



## Case report

# A case of squamous cell lung cancer presented as a cystic lesion and recurrent pneumothoraces

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## ABSTRACT

We report a rare case of a 70-year-old male with recurrent pneumothoraces within one year treated with intermittent insertion of chest tube on each occasion. Diagnostic testing was notable for a cystic lesion in the left lung that was initially interpreted as bulla on chest x-ray and chest computed tomographic scan. Due to thickening and nodularity changes of the thin wall of the cystic lesion, the patient underwent left upper lobectomy. Pathology showed poorly differentiated squamous cell carcinoma of the cystic lesion wall. This case emphasizes the importance of monitoring pulmonary cystic lesions especially in patients with a history of smoking and emphysema.

## 1. Introduction

There is little data about the prevalence of lung cancers associated with cystic diseases. The majority of cystic lung cancers are related to adenocarcinoma, although other types of carcinoma have been reported [1]. Pneumothorax is a rare manifestation of primary lung cancer. The approximate rate of pneumothorax in primary lung cancer patients is 0.32% [2]. However, pneumothorax can be the first sign of lung cancer [3]. The etiology of pneumothorax and cyst formation in primary lung cancer remains unclear. Prompt investigation and appropriate follow up is crucial to avoid delays in cancer diagnosis and treatment.

### 1.1. Case presentation

A 70 year-old man presented to the pulmonary clinic for evaluation of progressive exertional dyspnea for the past three weeks. His medical history was significant for paroxysmal atrial fibrillation, recurrent left-sided pneumothoraces, and rheumatoid arthritis on hydroxychloroquine. Family history was unremarkable. Social history was significant for smoking history of 1.5 packs per day for 52 years. However, he quit smoking one year ago.

Over the past year, the patient developed repeated episodes of pneumothoraces that required multiple hospitalizations. He presented

for the first time to the emergency room with dyspnea and nonproductive cough for two weeks. A chest x-ray revealed a moderate-sized left pneumothorax (Fig. 1) that required tube thoracostomy placement, which resulted in the complete resolution of the pneumothorax.

Three months later, a follow-up computed tomographic scan of the chest demonstrated no evidence of pneumothorax. However, it showed mild emphysematous changes and a 3.6 × 1.9 cm cyst (Fig. 2), which was interpreted as a bulla of the left upper lobe.

Ten months later, the patient presented again to the emergency room with dyspnea and left-sided chest pain. A chest x-ray showed a large left pneumothorax with a rightward shift of the mediastinum concerning for tension pneumothorax. A large-bore 32 French chest tube was placed by general surgery. His condition gradually improved and subsequently, he was discharged home after one week of hospitalization. The chest x-ray revealed mild and stable small left apical pneumothorax on the day of the discharge.

Three weeks later, the patient was readmitted with the third episode of spontaneous left pneumothorax. Therefore, surgical consultation was obtained and thoracoscopy with mechanical and talc pleurodesis was performed. Surgical exploration visualized no significant blebs on the lung surface near the apex. The post-surgical course was uneventful, and the patient was discharged home on postoperative day two.

Upon presentation at the pulmonary office, the patient was afebrile,

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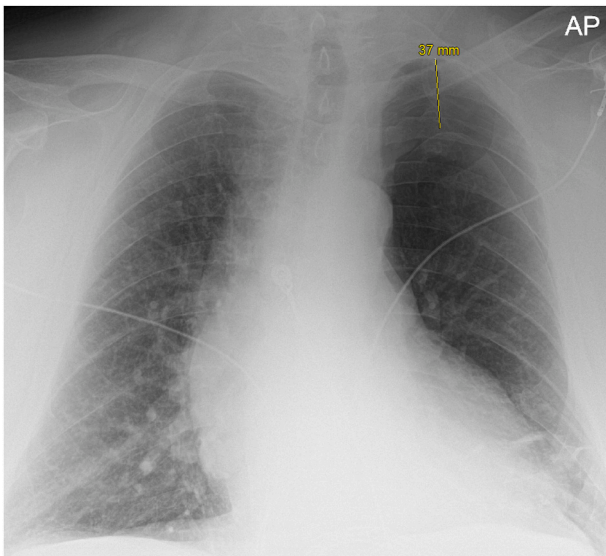


Fig. 1. Left sided pneumothorax.

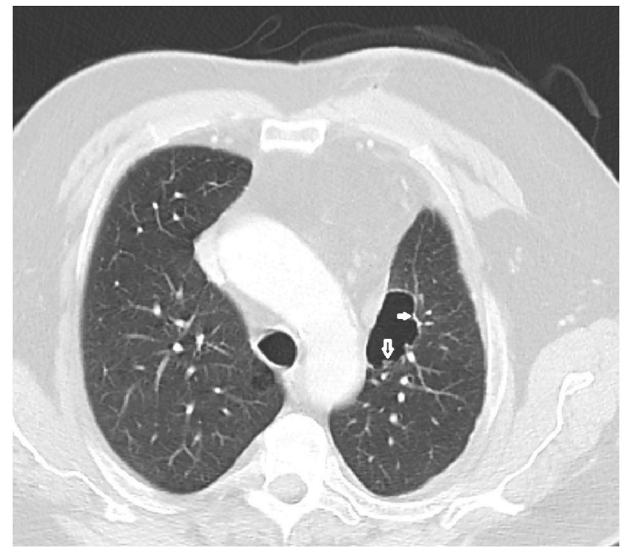


Fig. 3. CT chest: nodular thickening of the cyst wall. (white arrows).

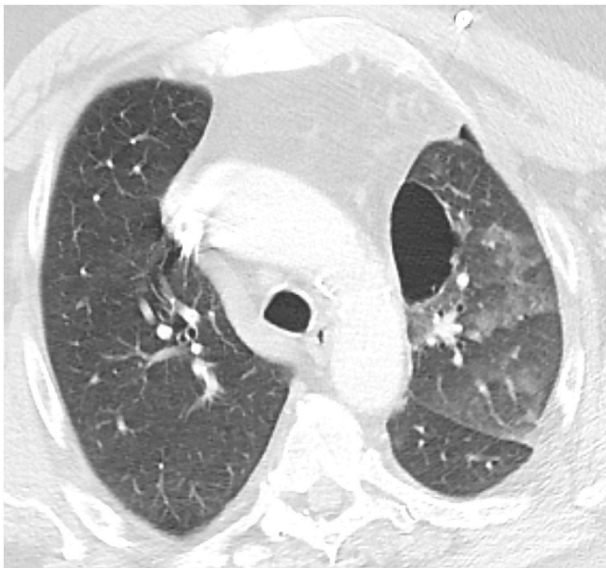


Fig. 2. CT chest: Left upper lobe cyst/Bulla with groundglass opacities.

his blood pressure was 126/73 mm Hg, and his pulse was 67 beats/min. The patient was not in respiratory distress with a respiratory rate of 16 breaths/min and his oxygen saturation was 92% on room air. Chest examination indicated decreased air entry over the left lung. There was no finger clubbing or palpable cervical lymph nodes. The patient was referred to the emergency room for suspected left-sided pneumothorax.

Complete blood count and liver and renal function tests were within normal limits. Chest radiograph showed a left-sided pneumothorax. CT of the chest showed a mild increase in the size of the cyst to  $3.9 \times 2.3$  cm with thickening and nodularity of the thin wall (Fig. 3). After consultation with thoracic surgical oncology, the patient underwent left video-assisted thoracoscopic surgery, left thoracotomy, lysis of adhesions and lingua-sparring left upper lobectomy with mediastinal lymph node dissection. Final pathology revealed poorly differentiated squamous cell carcinoma (Fig. 4A and B). The tumor involved 1 out of 11 lymph nodes. Therefore, the patient was staged as pT3 N1 M0, stage IIIA squamous cell carcinoma. Five percent of the tumor cells were positive for PD-L1 (membranous positivity). The patient was treated with adjuvant systemic therapy with cisplatin and gemcitabine.

## 1.2. Discussion

Pneumothorax is an uncommon presentation in lung cancer patients. One study reported that among patients with spontaneous pneumothorax, only 1.8% of patients had lung cancer [1]. In another report, pneumothorax was found in only 0.32% of primary lung cancer patients [2].

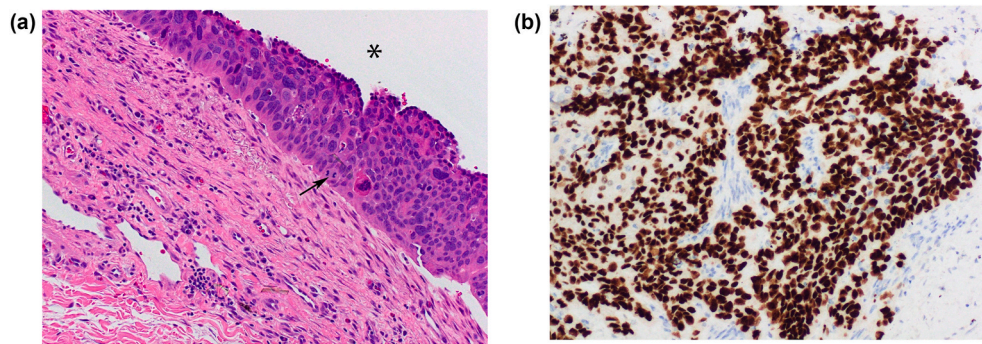
Cystic lesions associated with lung cancer have been reported in the literature and various etiologies have been proposed [3]. Ischemic necrosis, obstructive bronchiectasis, and air containing neoplastic space have been suggested as possible mechanisms to explain the formation of cystic lesions [4]. Also, a check-valve mechanism has been suggested as the etiology of the cystic dilatation and air trapping in the distal bronchioli, which subsequently can lead to an increase of the tension in the cyst and rupture [5]. This mechanism might explain the cause of pneumothorax as a complication of cystic lung cancer [6].

There is limited data about the prevalence of lung cancers associated with cystic diseases. Malignancy associated with cystic lesions was recognized as a cause of missed cancer in lung cancer screening trials [7, 8]. In an attempt to risk stratify cystic lung abnormalities, a classification system based on four morphologic types has been developed: Type I consist of nodule abutting the external aspect of a lung cyst. Type II is that of nodule arising from the cyst wall and projecting into the cystic space. Type III is that of cyst wall thickening. Type IV is that of a multicystic lesion that contains areas of soft-tissue attenuation [9]. However, none of these proposed classification systems proved to provide prognostic information. While adenocarcinoma is the predominant cell type in cystic related lung malignancy, squamous cell carcinoma has been reported as well [3].

We described a case of cystic squamous cell carcinoma presenting as recurrent pneumothoraces, which might be the initial presentation of lung cancer. Hence, this case underscores the importance of keeping a broad differential diagnosis and recognizing that pneumothorax can be an early complication of primary lung cancer.

## 2. Conclusion

Recurrent pneumothoraces may carry a serious underlying etiology and need to be investigated carefully. Lung cystic lesions should prompt a comprehensive evaluation and close interval follow-up. Characteristic CT finding of cystic lesions in the lungs with thickening or nodularity of the wall or increasing in size over time should raise concern for underlying malignancy and should be distinguished from a bulla in patients



**Fig. 4.** A): The cyst wall is lined by tumor cells with nuclear atypia, keratinization and abundant mitotic figures (black arrow). The cystic lumen is labeled with a black asterisk. (H&E, X10). (B): Immunohistochemical stain shows the cells are diffusely positive for p40. (  $\times 10$ ).

with underlying emphysema.

#### COI

This case report has not been published or presented elsewhere in part or in entirety and is not under consideration by another journal. All authors have seen and approved the case. I have read and understood your journal's policies, and there are no conflicts of interest to declare. Thank you for your consideration. I look forward to hearing from you.

#### Declaration of competing interest

No conflicts of interest.

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