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Mucinous Bronchioloalveolar Carcinoma Associated with Bronchiectatic Cyst
– A Brief Case Report –

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Herein we report a rare case of mucinous bronchioloalveolar carcinoma (BAC) associated with a solitary bronchiectatic cyst in a 29-year-old man. The patient presented with hemoptysis and had a history of pulmonary tuberculosis. Chest radiographs and computed tomography revealed a well-circumscribed, thin-walled cavitary lesion in the right upper pulmonary lobe. Gross examination of a lobectomy specimen showed a bronchiectatic cavity and a fungus ball within it. There were also several ill-defined small gray-white nodules around the cyst, nodules that were mucinous BAC. On microscopy, they were composed of columnar tumor cells along the intact alveolar walls in a single layer.

Key Words : Adenocarcinoma, bronchiolo-alveolar; Bronchiectasis; Lung neoplasm

Bronchiectasis is defined as a permanent dilatation of the cartilaginous airways. Cystic (saccular) bronchiectasis is characterized by severe and irreversible ballooning of the peripheral bronchus. It resembles a cyst because the dilatation or expansion of the airway tends to be spherical. Such a region can show single or multiple cyst-like structures. Complications and sequelae of bronchiectasis are various, such as pulmonary hypertension, hemoptysis, and pneumonia. However, carcinoma associated with bronchiectasis is extremely rare. We present here a case of mucinous bronchioloalveolar carcinoma (BAC) associated with a solitary bronchiectatic cyst.

CASE REPORT

A 29-year-old man was admitted for evaluation of hemoptysis. Over a period of ten months before admission, the patient began to have small amount of hemoptysis intermittently. Recently, the amount of hemoptysis increased up to 60 mL. The physical examination was unremarkable except for a chronic ill-looking appearance. Twenty years before admission, the patient was diagnosed with pulmonary tuberculosis, which was cured with medication. He has also been taking medication for hyperthyroidism that was diagnosed 2 years ago. He had no history of tobacco or alcohol use. There was no family history of allergic or neoplastic disease. Initial laboratory tests showed no abnormalities except for increases in microsomal antibodies, thyroid stimulating hormone (TSH) receptor antibodies, and TSH level. Chest radiographs and computed tomography (CT) revealed a well-circumscribed, thin-walled cavitary lesion in the right upper pulmonary lobe (Fig. 1A). Lobectomy with total removal of the cavitary lesion was performed. Gross examination showed a

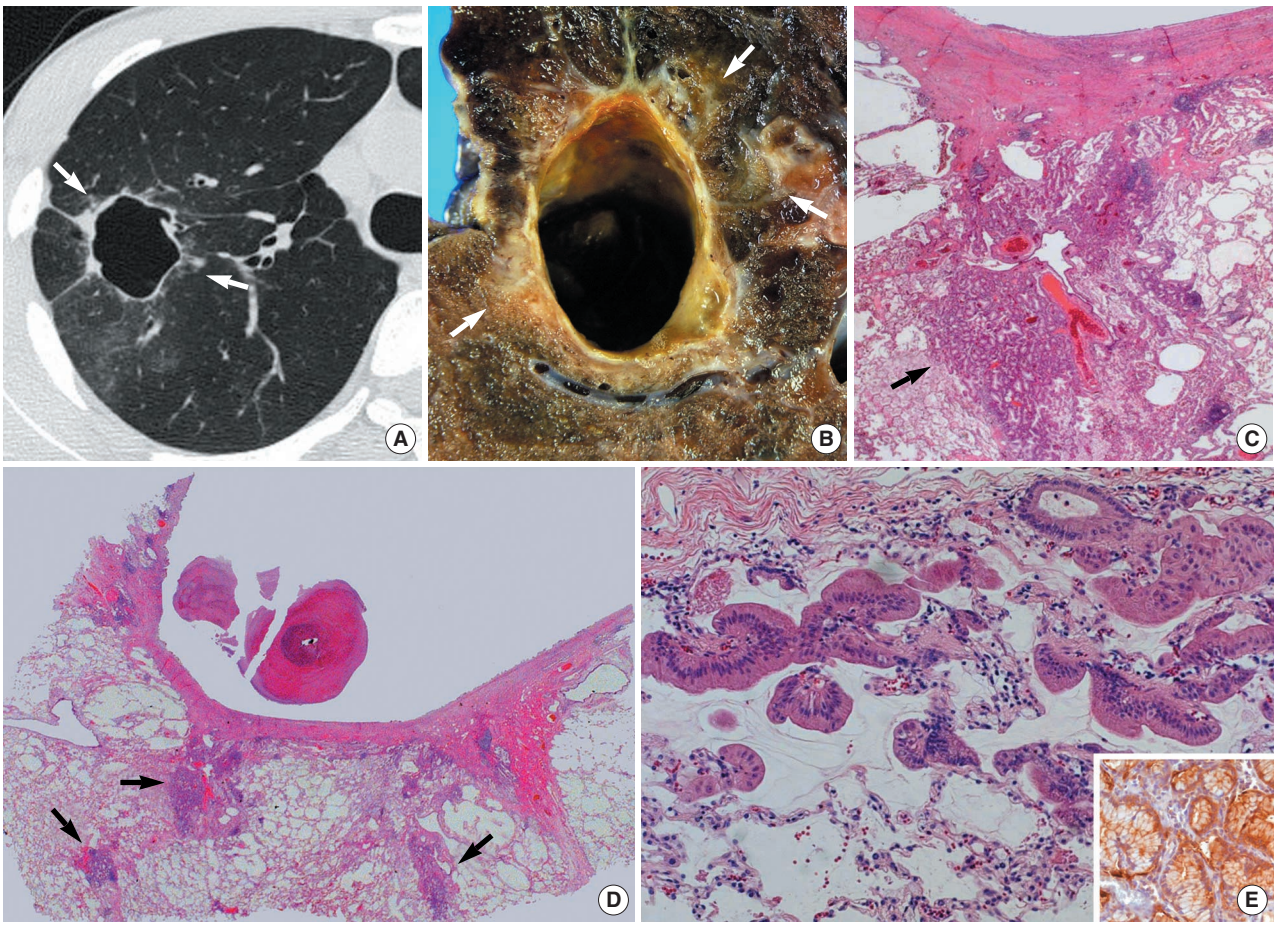

Fig. 1. (A) Computed tomography shows a well-demarcated cyst with ground-glass opacity around the lesion in the right upper lobe (arrows). (B) The cut section reveals a bronchiectatic cyst with a fungus ball. Also seen are several small gray-white nodules around the cyst (arrows). (C, D) Low magnification photomicrographs showing multiple discrete nodules of mucinous bronchioloalveolar carcinoma (arrows). (E) High magnification photomicrograph showing columnar tumor cells along the intact alveolar walls as a single layer. The tumor cells show immunoreactivity for carcinoembryonic antigen (inset).

bronchiectatic cyst with a fungus ball (Fig. 1B). Viewed with a microscope, the cyst was lined by fibrous tissue with pseudostratified ciliated columnar epithelium or erosion with loss of epithelium. Several small gray-white nodules up to 0.5 cm in diameter were identified around the cyst wall (Fig. 1B). The multiple discrete nodules were mucinous BAC composed of columnar tumor cells along the intact alveolar walls as a single layer (Fig. 1C-E). The tumor cells showed immunoreactivity for carcinoembryonic antigen (1 : 1,200, Dako, Glostrup, Denmark) (Fig. 1E, inset).

DISCUSSION

Causes of bronchiectasis include both congenital and acquired ones. Acquired bronchiectasis occurs more frequently, with one of the biggest causes being tuberculosis. Endobronchial tuberculosis commonly leads to bronchiectasis, either from bronchial stenosis or secondary traction from fibrosis. The cause of bronchiectasis in this patient may related to previous tuberculosis. And multifocal BACs were located around the bronchiectatic cyst. This suggests that occurrence of the tumor was related to metaplastic processes caused by recurrent irritation and inflammation.

There are a few reports of BAC making a pseudocavitary lesion.[1,2] One mechanism of pseudocavitation in BAC is that the low attenuating, mucin-containing air-space within the tumor produced the appearance of a central cavity on CT scans.[1] However, the cavitary lesion in this patient is different from the secondary pseudocavitary lesion seen in the a few reported cases of BAC. BAC in this patient developed secondarily in association with an acquired bronchiectatic cyst. There are a few reports of

pulmonary neoplasms associated with pre-existing cystic lesions. Dogan *et al.*[3] reported multiple neuroendocrine tumorlets associated with cystic bronchiectasis. Yoo *et al.*[4] described a mucinous adenocarcinoma arising in type 1 congenital cystic adenomatoid malformation (CCAM) of lung, and suggested that there was a predisposition of type 1 CCAMs toward malignancy. However, to our knowledge, there are no reports of multiple discrete mucinous BACs associated with cystic bronchiectasis.

Well-defined smooth, thin walled, air containing cavitary lesion in this patient suggested a benign lesion. This case clearly demonstrates the importance of being aware that such a benign looking cavitary lesion may be accompanied by a malignancy, even when the patient is young.

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