

**Case
Report**

Bronchial Pleomorphic Adenoma Coexisting with Lung Cancer

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Pleomorphic adenoma usually occurs in the salivary glands but rarely in the trachea or bronchi. A 71-year-old man had abnormal shadows on a chest X-ray. Chest CT revealed one tumor in the right basal segment of the lung and another, in the left main bronchus. Bronchoscopic biopsy of the right tumor revealed well-differentiated squamous cell carcinoma. Right lower lobectomy and lymph node dissection were performed (pT2N0M0, stage IB). At the orifice of the left main bronchus, bronchoscopy identified a polypoid lesion nearly obstructing the airway. The lesion was resected with hot snare ablation. The histological examination revealed a mixture of epithelial and myxoid mesenchymal elements, characterized by ductal structures, squamous metaplasia, and cartilage tissue. The diagnosis was bronchial pleomorphic adenoma coexisting with squamous cell carcinoma of the lung.

Key words: pleomorphic adenoma, bronchoscopy, airway obstruction, lung cancer

Introduction

Although pleomorphic adenoma is the most common type of tumor in salivary glands, tracheobronchial pleomorphic adenoma is extremely rare.¹⁾ Here, we report a rare case of tracheobronchial pleomorphic adenoma coexisting with lung cancer. We performed endobronchial resection of the pleomorphic adenoma using an electro-surgical snare, and curative surgery for the lung cancer.

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Case Report

A 71-year-old man was referred to our department for progressive dyspnea. He had a 50-year history of smoking 2.5 packs per day and was receiving bronchodilator inhalation therapy for chronic obstructive pulmonary disease (COPD).

Blood chemistry showed no abnormalities except for elevations in SCC (6.0 ng/ml; normal range, 0–2 ng/ml) and Cyf1a (4.3 ng/ml; normal range, 0.1–3.3 ng/ml). Chest X-ray revealed a mass shadow in the right lower lung field and a low-radiolucent area at the orifice of the left main bronchus (**Fig. 1**). Chest CT revealed an irregularly margined tumor of 4.8 cm in diameter in the right basal segment and a tumor of 2.5 cm in diameter, in the left main bronchus (**Fig. 2**). Bronchoscopy identified a polypoid lesion nearly obstructing the airway at the orifice of the left main bronchus (**Fig. 3A**). Since the blood vessels on the tumor surface were engorged, the mediastinal side of the tumor pedicle was ablated using a hot snare, followed by tumorectomy. At the same time, another tumor was visible at B9 and B10 and was biopsied bronchoscopically. Bronchoscopic treatment resulted in the resolution

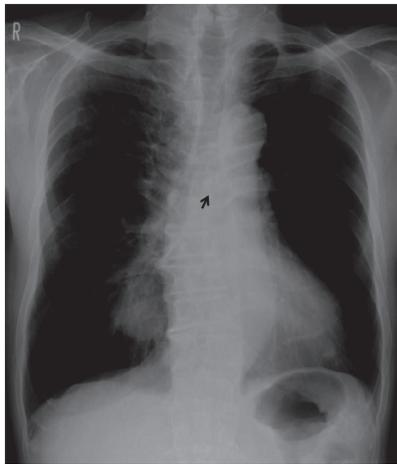


Fig. 1 Chest X-ray on referral to our department. A tumor was observed in the right lower lung field. A low-radiolucency area was also noted at the orifice of the left main bronchus (arrow).

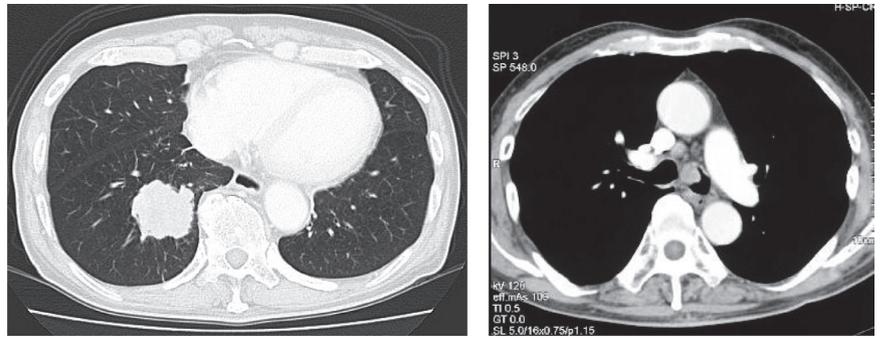


Fig. 2 Chest CT showing an irregularly margined tumor of 4.8 cm in diameter in the right basal segment and a tumor of 2.5 cm in diameter in the left main bronchus. CT, computed tomography

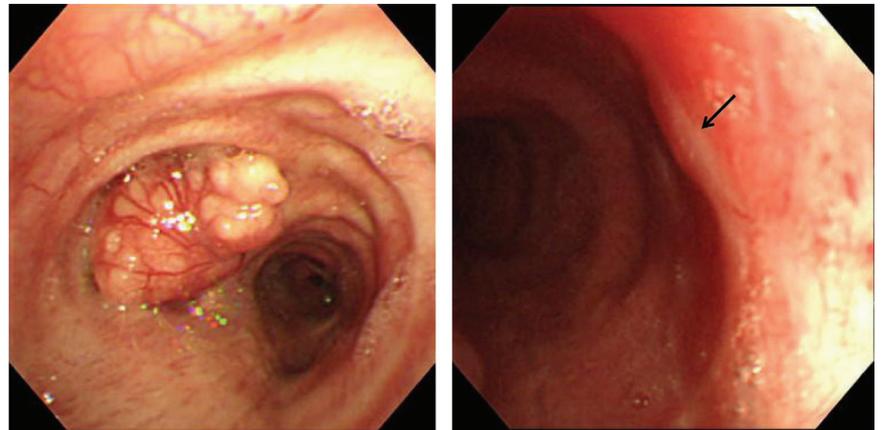


Fig. 3 Bronchoscopic findings of the tumor.

A: The tumor surface was lustrous and smooth, and had engorged branching vessels.

B: Bronchoscopy, performed 2 months after electrocautery snaring, showed no tumor regrowth, and only redness of the regenerated epithelium (arrow).

of the left main bronchial obstruction and marked improvement of respiratory distress, which the patient had thought was due to COPD. The tumor in the airway measured 2.5×2 cm and had a smooth surface and yellowish heterogeneous components (**Fig. 4A**). Histologically, the surface of the tumor was covered with bronchial epithelium and partly with squamous epithelium. Submucosal tumor consisted of ductal structures and myxoid stromal components (**Fig. 4B**). The tumor was composed of a mixture of epithelial and myxoid mesenchymal elements, and characterized by the presence of ductal structures with two layers (cuboidal luminal cells and myoepithelial cells), squamous metaplasia of ductal components, adipose tissue, cartilage, and hyaline fibers, suggesting pleomorphic adenoma (**Fig. 4C and 4D**). There was no evidence of malignancy in the epithelial component. The surgical margin was positive for the

tumor. On immunohistochemical staining, the inner and outer layers of ducts were positive for cytokeratin AE1/3 and α -smooth muscle actin, respectively (**Fig. 4E**). On the other hand, the tumor in the right basal segment suggested squamous cell carcinoma. Pulmonary function tests after electrocautery snaring were as follows: VC 3.05 L, %VC 96.5%, FEV_{1.0} 1.27 L, %FEV_{1.0} 58.3%, and FEV_{1.0}% 43.6%, which indicated grade II COPD. With cT2N0M0 lung cancer, the patient underwent right lower lobectomy with hilar and mediastinal lymph node dissection. Histologically, the tumor showed a marked tendency toward keratinization, suggesting well-differentiated squamous cell carcinoma. In the absence of lymph node metastasis, the tumor was diagnosed as squamous cell carcinoma, pT2N0M0, stage IB. The postoperative course of the patient was uneventful, and he has remained in good health since discharge. Postoperative bronchoscopy

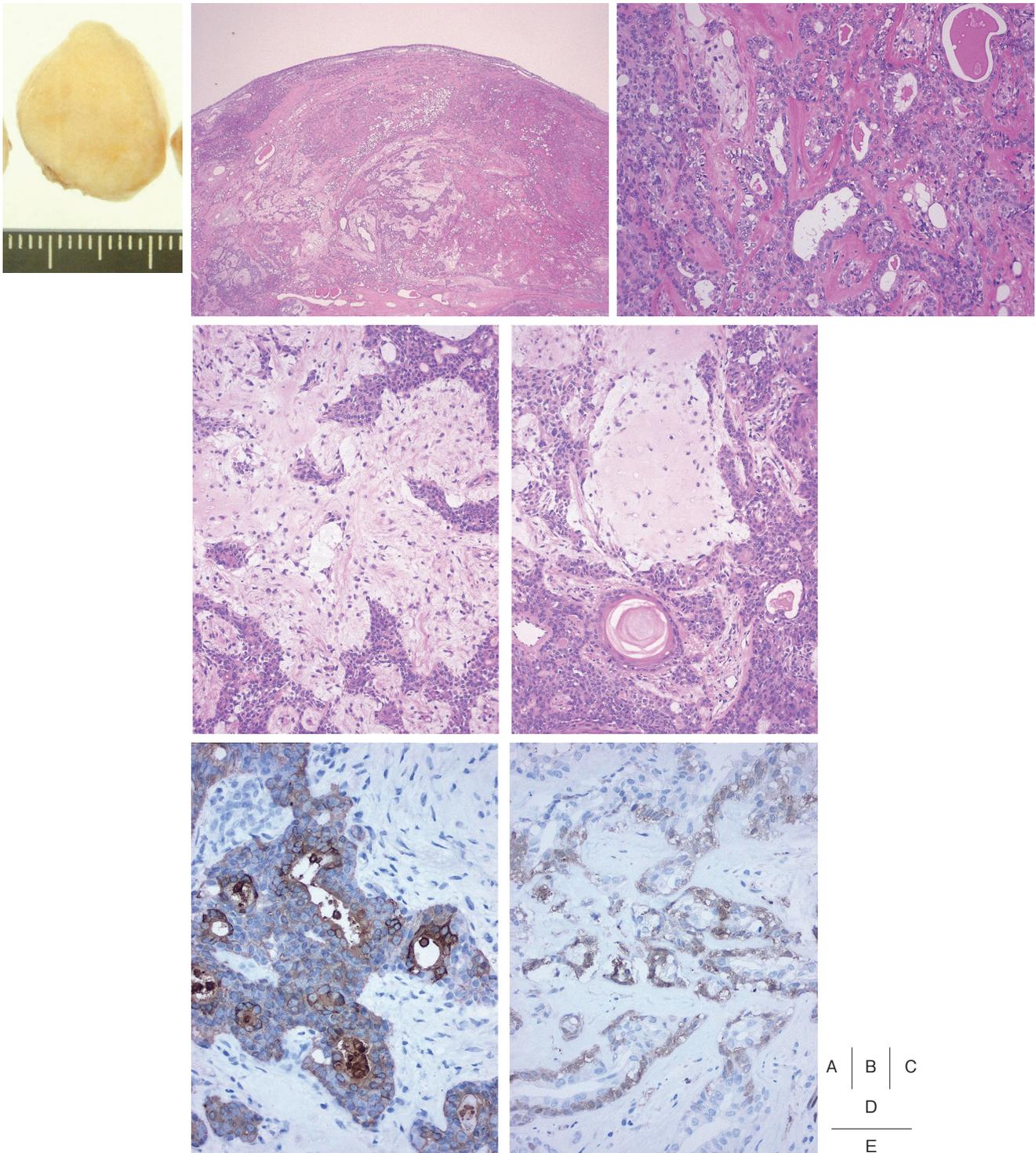


Fig. 4

A: Macroscopic appearance of the pleomorphic adenoma.

B: Submucosal tumor consisted of ductal structures and myxoid stromal components.

C: Ductal components consisted of eosinophilic cuboidal epithelium on the luminal side and myoepithelium with clear cytoplasm on the basal side.

D: Ductal components with partially solid nests scattered in the background of a myxoid stroma (**left**). A chondroid matrix and squamous epithelial metaplasia (**right**).

E: Immunostaining for cytokeratin AE1/3 (**left**) and α -smooth muscle actin (**right**).

showed only redness of the regenerated epithelium at the resected site, and no regrowth of the pleomorphic adenoma (**Fig. 3B**). The patient will continue to have follow-up examinations as an outpatient.

Discussion

Pleomorphic adenoma, which is the most common tumor of the major salivary glands,²⁾ occurs in the lung and typically arises from the tracheal and bronchial seromucous glands.³⁾ These tumors are extremely rare, and their incidence, etiology and prognosis are unknown.⁴⁾ Only 35 cases of tracheobronchial pleomorphic adenoma have been reported in the literature.⁵⁻⁷⁾ There is no clear gender or age predominance: the ages ranging between 8 and 74 years.^{1, 3, 6, 8)} Pleomorphic adenomas are benign neoplasms with mixed epithelial and mesenchymal components.⁹⁾ Patients presenting with obstructive symptoms usually have endobronchial lesions, generally in a main or secondary bronchus.¹⁾ The lesions usually develop on the posterolateral surface of the trachea or bronchus, where the glandular elements are most densely concentrated.⁸⁾ The present case was unusual in that the tumor arose from the anteromedial side of the left main bronchus. Occasionally, the tumor surface shows a prominent vascular pattern, as observed in this case.¹⁰⁾

The histological features are similar to those seen in salivary gland tumors except that the ducts are relatively sparse.^{1, 3)} Microscopy shows sheets, trabecular, or islands of epithelial and myoepithelial cells in a myxoid or chondroid matrix.^{1, 9)} Ducts consist of an outer layer of myoepithelial and an inner layer of epithelial cells.^{1, 9)}

Here, we reported a rare case of tracheobronchial pleomorphic adenoma coexisting with lung cancer. Although the initial examination suggested right lung cancer with metastasis to the left main bronchus, bronchoscopy confirmed the diagnosis of benign pleomorphic adenoma at the left main bronchus. Thus, the lung cancer could be treated by curative surgery.

Electrosurgical snare has been widely used and

accepted for the management of benign airway stenosis including a tracheobronchial pleomorphic adenoma.¹⁰⁾ In this patient, too, we successfully performed an endobronchial resection of the adenoma with an electrosurgical snare. In the absence of histological features of malignancy, such as necrosis and a high mitotic rate,¹⁾ the remaining pleomorphic adenoma is unlikely to grow into a malignant phenotype. We will follow the patient by employing periodic bronchoscopy to check for any regrowth of pleomorphic adenoma.

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