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CASE REPORT

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Giant bullous emphysema successfully treated with percutaneous drainage followed by resection: A case complicated by lung cancer diagnosed by intraoperative biopsy

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Abstract

We present a case of bilateral giant bullous emphysema (GBE) with rapidly progressive dyspnea. The dyspnea was thought to be due to tension bullae caused by the check valve mechanism in COVID-19 bronchitis. Multiple nodules were also detected on both sides of the lung. As the patient had poor pulmonary reserve for surgical bullectomy, we first performed percutaneous intracavitary drainage. Prior to this procedure, we placed a chest tube in the thoracic cavity to avoid tension pneumothorax. As a result, the patient's remaining lung expanded and respiratory status improved, allowing him to undergo surgical bullectomy. Intraoperatively, needle biopsy of the lung nodule was directly performed, which led to a diagnosis of adenocarcinoma. Despite multiple distant metastases, the patient's general condition improved postoperatively, and chemotherapy was successfully initiated.

KEYWORDS

giant bullous emphysema, lung cancer, percutaneous drainage, respiratory failure

INTRODUCTION

Giant bullous emphysema (GBE) is a rare disease that typically presents in middle-aged smokers. A giant bulla is defined as a bulla occupying at least one-third of the hemithorax and compressing normal surrounding parenchyma. The most common indications for surgical bullectomy include progressive bulla enlargement, dyspnea due to a giant bulla, secondary pneumothorax, and infection or bleeding in a bulla.[1] However, bullectomy under general anaesthesia might not be recommended in patients with poor pulmonary reserve. For such patients, intracavitary drainage provides less invasive treatment. Here, we report a case of bilateral GBE with multiple lung nodules, who was treated with percutaneous drainage followed by surgical bullectomy and was diagnosed with lung cancer by intraoperative needle biopsy.

CASE REPORT

A 57-year-old man presented to the emergency department with fever, dry cough, and rapidly progressive dyspnea. His modified Medical Research Council (mMRC) dyspnea scale score was 4. He was a current smoker with a smoking history of 39 pack-years. He had been diagnosed with bilateral GBE 12 years earlier (Figure 1), but because he was asymptomatic, he had been followed by only annual check-ups without detailed investigations.

On admission, he had an oxygen saturation of 96% while breathing 5 L/min oxygen with a face mask. His body temperature, blood pressure, heart rate, and respiratory rate were 38.0°C, 102/75 mmHg, 110 beats/min, and 33 breaths/min, respectively. Auscultation revealed decreased breath sounds on both sides of the chest. Compared to the chest x-ray taken 18 months earlier, the admission chest x-ray

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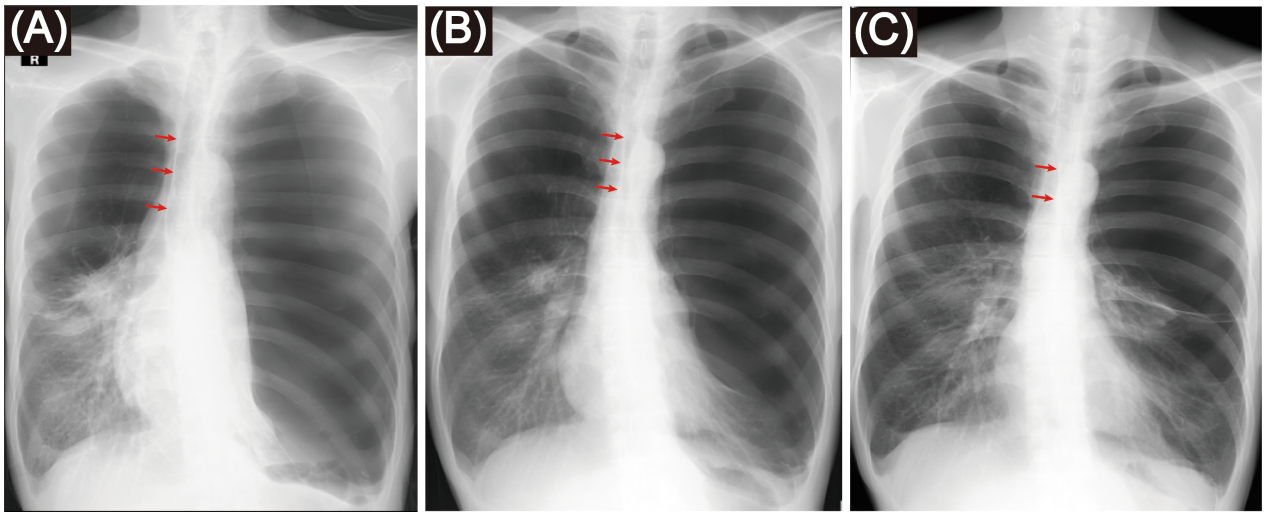

FIGURE 1 (A) Chest-x ray on admission showing bilateral giant bullous emphysema, mediastinal shift to the right, and a mass in the right hilum. (B) Chest x-ray taken 18 months earlier. (C) Chest x-ray taken 12 years earlier. Red arrows indicate anterior junction lines.

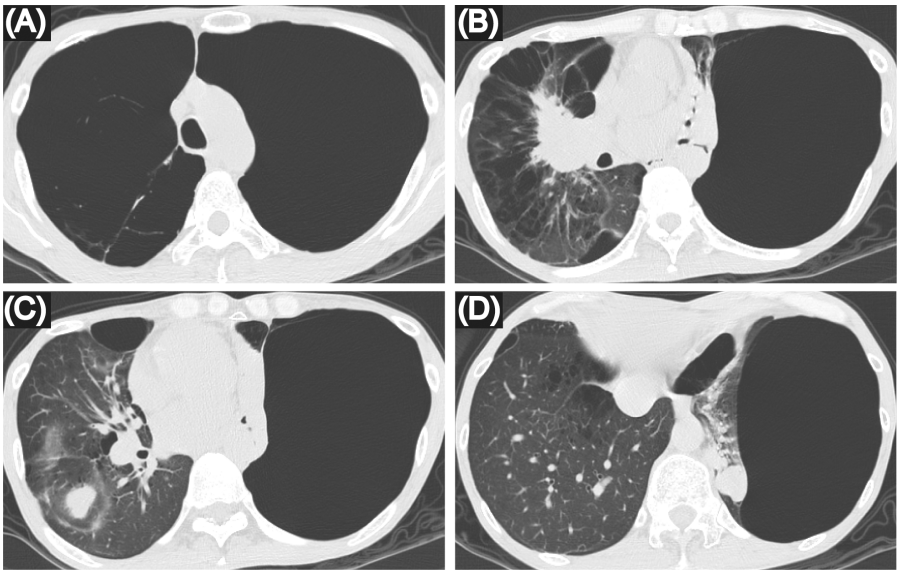

FIGURE 2 (A–D) Computed tomography scan on admission showed bilateral giant bullous emphysema. Three separate nodules were found in the right lower (B), right upper (C), and left lower (D) lobes.

showed that the left lower lobe and heart were significantly compressed to the right, and the anterior junction line was also displaced to the right, which indicated rapid enlargement of the left bulla (Figure 1). Computed tomography (CT) subsequently revealed that the left-sided bulla was monocystic, significantly compressing the lower lobe, and that the right upper lobe was severely destroyed, forming GBE with multiple septations (Figure 2). Three separate nodules were found in the right upper, right lower, and left lower lobes (Figure 2).

The patient was positive for SARS-CoV-2. His C-reactive protein level was 1.41 mg/dL. His pro-gastrin-releasing peptide level was 325 pg/mL (normal range <80.9 pg/mL) and carcinoembryonic antigen level was 5.3 ng/mL (normal range <5.0 ng/mL). Serum alpha-1-antitrypsin level was normal (225 mg/dL).

Assuming that the rapidly progressive dyspnea was due to tension bullae caused by the check valve mechanism in COVID-19 bronchitis, he was initially treated with remdesivir and nasal high-flow oxygen therapy ($FiO_2 = 40\%$). But his respiratory failure persisted. Because he had been asymptomatic prior to this admission, surgical intervention was considered possible if the positive pressure in the bullae was relieved and the respiratory status improved. Therefore, we decided to perform intracavitary drainage to relieve the tension in the left bulla. Prior to the drainage, we placed a chest tube in the left thoracic cavity to avoid tension pneumothorax. Under CT guidance, we inserted a cannula into

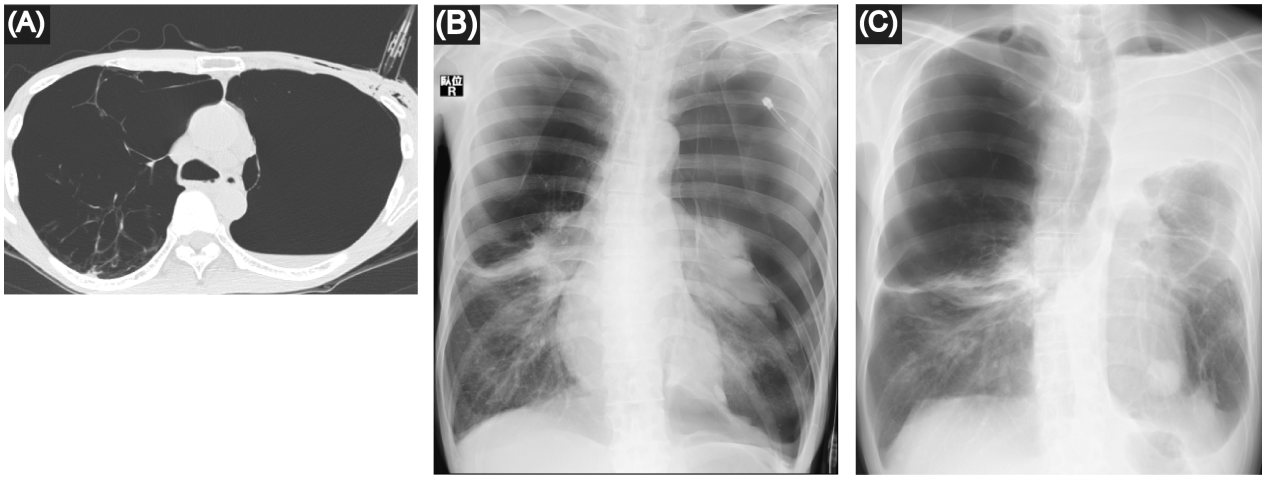

FIGURE 3 (A) A computed tomography scan showing intracavitary drainage with a cannula. (B) Chest x-ray after the intracavitary drainage. (C) Chest x-ray just before chemotherapy.

the left bulla and drained 2000 mL of air (Figure 3A). Immediately after drainage, his respiratory status improved rapidly. The left lower lobe expanded with mild pneumothorax (Figure 3B). A slight air leak was observed from the chest tube. However, this kept the pressure in both the bulla and the thoracic cavity close to atmospheric pressure. By day 14 after this procedure, his mMRC scale score improved to 3 while breathing 1 L/min oxygen. He was subsequently able to undergo left-sided bullectomy. Intraoperatively, needle biopsy of the left lower lobe nodule was directly performed. Histopathological evaluation indicated a diagnosis of adenocarcinoma. The patient recovered uneventfully and was discharged 16 days after the surgery. His mMRC scale score on discharge was 2 on room air. Pulmonary function tests on discharge indicated: forced vital capacity of 1.76 L (46.1% of predicted); and forced expiratory volume in 1 s of 1.26 L (38.2% of predicted). Although the patient had multiple distant metastases, he was in good condition and was started on chemotherapy 57 days after the surgery. His chest x-ray just before chemotherapy is shown in Figure 3C.

DISCUSSION

Surgical bullectomy has generally been advocated as the best management option for GBE with respiratory symptoms in fit patients. However, in this case, we first performed intracavitary drainage because the patient was too critically ill to undergo surgery. Intracavitary drainage for GBE along with pleurodeses was originally described by Head and Avery.[2] Recently, non-invasive intracavitary drainage followed by bullae reduction procedures with endobronchial occlusion, etc. have been adopted.[3,4] In the present case, both pleurodesis and bullae reduction procedures were not performed because we thought that surgical bullectomy would be possible after intracavitary drainage. As long as a chest tube is placed in the thoracic cavity, as in this case, intracavitary drainage can be performed relatively safely.

The incidence of lung cancer in patients with bullous emphysema is reportedly very high for the following reasons:[5] smoking is a common risk factor for both diseases, carcinogens tend to accumulate in bullae, and carcinomas are more likely to develop from scars of bulla walls. However, it is very difficult to diagnose cancers using bronchoscopy in patients with GBE because the bronchi involved are severely compressed. In the present case, the diagnosis was made by intraoperative needle biopsy. To the best of our knowledge, this is the first case of GBE with lung nodules that was treated with percutaneous drainage followed by bullectomy, and was successfully diagnosed with lung cancer by intraoperative needle biopsy.

AUTHOR CONTRIBUTIONS

Masaki Kimura and Susumu Yoshida contributed to the surgical bullectomy. Sachie Hasegawa and Tohru Sakamoto contributed substantially to the writing of the manuscript. Mako Yokoyama, Toshihide Inui, Hiroaki Ishikawa, Hiroko Watanabe, Masaki Kimura, and Susumu Yoshida contributed substantially to critical review of the manuscript. All authors read and approved the final manuscript.

CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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