

CASE REPORT

Treatment strategy for primary lung cancer in a lung highly compressed by giant emphysematous bullae: A case report

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Keywords

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Abstract

Lung cancer sometimes develops on the wall of a giant emphysematous bulla (GEB). Herein, we describe a rare case in which lung cancer developed in lung tissue compressed by GEBs. A 62-year-old man underwent a computed tomography (CT) scan that revealed two right GEBs. A tumor was suspected in the highly compressed right upper lobe. Since the right bronchus was significantly shifted toward the mediastinum, it was difficult to perform a bronchoscopy. We inserted thoracic drains into the GEBs, and a subsequent CT scan revealed re-expansion of the remaining right lung and a 3.3 cm tumor in the right upper lobe. The shift of the right bronchus was improved, and bronchoscopy was performed. The tumor was diagnosed as non-small cell lung cancer (NSCLC). Additionally, the GEBs were found to have originated from the right lower lobe. We performed a right upper lobectomy, mediastinal lymph node dissection, and bullectomy of the GEBs via video-assisted thoracoscopic surgery. In preoperative evaluation of a GEB, assessing re-expansion and lung lesions of the remaining lung is important, and intracavity drainage of a GEB may be useful.

Key points:

Significant findings of the study

- Cancer that develops in lung tissue highly compressed by a giant emphysematous bulla is difficult to diagnose.
- In the preoperative evaluation of a giant emphysematous bulla, assessing re-expansion and lung lesions of the remaining lung is important.

What this study adds

- After performing intracavity drainage of a giant emphysematous bulla, the remaining lung re-expands, and the bronchial shift improves; subsequently, bronchoscopy makes it possible to diagnose lung cancer in the remaining lung.

Introduction

A giant emphysematous bulla (GEB) is defined as a large bulla occupying at least one-third of a hemithorax.¹ Enlargement of a GEB often causes respiratory distress, and surgical treatment of GEBs improves respiratory symptoms.² There are some reports of lung cancer occurring on the cyst wall of a GEB.^{3–5} Herein, we report a rare surgical case of primary lung cancer that developed in lung tissue that was compressed by GEBs.

To our knowledge, this is the first report of a treatment strategy for primary lung cancer that developed in lung tissue highly compressed by GEBs.

Case report

A 62-year-old man with hypertension, rheumatoid arthritis, and a 43 pack-year smoking history experienced

exertional dyspnea. Spirometry indicated a vital capacity (VC) of 2.26 L (68.5%) and a forced expiratory volume in the first second (FEV1.0) of 1.16 L (43.3%). Transthoracic echocardiography revealed normal heart function. Laboratory tests revealed a tumor marker carcinoembryonic antigen level of 6.1 ng/mL (<5.0 ng/mL). Computed tomography (CT) revealed two GEBs occupying about three-quarters of the patient's right thorax, and the remaining lung was highly compressed (Fig 1a,b). A 3.1 × 1.4 cm irregular-shaped lung lesion was found in the right upper lobe. However, the right bronchus was significantly shifted toward the mediastinum (Fig 1c), and we expected difficulty performing bronchoscopy due to flexion of the right upper lobe bronchus. To allow re-expansion of the remaining lung and identify the localization of the GEBs, 16Fr and 8Fr thoracic drains were inserted into the GEBs under CT guidance. The thoracic under-water seal drains were not suctioned. The patient's respiratory distress improved, and he did not require oxygen therapy. A CT scan subsequently revealed re-expansion of the right lung and a 3.3 × 1.8 cm lung tumor with pleural indentation in the right upper lobe (Fig 1d,e). Since the shift of the right bronchus was improved (Fig 1f), bronchoscopy

was performed, and the tumor was diagnosed as non-small cell lung cancer (NSCLC) at clinical stage IB by bronchial brushing cytology. The GEBs were found to have originated from the right lower lobe. ¹⁸F-fluoro-2-deoxy-d-glucose positron emission tomography (FDG-PET)/CT was not performed.

Video-assisted thoracoscopic surgery (VATS) was performed for right upper lobe lung cancer and right lower GEBs six days after drainage. Surgical findings included extensive adhesions between the cyst wall and chest wall, and adhesiotomy was performed (Fig 2a). The tumor was found in the right upper lobe (Fig 2b), and no pleural dissemination was observed macroscopically. Right upper lobectomy, mediastinal lymph node dissection (ND2a-1), and bullectomy of the GEBs using autosuture devices were subsequently performed (Fig 2c,d). The operative time was 180 minutes, and the estimated blood loss was 20 mL. Postoperatively, pleurodesis was performed with a 50% glucose solution for a prolonged air leak, and the chest drain was removed on postoperative day 13. On histopathological examination, the tumor was diagnosed as invasive adenocarcinoma (Fig 3a,b). Pleural dissemination

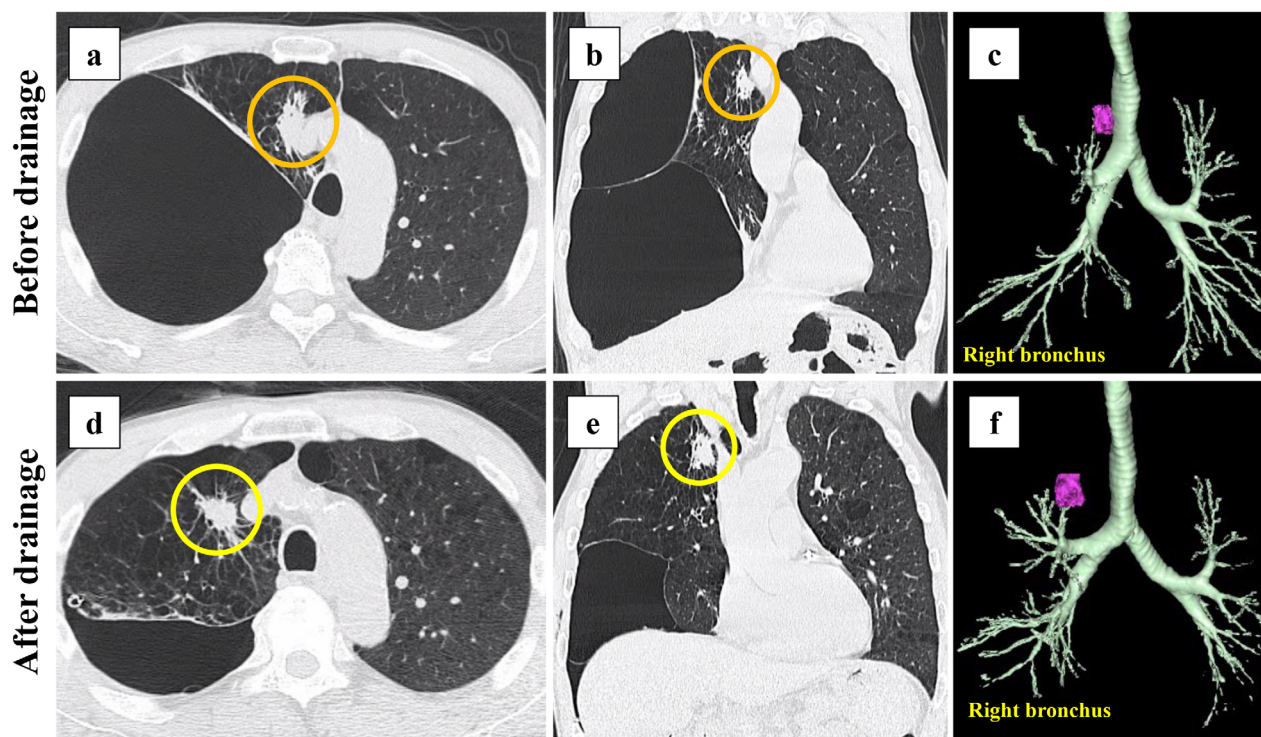


Figure 1 Imaging findings before and after intracavity drainage of the giant emphysematous bullae. (a, b) Computed tomography (CT) scan showing that giant emphysematous bullae occupy about three-quarters of the right thorax. The remaining right lung is highly compressed, and a 3.1 × 1.4 cm lung tumor is suspected in the right upper lobe (orange circles). (c) The right bronchus is significantly shifted toward the mediastinum (pink lesion: lung tumor). After intracavity drainage of the giant emphysematous bullae, a CT scan (d, e) shows that the right remaining lung is re-expanded, a 3.3 × 1.8 cm lung tumor with pleural indentation is revealed in the right upper lobe (yellow circles), and (f) the shift of the right bronchus is improved (pink lesion: lung tumor).

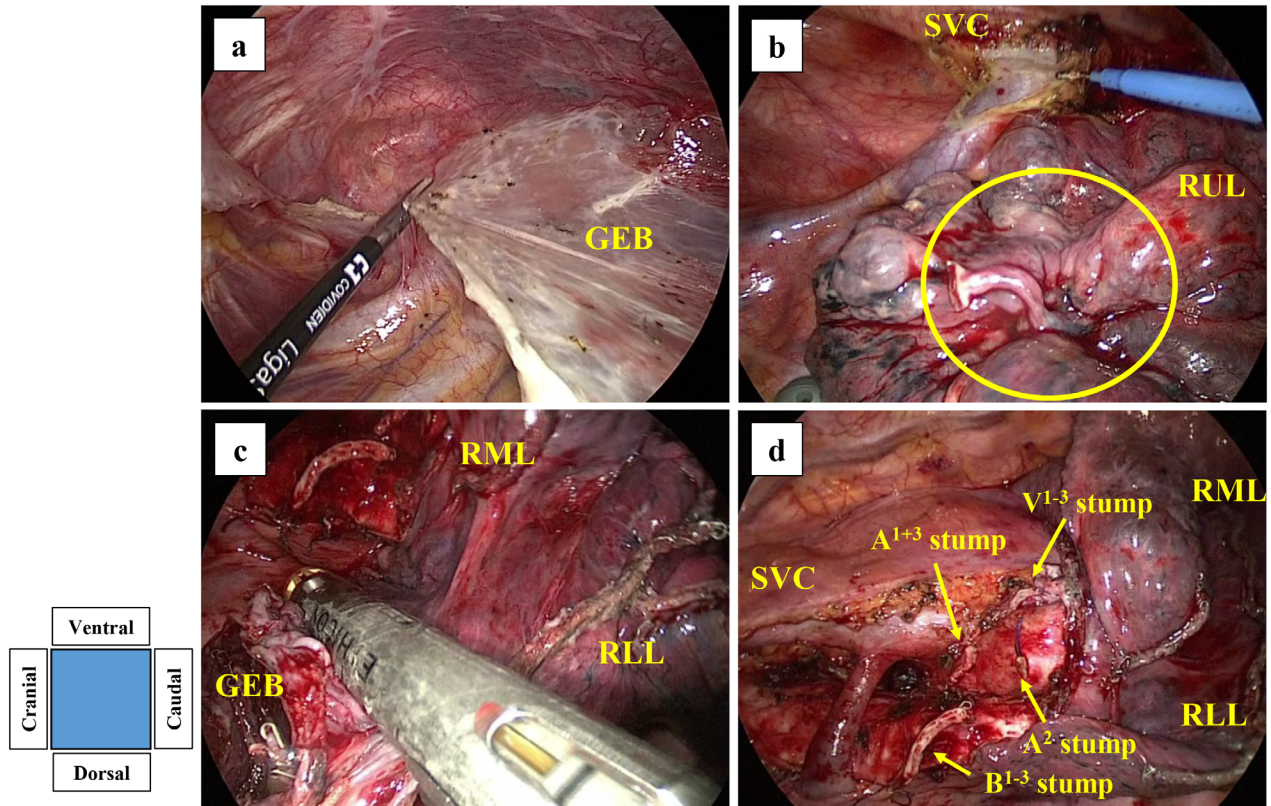


Figure 2 Surgical findings. (a) The cyst wall shows extensive adhesions to the chest wall. (b) Lung cancer is found in the right upper lobe (yellow circle), and no pleural dissemination is observed macroscopically. (c) Giant emphysematous bullae are resected using autosuture devices. (d) Surgical findings after right upper lobectomy, mediastinal lymph node dissection (ND2a-1), and bullectomy of the giant emphysematous bullae. GEB, giant emphysematous bulla; SVC, superior vena cava; RUL, right upper lobe; RML, right middle lobe; RLL, right lower lobe.

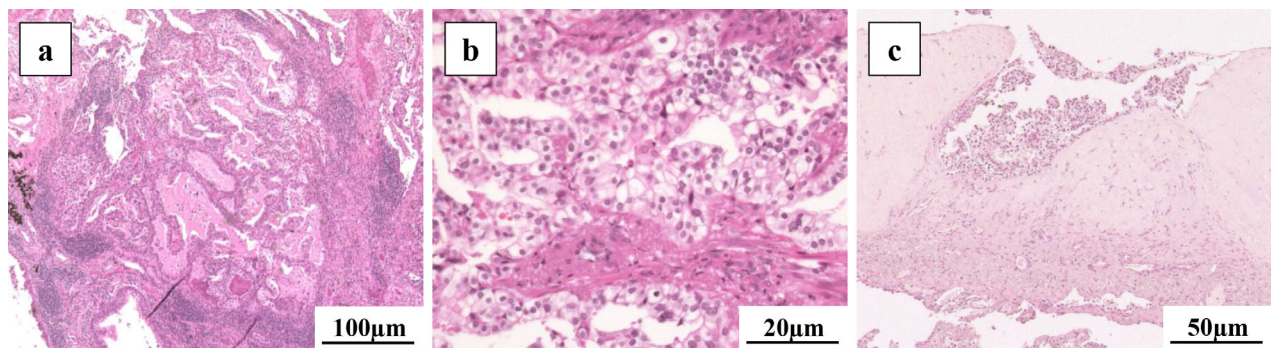


Figure 3 Histopathological findings. Hematoxylin and eosin staining showing that (a) lung adenocarcinoma has a papillary-predominant pattern and (b) some tumor cells contain clear cytoplasm. (c) Tumor cells containing clear cytoplasm are observed on the surface of the cyst wall of the giant emphysematous bulla; this lesion was diagnosed as pleural dissemination.

was found in part of the cyst wall of the GEB (Fig 3c), and the lung cancer was staged as IVA (T2aN0M1a). The patient received four courses of chemotherapy with cisplatin, pemetrexed, and bevacizumab followed by maintenance chemotherapy with pemetrexed and bevacizumab.

At six-month follow-up, no exertional dyspnea was observed. Spirometry indicated that VC was improved to 2.36 L (71.5%) and FEV1.0 was improved to 1.52 L (57.1%). CT showed that the remaining right lung was fully inflated, and no recurrence of lung cancer was observed (Fig 4a–c).



Figure 4 Computed tomography findings at five-month follow-up examination after surgery. (a, b, c) No new giant emphysematous bulla or recurrence of lung cancer is observed.

Discussion

In patients with a GEB occupying at least half of the hemithorax, surgery is recommended to improve lung function.² Since the remaining lung is often in a collapsed condition for a prolonged period, it is difficult to predict the degree of re-expansion preoperatively.⁶ In this case, thoracic drains were placed in the GEBs before surgery. This procedure, termed “intracavity drainage” was reported for the first time as a treatment for patients with GEB who cannot tolerate surgery.⁷ Thereafter, this procedure has also been reported as a preoperative treatment for GEB.^{8,9} The purpose of intracavity drainage is to allow re-expansion of the remaining lung and reveal any lung lesion, identify the localization of the GEB, and prevent perioperative complications such as tension bullae and re-expansion pulmonary edema. In this case, a lung tumor was revealed after re-expansion of the remaining lung, and the preoperative histological diagnosis of lung cancer by bronchoscopy became feasible. Additionally, identifying the localization of the GEBs facilitated the performance of VATS. Since re-expansion pulmonary edema and pneumothorax have been reported as complications of intracavity drainage of a GEB,^{9,10} careful observation of the patient’s respiratory condition is required after this procedure.

In conclusion, primary lung cancer that develops in lung tissue compressed by a GEB is rare. In the preoperative evaluation of a GEB, assessing re-expansion and lung lesions of the remaining lung is important, and intracavity drainage of a GEB may be useful.

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Disclosure

None of the authors has any potential conflicts of interest relevant to this report.

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