Spontaneous Regression of a Bulla with the Development of Adenocarcinoma of the Lung

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Spontaneous regression of a bulla in the lung is rare. We describe a case of spontaneous regression associated with the development of adenocarcinoma of the lung in a 59-year-old male smoker. The bulla had begun to regress spontaneously at least six months before lung cancer was detected on a chest radiograph. He underwent left upper lobe lobectomy with mediastinal node dissection. The tumor arose within the bulla, extending along the bulla wall. He has been alive for more than eight years with no evidence of recurrence. This case suggests that spontaneous regression of a bulla should be recognized as one of the early radiographic signs of the development of lung cancer in patients with bullous lung disease.

Key words: Bullous lung disease, lung cancer, non-small cell

Introduction

Bullous disease of the lung is frequently noted in clinical practice. The association of lung cancer and bullous lung disease has been increasingly recognized (1). In most cases, the radiographic features of lung cancer associated with bullous disease are a tumor mass within or adjacent to the bulla or thickening of the bulla wall. However, spontaneous regression of a bulla with the development of lung cancer has been rarely reported. We herein report a case of a male smoker with bilateral apical bullae, in whom the bulla in the left lung regressed spontaneously with the development of lung cancer.

Case Report

A 59-year-old man was referred to the Aichi Hospital for evaluation of a mass in the left upper lung. A chest radiograph taken 10 years before presentation showed bilateral apical bullae, which had been essentially unchanged until one year before presentation (Fig. 1). A chest radiograph taken six months before presentation revealed a decrease in the size of the left apical bulla (Fig. 2). Further evaluation, however, was not done at that time. A chest radiograph taken just before presentation demonstrated the new development of a mass in the left upper lobe and further regression of the left bulla (Fig. 3). He had worked as a miner at a copper mine for 10 years until he developed left pleuritis at the age of 40, and he had a 30 pack-year history of smoking. On presentation, he was asymptomatic. Physical examination was unremarkable. Pulmonary function tests showed forced vital capacity and forced expiratory volume in one second of 112% and 72% of the predicted values, respectively. The serum carcinoembryonic antigen level was normal. Chest computed tomography (CT) scan showed a 4 x 4 cm mass with small multiple bullae in the left upper lung and a large bulla in the right lung. Flexible fiberoptic bronchoscopy was performed twice. However, cytology of bronchial brushings and washings was negative. He underwent left upper lobe lobectomy with mediastinal node dissection. The bulla decreased in size and was filled with fluid. The tumor arose within the bulla, extending along the bulla wall (Fig. 4). The histology of the tumor revealed poorly differentiated adenocarcinoma without nodal involvement (Fig. 5). He received adjuvant chemotherapy and has been alive for more than eight years with no evidence of recurrence.

Discussion

The incidence of bullous lung disease in patients with lung cancer has been reported at 2.5 to 3.9% (2-4). Stoloff and colleagues reported that the prevalence of lung cancer in those with bullous lung disease was 6.1 per 1000 and the relative risk of the development of lung cancer in this group was 32 times higher than in those without bullous lung disease (3). Venuta and colleagues reported that four (4.2%) of 95 patients with bullous lung disease who underwent surgical bullectomy had occult lung cancer (5). Lung cancer may develop at a
Saito and Okuno

Figure 1. A chest radiograph taken one year before presentation showing bilateral apical bullae.

Figure 2. A chest radiograph taken six months before presentation revealing regression of the left apical bulla.

Figure 3. A chest radiograph taken at presentation demonstrating a tumor mass in the left apex with further regression of the bulla.

Figure 4. Resected bulla with a tumor arising within the bulla and extending along the bull wall. The distal part of the tumor is necrotic (HE stain, ×4).

younger age in those with bullous disease (1). Based on these clinical findings, bullous lung disease is currently recognized as one of the major predictors of lung cancer.

In order to diagnose lung cancer associated with bullous lung disease at an early stage, it is important to recognize the specific radiographic features. Tsutsui and colleagues reviewed 26 cases of lung cancer associated with bullous lung disease and reported their findings of the radiographic features of lung cancer (4); these include nodular opacity within or adjacent to bullous disease, focal or diffuse thickening of the bulla wall, an increase or decrease in the size of the bulla, straightening of the hairline arcuate shadow of the bulla wall, fluid retention, and pneumothorax. However, regression of the bulla with the development of lung cancer was observed in only three out of the 26 patients in their series. Furthermore, the details were
Spontaneous regression of the bulla without any obvious cause has also been reported (6).

Because of increased risk of lung cancer in those with bullous lung disease, some have advocated regular screening by chest radiograph to detect lung cancer at an early stage in this group (2, 3). Clearly, further prospective study is necessary to define the role of regular radiographic screening in terms of early detection of lung cancer. In the meantime, regular screening of the chest radiograph, at least annually, should be considered with special attention to specific radiographic features of lung cancer associated with bullous disease. When the above-mentioned radiological features are found, further evaluation including sputum cytology, chest CT scan and bronchoscopic examination should be considered. However, preoperative diagnosis of lung cancer associated with bullous disease is often difficult. Even when histologic diagnosis is not made preoperatively, thoracotomy should be considered in doubtful cases. Furthermore, bullectomy is recommended in patients with bullous lung disease who have functional impairment because occult lung cancer has been reported in patients with bullous lung disease (5).

**References**