

Squamous Cell Carcinoma Presenting as a Solitary Growing Cyst in Lung: A Diagnostic Pitfall in Daily Clinical Practice

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Gradually growing cystic lesion of the lung is commonly encountered in daily clinical practice. Thin-walled cavitory lung cancer is a rare entity; however, it could be a pitfall in the diagnosis of such radiographically benign-looking lesions, especially without an obvious solid, nodular, or tumorous appearance in the lesion. We herein report a rare case of lung cancer successfully treated by surgical resection that appeared as a gradually growing cystic lesion mimicking benign emphysematous disease, with a review of the literature. A 68-year-old man with a 24-year history of hypothyroidism presented with an abnormal cystic shadow in the left lung on routine chest X-ray. Twelve months later, occasional bloody sputa had started and was gradually getting worse. The patient was then referred to our department for surgical intervention. He received clarithromycin by daily oral administration, and the bloody sputa soon disappeared. However, a malignancy was still suspected because the wall was slightly thickened unevenly in comparison with the previous chest X-ray and computed tomography findings. Thus we performed a left lower lobectomy followed by mediastinal dissection because a squamous cell carcinoma was diagnosed by intraoperative frozen section. The patient postoperatively received 4 courses of paclitaxel-carboplatin therapy. Twelve months after surgery, he survives without recurrence. (*Ann Thorac Cardiovasc Surg* 2009; 15: 174–177)

Key words: lung cancer, bulla, diagnosis, thin-wall, computed tomography

Introduction

Gradually growing thin-walled cavitory lesion of the lung is commonly encountered in daily clinical practice. Vari-

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ous conditions could be listed in the differential diagnosis. Most of these lesions are a benign disease, and emphysematous bullae are the most common. Thin-walled cavitory lung cancer is a rare entity, however, and could be a pitfall in the diagnosis of such radiographically benign-looking cystic lesions. Transbronchial biopsy or percutaneous needle biopsy is usually not scheduled because of a risk of pneumothorax by rupturing the lung cyst when obvious solid, nodular, or tumorous appearance is absent. Therefore it is sometimes problematic to make a decision on whether follow-up or surgical treatment should be chosen for such growing thin-walled cystic lesion in lung. We herein report a rare case with lung cancer successfully treated by surgical resection that

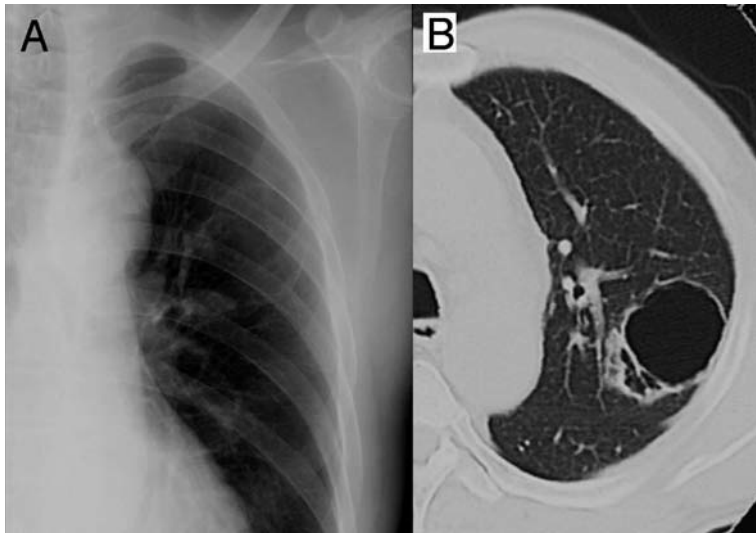


Fig. 1.
A: Chest X-ray shows a cystic lesion in the left middle lung field.
B: Chest computed tomography (CT) revealed the same thin-walled cystic lesion in the left upper lobe.

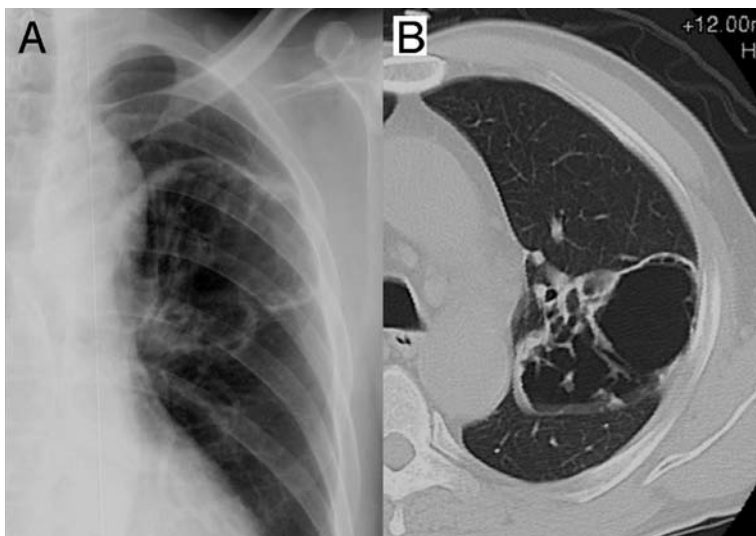


Fig. 2.
A: Chest X-ray taken 12 months later. An enlarged cystic lesion with slight partial thickness of the cystic wall is demonstrated.
B: Chest computed tomography (CT). Smooth-surfaced, multiseptal, and extended lumen of the cystic lesion is shown.

appeared as a gradually growing and very thin-walled cystic lesion mimicking benign emphysematous disease, with a review of the literature.

Case Report

A 68-year-old man, with a 24-year history of hypothyroidism after radioisotope therapy for Basedow's disease, presented abnormal cystic shadow in the left lung on routine chest X-ray and computed tomography (CT) in December 2004 at a local hospital (Fig. 1). Culture of the sputum demonstrated no evidence of tuberculosis or bacterial infection; it was positive only for *Candida albicans*. Antimycotic drugs had been administered for a week,

but the treatment was stopped by induced liver dysfunction. Thus the lung shadow had been followed by routine chest X-rays for 12 months after that. The size of the pulmonary cyst gradually increased; however, the wall had not been thickened, and the mass lesion had never been observed, even in the chest CT. Since December 2005, occasional bloody sputum has started, and it was gradually getting worse. In June 2006, the patient was referred to our department for surgery. He had smoked 2 packs of cigarettes a day since he was 20 years old (96 pack-years). Chest X-ray revealed a large cystic lesion approximately 9 cm in diameter in the middle lung field (Fig. 2A). The wall was very thin, like emphysematous bullae; however, it was nonuniform, and we found some parts of it to be

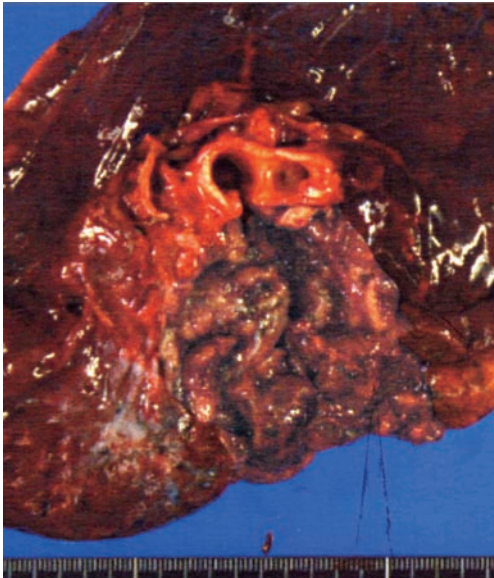


Fig. 3. Gross specimen of the cystic lesion.
The inner surface of the cystic lesion was totally lined with thin and fragile tumor tissue.

comparatively thick and uneven. No fluid content or nodular component was visualized inside. In chest CT, the cystic lesion showed no spiculations or indentations (Fig. 2B). The cyst lumen seemed smooth-surfaced and multi-septal; however, no tumorlike lesion could be demonstrated along the wall. As compared to the previous chest X-ray and CT (Fig. 1), the posterior half of the bullous lesion was enlarged, and part of the cyst wall was slightly thickened. Mediastinal lymph nodes did not swell in CT, and the patient was not febrile. The white blood cell count was 9,500/ μ L, and C-reactive protein was 2.0 mg/dL. Serum values of squamous cell carcinoma-associated antigen and carcinoembryonic antigen were within normal range. After daily oral administration of 400 mg of clarithromycin had been started, the bloody sputa disappeared. Bronchoscopic biopsy was not scheduled because of a risk of rupturing the cystic lesion. However, malignancy was still suspected because the wall was becoming slightly thickened irregularly as compared with the previous chest X-ray and CT findings. So after a month, a left lower lobectomy was carried out for refractory infectious bullae and possible malignancy. The cut surface of the specimen revealed a cystic lesion lined by very thin and fragile tumor tissue (Fig. 3). No debris or purulent content was found inside. Squamous cell carcinoma was diagnosed by intraoperative frozen section. Therefore mediastinal lymph node dissection was added. Pathology

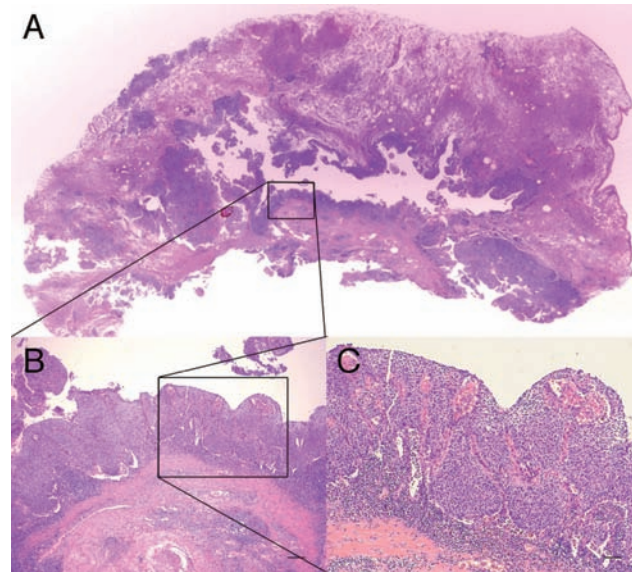


Fig. 4. Photomicrographs of the specimen. (hematoxylin-eosin stain)

- A: The cystic lumen was lined with tumor tissue in various uneven thicknesses.
- B: The tumor tissue presented mainly alongside the lumen at low power.
- C: This tumor was diagnosed as a poorly differentiated squamous cell carcinoma.

postoperatively confirmed that the cystic lumen was totally lined with tumor tissue in various uneven thicknesses (Fig. 4A). This lung cancer was diagnosed as poorly differentiated squamous cell carcinoma with lymph node metastasis in the pretracheal, lower paratracheal, and lobar nodes (Figs. 4B and 4C). Thus the patient was diagnosed as T2N2M0 stage IIIA disease, according to the International Union Against Cancer staging system, and postoperatively received paclitaxel-carboplatin therapy for 4 courses. Twelve months after surgery, the patient survives without recurrence.

Discussion

Generally, solitary thin-walled cavitory lesion in the lung was considered as benign disease. Lung cancer presenting as cystic lesion was first described by Anderson and Pierce in 1954.¹⁾ Ninety-five percent of solitary cavitory lesions in lung that showed a maximum wall thickness of more than 15 mm was malignant, though lesion with a maximum wall thickness of 4 mm or less would be benign.²⁾ Possible mechanisms forming cystic lung cancer have been discussed in the Japanese literature.³⁾ (1) A

check-valve mechanism of the responsible bronchus formed by inflammatory or neoplastic stenosis leads to cystic formation of the lesion. (2) Liquidized necrotic content is excreted through the responsible bronchus from ischemic centric necrosis of the tumor. (3) A tumor arising in the existing pulmonary emphysematous bullae is infiltrating alongside the cystic wall. (4) Cavitation is extending and the wall is thinned by an elastic retraction of surrounding pulmonary tissue.

Lung cancer arising in a bronchogenic cyst has been reported showing radiographic appearance similar to our case.⁴⁾ In the present case, however, concerning this patient who has been followed up by a single physician for more than two decades, cystic lesion has been confirmed to have newly arisen in the recent follow-up period. Therefore the possibility of lung cancer arising in the congenital bronchogenic cyst could be excluded. The occurrence of lung cancer in the wall of emphysematous bullae has been reported.⁵⁾ The cystic lesion was not located in the frequent site of emphysematous bullae, such as the apical area. Neither was prior bullous disease observed in the previous follow-up period. Thus the cancer was also believed not to arise in the emphysematous bullae. A combination of the check valve mechanism and elastic retraction by surrounding tissue because of the very soft and fragile consistency of the tumor tissue is considered as the most likely mechanism to form such cystic lesion in our case. Heavy smoking might have also caused an emphysematous change in the lung tissue, and decreased intensity and strength of the surrounding tissue might affect the formation and growth of the cystic lesion.

We had decided to perform surgical treatment in this case because of refractory infectious bullae and possible malignancy. The first reason was because the lesion was growing radiographically and accompanying bloody sputae that had recently started and were constantly continuing. Nevertheless, the bloody sputae had been dis-

appearing preoperatively by daily administration of clarithromycin. The bloody sputa were likely resulting from a secondary infection in the cystic lumen of the malignancy with a check-valve mechanism. The second reason was because although the internal surface of the cystic wall seemed generally smooth surfaced without any malignant findings such as spiculation, pleural indentation, or nodular or mass-like lesions, a part of the wall looked as if it were becoming slightly thickened in the follow-up CT. A growing bullous cystic lesion in an unusual anatomic site should be followed up carefully. A careful detection of the slight changes in clinical findings and periodical radiographies is important in decision making for surgical treatment of this clinically ambiguous and radiographically benign-looking malignancy.

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