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Endobronchial Hamartoma as a Cause of Pneumonia

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Conflict of interest: None declared

Patient: Male, 66
Final Diagnosis: Endobronchial hamartoma
Symptoms: Fever
Medication: —
Clinical Procedure: Flexible bronchoscopy • surgical resection
Specialty: Pulmonology

Objective: Unusual clinical course

Background: Post-obstructive pneumonia occurs in the presence of airway obstruction, usually caused by lung cancer. However, there are cases of bronchial obstruction due to benign origin such as foreign bodies and benign endobronchial tumors, which are often misdiagnosed.

Case Report: A 66-year-old man was referred to our hospital due to high fever with abnormal shadow in the right lung. Chest computed tomography after a course of antibiotic treatment showed an intra-bronchial tumor obstructing the right upper bronchus. Part of the tumor was removed with flexible bronchoscopy, and histopathological examination revealed cartilage tissue but not fat or other components. Lobectomy of the right upper lobe of the lung was performed to make a definite diagnosis and prevent recurrent obstructive pneumonia. The resected tumor contained mature cartilage and fat tissues, and was diagnosed as endobronchial hamartoma.

Conclusions: Benign endobronchial tumors such as hamartomas should be considered in the differential diagnosis of post-obstructive pneumonia.

MeSH Keywords: Bronchial Neoplasms • Hamartoma • Pneumonia

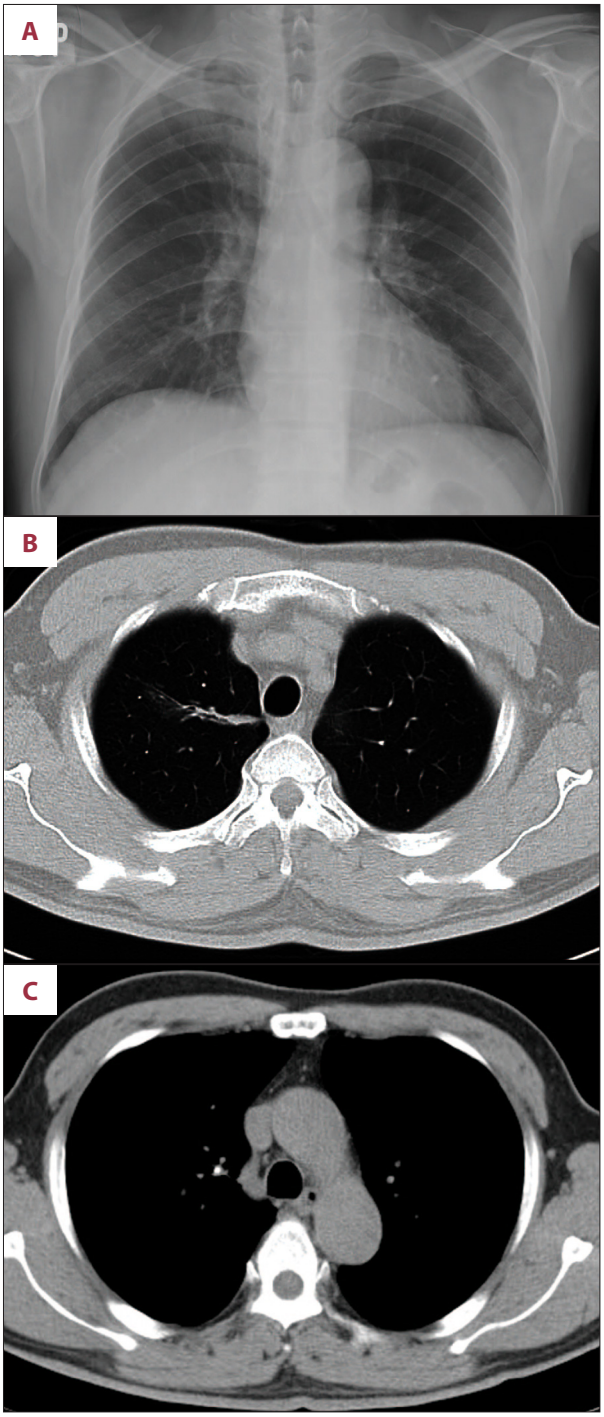
Full-text PDF: http://www.amjcaserep.com/abstract/index/idArt/890869

Background

Post-obstructive pneumonia occurs in the presence of airway obstruction, usually caused by lung cancer [1]. However, there are cases of bronchial obstruction due to benign origin such as foreign bodies and benign endobronchial tumors, which are often misdiagnosed. We report a case of endobronchial hamartoma that caused post-obstructive pneumonia.

Case Report

A 66-year-old man was referred to our hospital due to high fever with an abnormal shadow in the upper field of the right lung on chest radiograph. Based on the diagnosis of pneumonia, he was treated with antibiotics for 3 days prior to the visit to our hospital. On examination, he was afebrile and showed no abnormal physical findings, including breath sounds. Laboratory tests showed an elevated level of C-reactive protein with no abnormality in serum tumor makers or other data. Chest radiography showed a localized consolidation in the right upper lung (Figure 1A) and computed tomography (CT) showed a linear atelectasis. A follow-up CT scan 6 weeks later revealed a



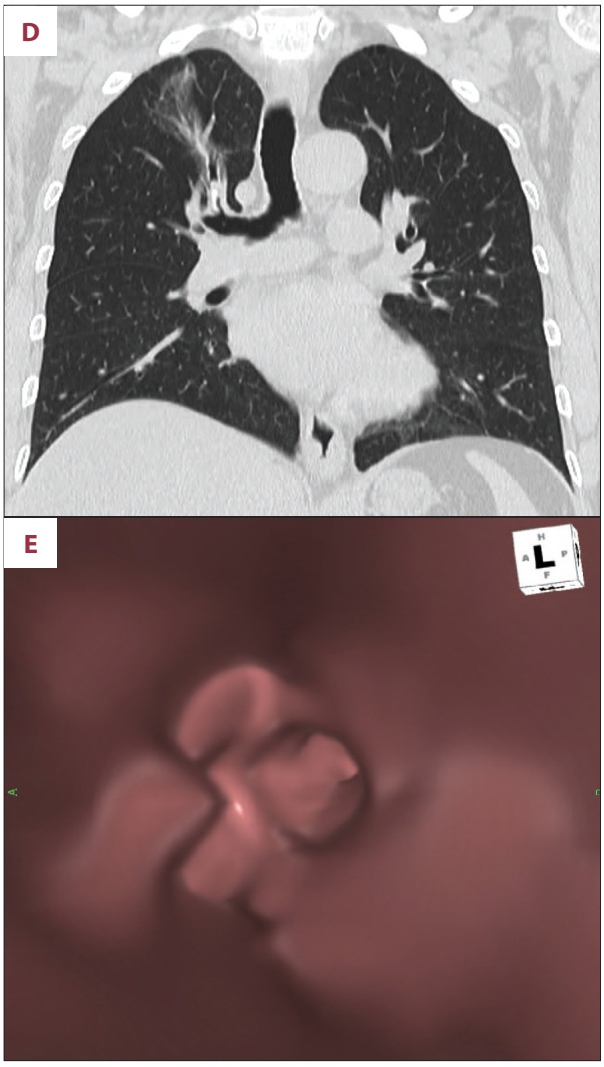


Figure 1. Chest radiograph (**A**) on the first visit and follow-up chest CT scan (**B**) 6 weeks later. Linear atelectasis was present in the right upper lobe (**B**) with a small calcified nodule in the right upper bronchus (**C, D**). Virtual CT bronchoscopy (**E**) demonstrated the tumor in the right upper bronchus.

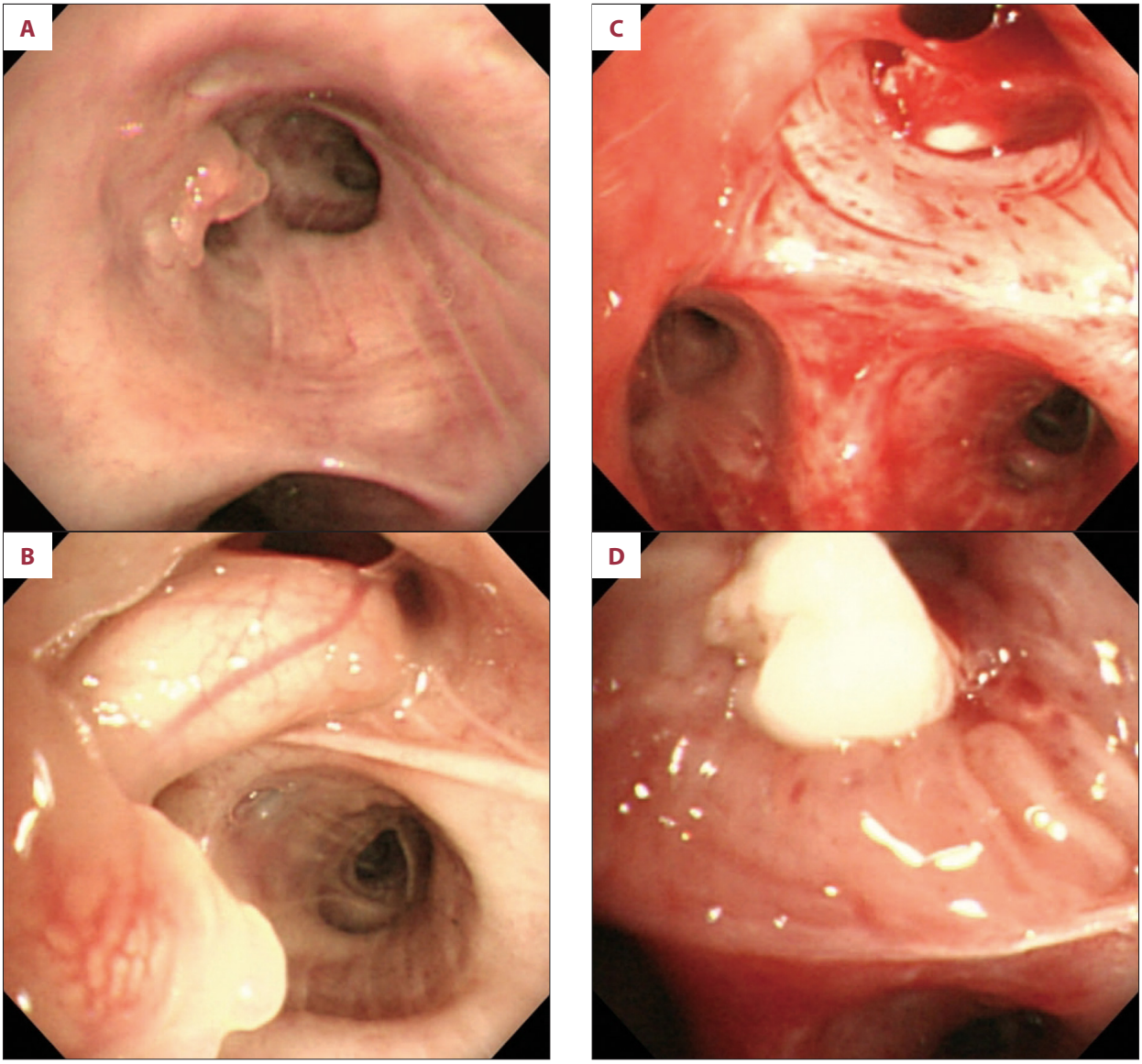


Figure 2. Bronchoscopic examination showed an intra-bronchial tumor in the right upper bronchus (**A, B**). The right upper bronchus was still partially obstructed by the remnant tumor after removal with a biopsy forceps (**C, D**).

reduced size of the linear atelectasis in the right upper lobe (Figure 1B), and a small, calcified lesion in the right upper bronchus was detected (Figure 1C and 1D). Virtual CT bronchoscopy demonstrated an endobronchial lesion in the right upper bronchus (Figure 1E), suggesting the possibility of broncholithiasis, granuloma, or an endobronchial tumor.

Flexible bronchoscopy was performed to investigate the endobronchial lesion, identifying a polypoid tumor in the right upper bronchus that partially obstructed it (Figure 2A and 2B). Part of the tumor was removed with biopsy forceps (Figure 2C), but the right upper bronchus remained obstructed by the remnant tumor (Figure 2C and 2D). Pathological examination of the removed tumor showed cartilage, but not fat or other components (Figure 3). Examination with bronchoscopy and CT scan was repeated 2 and 6 months later, showing no regrowth of the tumor that still obstructed the upper bronchus. Approach to remove the tumor with biopsy forceps through thin flexible bronchoscopy was unsuccessful due to the peripheral localization of the tumor. The pathological examinations of specimens obtained by bronchoscopy did not demonstrate the definitive diagnosis.

Because obstruction of the right upper bronchus could lead to recurrent post-obstructive pneumonia, we performed lobectomy of the right upper lung. Pathological examination identified a tumor protruding into the bronchial lumen and partially obstructing the bronchus, which contained mature cartilage

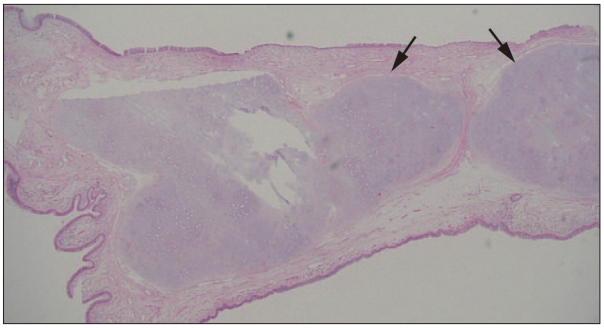


Figure 3. Histopathology of the tumor removed with bronchoscopy. Hematoxylin-eosin staining. Low-magnification view. Histopathology of the tumor removed with bronchoscopy shows cartilage component (arrow).

and fat tissue in the deeper layer, and was finally diagnosed as endobronchial chondroid hamartoma (Figure 4A and 4B). Several peripheral bronchi were filled with mucus and infiltrated with inflammatory cells, suggesting recurrent bronchitis or localized pneumonia. There was no evidence of recurrence of the tumor or post-obstructive pneumonia during the 6-month post-operative period.

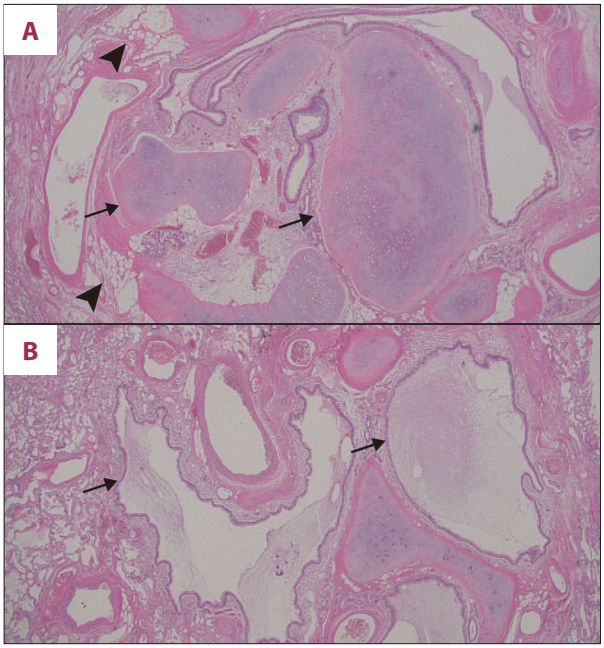


Figure 4. Histopathology of the surgically-resected specimen. The endobronchial tumor contained calcified cartilage (arrow) and fat tissues (arrowhead) (**A**). The peripheral bronchi (arrow) exhibited mucus collection and inflammatory cell infiltration (**B**). Hematoxylin-eosin staining. High-magnification view.

Discussion

Post-obstructive pneumonia is difficult to treat with antibiotics alone because of poor drainage, and can lead to recurrent hospitalization, increased medical costs, and poor outcomes [1]. Actual incidence and cause of obstructive pneumonia has not been well elucidated; however, lung cancer has been considered as the most common cause of post-obstructive pneumonia, occurring in approximately 20% of the patients with proximal airway obstruction [1]. Benign diseases (e.g., foreign bodies, benign tumors, granulomatous diseases such as tuberculosis, or broncholithiasis) can obstruct the airways and induce post-obstructive pneumonia [1–5]. Jhun et al. reported that 31% of the patients with benign endobronchial tumors exhibited obstructive pneumonia on chest CT scan [3].

Hamartomas occurs anywhere in the lung [6], but are rarely identified as an endobronchial lesion; in a series of 215 pulmonary hamartomas, only 1.4% were located endobronchially [6]. Although most patients with parenchymal hamartoma are asymptomatic [6], it is important to consider the possibility of endobronchial hamartoma as a cause of post-obstructive pneumonia for 2 reasons. Firstly, hamartoma is a relatively common pathology among endobronchial benign tumors. In a review of 185 benign tracheobronchial tumors, 45 hamartomas and 15 hamartochondromas were reported [7]. Another case series from Korea reported that hamartomas accounts for 44% (24/55 cases) of benign endobronchial tumors, and two-thirds of these were chondroid hamartomas [3]. Secondly, many endobronchial hamartomas are accompanied by recurrent pulmonary infection. In the largest case series, Cosio et al. [8] reported 43 cases of endobronchial hamartoma, most diagnosed during examination of recurrent respiratory infection (37%) or bloody sputum (32%) [8]. In a Korean study that reviewed 24 cases of endobronchial hamartoma, 10 cases (42%) showed obstructive pneumonia [3].

Recent reports demonstrate successful endobronchial resection of endobronchial hamartomas using laser or snare, without recurrence or regrowth of the tumor [9–11]. Several reports demonstrated successful endobronchial resection of endobronchial hamartomas using laser or electrocautery snare without recurrence or regrowth of the tumor [9–11]. Recently, other methods such as cryotherapy [12], argon plasma coagulation (APC) [13], or combination of different methods [13] have been reported.

Because hamartomas rarely recur or transform into malignancy [6–11,14], a minimally invasive procedure should be selected. In the present case, however, it was impossible to remove it endobronchially because we could not identify its stoke, which separates normal tissue from a tumor. Surgical approach is still an effective therapy for an intra-bronchial tumor that is difficult diagnose or remove by bronchoscopic approach [15,16],

especially when the tumor causes irreversible lung damage and/or recurrent infections at the distal site of obstruction, as in the present case.

Conclusions

Clinicians should consider the possibility of an intra-bronchial benign tumor such as hamartoma in the differential diagnosis of post-obstructive pneumonia. Bronchoscopic or surgical intervention to a remove intra-bronchial tumor is often required to avoid recurrent respiratory infections.

Conflict of interest statement

The authors have no potential conflict of interest to disclose.

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