

## Idiopathic Localized Bronchostenosis in an Adult Man with Frequent Recurring Pneumonia

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### Abstract

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The case of a 48-year-old Japanese man with idiopathic bronchostenosis in the right lower lobe is reported. The patient had fourteen episodes of pneumonia in two years and therefore surgical resection of the right lower lobe was performed for both diagnosis and treatment. Histopathology demonstrated no evidence of malignancy, tuberculosis, sarcoidosis or amyloid deposition. Despite an exhaustive evaluation, a specific etiology was never determined. The patient was given the diagnosis of acquired idiopathic localized bronchostenosis with frequent recurrence of pneumonia.

**Key words:** idiopathic, bronchostenosis, pneumonia, lobectomy, adult

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### Introduction

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The reported cases of idiopathic tracheal stenosis are few and cases of bronchial stenosis are even more rare. Both are characterized by chronic inflammation with fibrotic tissue formation and patients present with dyspnea, wheezing or stridor. A case of idiopathic bronchostenosis with unusual presentation is reported. Secondary bronchial stenosis, such as a congenital lesion, effects of granulomatous diseases, bronchomalacia and recurrent aspiration all needed to be considered in the differential diagnosis.

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### Case Report

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A 48-year-old Japanese man visited the Department of Respiratory Medicine in our hospital for evaluation of hemoptysis and purulent sputum. In the previous 14 months he had been diagnosed with bacterial pneumonia in the right lower lobe on seven occasions and had been treated with multiple courses of empiric antibiotics. He had a fifty pack-year smoking history but no exposure to dust or fumes. His past medical history was notable for a subarachnoid hemor-

rhage five years prior from which he recovered without residual defects. He denied a history of major chest trauma, cardiac disease or thoracic surgery and had no family history of heritable illness.

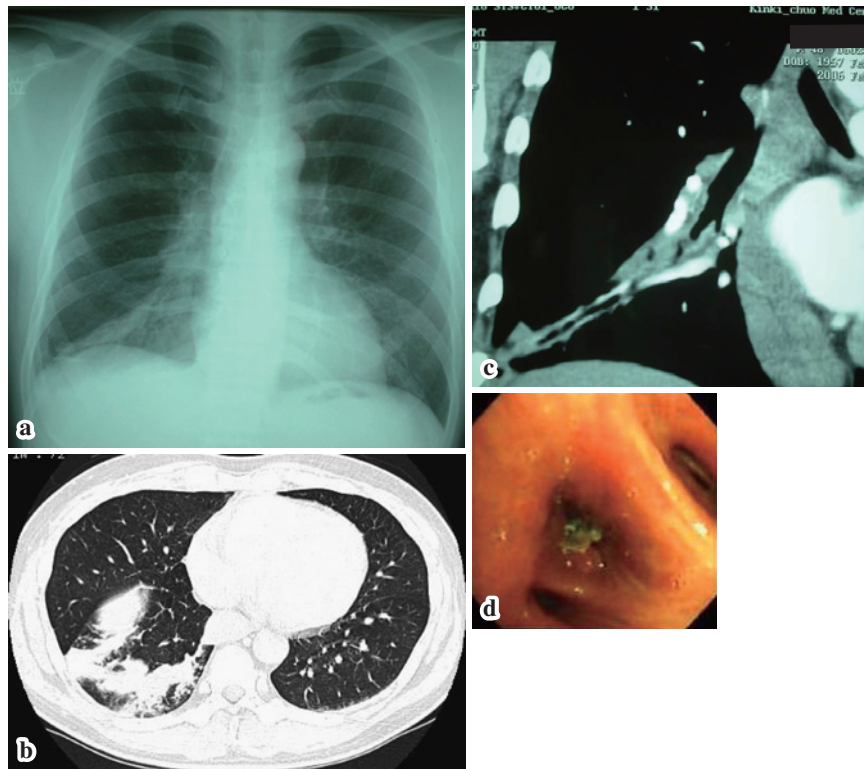
The patient had a temperature of 37.6°C and a pulse of 98 beats per minute. He had diminished respiratory sounds in the right lower lung field, but no wheezing or egophony. Percussion was dull in same field. A chest radiograph demonstrated an infiltrate in the right lower lobe (Fig. 1a). Laboratory investigation revealed a white blood cell count of 9,300/mm<sup>3</sup> (74.2% neutrophils) and a C-reactive protein level of 1.4 mg/dL (normal, <0.04 mg/dL). Liver and renal function tests were normal. The patient was diagnosed with right lower lobe pneumonia. Since he had been treated for pneumonia in the same region on multiple occasions he was admitted to the hospital for further evaluation and definitive therapy. Microbiological examinations of his sputum were negative, including bacterial culture, acid fast staining and polymerase chain reaction for mycobacteria. Spirometry was normal with a forced vital capacity (FVC) that measured 3,250 mL (93% predicted), a forced expiratory volume at 1 second (FEV<sub>1</sub>) that measured 2,390 mL (78.6% predicted) and a FEV<sub>1</sub>/FVC ratio that was 73.5%.

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**Figure 1.** a, b: Chest radiograph and CT scan demonstrated an infiltrate in the right lower lobe on admission. c: CT scanning demonstrated a stenotic region in the right lower lobe bronchus with a thickened bronchial wall. d: FB demonstrated severely narrowed right lower lobe bronchi with smooth mucosal surfaces and scant amounts of purulent white material. The patency of airway was not visible.

CT scanning of the thorax demonstrated a stenotic region in the right lower lobe bronchus with a thickened bronchial wall and peripheral airspace consolidation (Fig. 1b, c). The stenosis was confined to the lobar and segmental bronchus and did not involve sub-segmental bronchi.

Evaluation with fiberoptic bronchoscopy (FB) demonstrated severely narrowed right lower lobe bronchi with smooth mucosal surfaces and scant amounts of purulent white material (Fig. 1d). Visualization of sub-segmental bronchi in the right lower lobe was impossible due to edematous mucosa caused by acute infection. The appearance of the airways in other lobes was normal. Bronchial mucosal biopsies were performed in the stenotic region. Tissue histology was normal and demonstrated no evidence of malignancy or inflammation.

Since the patient had a history of tobacco use, positron emission tomography (PET)-CT was performed. No evidence of malignancy was found. The patient reportedly denied dysphagia and recurrent aspiration. Evaluation by a speech therapist revealed normal swallowing function.

The patient had six more episodes of pneumonia in the right lower lobe of his lung after his initial visit to our hospital. On each occasion, he was treated and cured with antibiotics. Due to the high frequency of recurrent pneumonias, the patient was referred for surgical resection of right lower lobe.

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## Histological Findings

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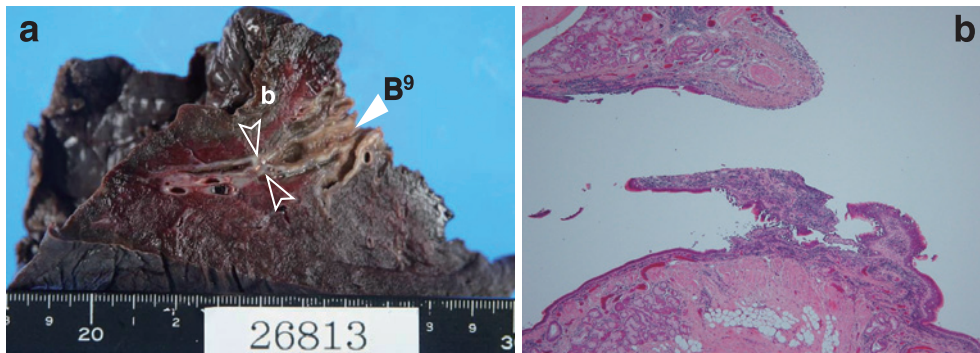
The resected lower lobe of the right lung revealed a focal luminal narrowing without submucosal scarring in a segmental bronchus (Fig. 2a). The stenotic portion showed a valve-like projection of the mucosa which was covered by pseudostratified columnar epithelium. Cilia were present and histologically unremarkable. Moderate infiltration of lymphocytes, lymphoid follicles and bundles of smooth muscle cells were seen in the subepithelial layer (Fig. 2b). Mucous glands were slightly enlarged in the bronchial wall adjacent to the narrowing portion. In the mucosa of the main and lobar bronchus, other segmental bronchi and bronchioles, lymphocytic infiltration and lymphoid follicles were present, but there was no evidence of acute and chronic severe inflammation, malignancy, tuberculous lesion, sarcoidosis or amyloid deposition. Ultrastructural examination was not performed. Consequently, the patient was given the diagnosis of acquired localized bronchostenosis.

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## Discussion

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We believe that this case represents a rare occurrence of adult idiopathic bronchostenosis without severe inflammation. Reported cases of idiopathic tracheal stenosis are few



**Figure 2.** a: Histological findings. Resected lower lobe of the right lung revealed a focal luminal narrowing without submucosal scarring in a segmental bronchus. b: Hematoxylin and Eosin staining 4×. The stenotic portion showed a valve-like projection of the mucosa which was covered by pseudostratified columnar epithelium. Moderate infiltration of lymphocytes, lymphoid follicles and bundles of smooth muscle cells were seen in the subepithelial layer. Mucous glands were slightly enlarged in the bronchial wall adjacent to the narrowing portion.

and cases of bronchial stenosis are even more rare. According to those reports, idiopathic stenosis of the airway usually shows evidence of severe inflammation such as fibrous expansion of the mucosa and ulceration with granulation tissue formation (1). Secondary bronchial stenosis can be seen as a complication of surgical procedures such as lung transplantation and bronchoplasty, following tissue injury, as a congenital lesion, and as a result of granulomatous diseases such as sarcoidosis and tuberculosis (2, 3). In the present patient, a congenital lesion seems unlikely since the patient had no pulmonary problems as a child or young adult. Similarly, there was no evidence of granulomatous disease or tuberculosis. The absence of inflammatory cells and fibrovascular connective tissue deposition in the bronchial submucosa from our patient is noteworthy since most cases of localized airway stenosis are caused by active inflammatory processes (1). Although lymphocytes in the subepithelial region were observed, it is unlikely that they induced severe airway stenosis. Rather, we suggest that the lymphocytic infiltration resulted from recurrent infections.

Histologically, the cartilage was moderately torn in the stenotic region, suggesting the possibility of bronchomalacia. Bronchomalacia is an unusual cause of bronchostenosis and recurrent pneumonia. The most common cause of acquired bronchomalacia is chronic tobacco smoke exposure in association with chronic bronchitis and/or emphysema (4, 5). Although the present patient had an extensive smoking history, he did not have any symptoms of COPD and his

FEV<sub>1</sub>/FVC ratio was not significantly decreased. Moreover, his CT scan showed no evidence of smoking-related lung disease. While it is possible that chronic irritation from tobacco smoke might have contributed to the development of bronchomalacia it is unlikely that it was the sole contributor since the patient had no clinical evidence of bronchial disease in other regions of the lung.

Surgical resection is the most common method used to treat localized bronchial stenosis. For patients who cannot tolerate surgery, alternative approaches exist. Mayse et al reported successful bronchoscopic balloon dilatation on non-malignant stenosis of bronchi (6) and Ball et al used steroids for two of four cases in which stenosis recurred at short intervals after bronchoscopic dilatation to maintain clinical improvement (2). Recent reports suggest that bronchial laceration is relatively common following balloon dilatation, but that transmural laceration is rare (7). Therefore, dilatation is best performed by experienced operators and patients should be monitored closely following the procedure. Our patient did well following surgical resection of the affected region. Thirty-six months after surgical intervention the patient is doing well with no evidence of recurrence.

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