-0 Vicryl (Ethicon Inc., Johnson and Johnson company, Somerville, NJ, USA) suture and then end to side anastomosis between the remnant distal portion of the left main bronchus and stoma of the carina (modified one-stoma-type carinoplasty) [8] (Fig. 2). We applied the hilar release technique by U-shaped pericardial incision below the left inferior pulmonary ligament for tension free anastomosis.

Modified one-stoma-type carinoplasty could make surgeons avoid an aggressive carinal resection for lung-saving and showed a good result without luminal narrowing.

There is much debate about the therapeutic effectiveness of radiotherapy, chemotherapy, and corticosteroid therapy when complete resection is not tolerable. In general, complete resection results in a better prognosis [2,7], but a few delayed recurrence after surgical resection implies the need for long-term follow-up.

Here we reported a rare case of endobronchial myofibroblastic tumor invading carina, even the access to the carina from left side is troublesome because of the anatomy of the aortic arch and narrow surgical field, we successfully treated

by complete resection with lung-saving modified left one-stoma-type carinoplasty.

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