

Surgery for giant emphysematous bullae: case report and a short literature review

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Abstract: Giant bullous emphysema (GBE), known as ‘vanishing lung syndrome’, usually occurs in association with chronic obstructive pulmonary disease (COPD). In this report, we describe a patient with giant emphysematous bulla occupying approximately 95% of the right hemithorax, who had history of repeated attacks of acute exacerbation of COPD. About 95% of right pulmonary parenchyma was removed. Delightfully, the 5% of residual lung compressed for 4 years gradually inflated, and occupied the whole hemithorax. Preoperatively he was functionally and clinically severely disabled while improved markedly after bullectomy.

Keywords: Giant pulmonary bulla; video-assisted thoracoscopic surgery; preoperative evaluation; lung function

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Introduction

Giant bullous emphysema (GBE), referred to as vanishing lung syndrome as a clinical syndrome, was first described by Burke in 1937 (1). Fifty years after that, Roberts *et al.* established the radiographic criteria for this syndrome as the presence of giant bullae in one or both upper lobes occupying at least one-third of the hemithorax and compressing the normal surrounding parenchyma (2). Chronic obstructive pulmonary disease (COPD)-related emphysematous bullae are the most common type lead to GBE (3). With progression of COPD, the obstruction increases in severity and eventually becomes irreversible. When the giant bulla occupies the entire hemithorax, and the remaining lung have been collapsed for a long period, it is difficult to predict the postoperative outcome of a bullectomy, and greatly increases the risks of surgery. Surgical bullectomy is a valid treatment option for patients with GBE (4), whereas bullectomy with end-stage COPD have very rarely been reported. We present a case of a patient with severely impaired lung function underwent successful bullectomy and is currently without residual symptoms.

Case report

A 59-year-old Chinese man, a nonsmoker presented in February 2012 with increasing cough and exertional dyspnea for 7 days, has suffered from repeated attacks of COPD over the past 4 years.

On admission, there was severe orthopnea and he was too breathless to leave the house (MRC grade 4) (5). Oxygen saturation was 93 percent while he was breathing ambient air. Arterial blood gas analysis revealed that his PaO₂ was low at 67 mmHg, with PaCO₂ 47 mmHg. Chest computed tomography (CT) (*Figure 1*) revealed the giant bulla had occupied approximately 95% of his right hemithorax, with only a small volume of ventilated lung in the lower lung field. We were not able to measure the ventilatory function, because the severity of his dyspnea prohibited him from holding his breath long enough. However, a previous test on March 7, 2011 showed: forced expiratory volume in one second (FEV₁) 0.93 L (31.30% predicted); forced vital capacity (FVC) 1.81 L; FEV₁/FVC 51.27%; and maximum ventilatory volume (MVV) 34.75 L (30.80% predicted). We also used the physical functioning domain of the Medical Outcomes study 36-Item Short-Form Health

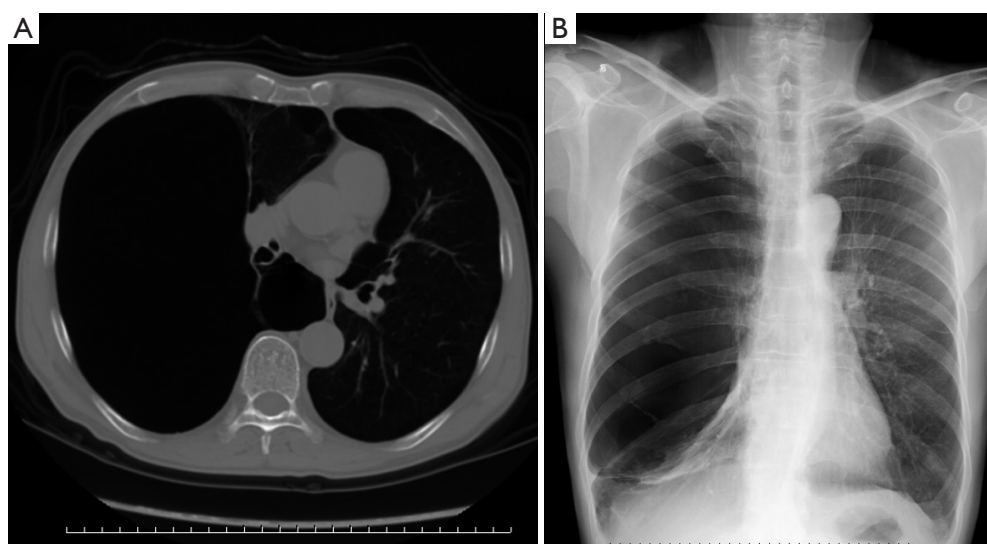


Figure 1 Chest computed tomography (A) and radiograph (B) revealed bilateral giant emphysematous changes. In particular, the giant emphysematous bulla occupied approximately 95% of his right hemithorax, with only a small volume of ventilated lung in the lower lung field.

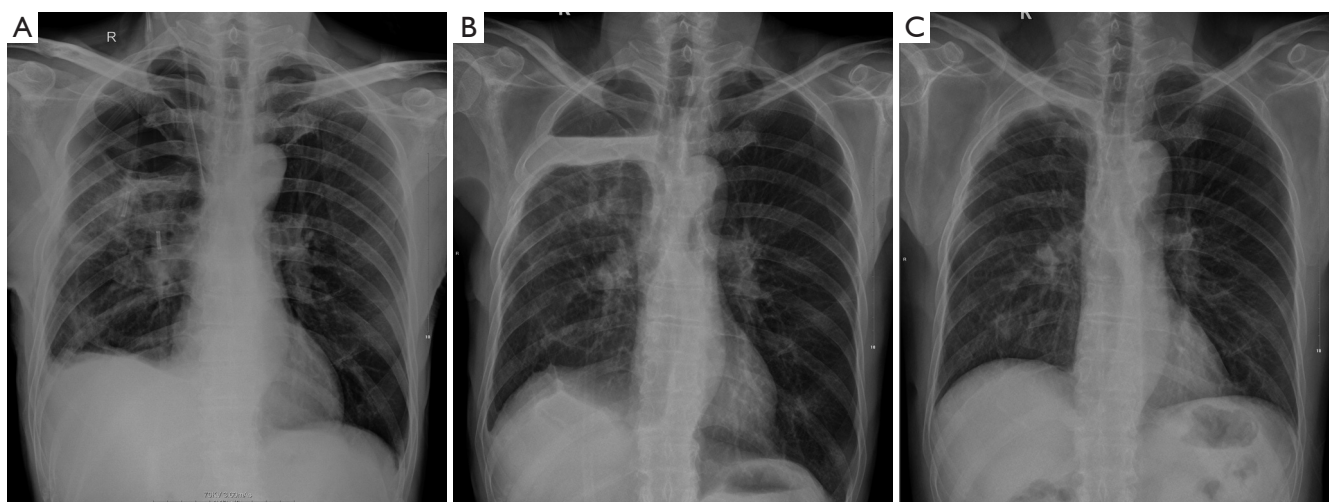


Figure 2 (A) The chest radiograph showed there was a residual cavity in the upper right thoracic cavity and the lung compressed about 30% when discharged; (B) the chest radiograph performed 2 months after surgery showed the right lung inflated well, but an air-fluid level was reserved; (C) chest radiography performed at the last follow-up, ten months after surgery.

Survey (6) to assessed the quality of life, which score was 0. The differential diagnosis was made against lung cancer. According to the CT scanning results, there's no obvious mass growth in the lung, so the diagnosis of cancer can be excluded.

Bullectomy was performed using the video-assisted thoracoscopic surgery approach. Intraoperatively, we saw multiple bullae in the upper, middle, and lower lobe. About 95% of right pulmonary parenchyma was removed. Soon

after the operation, the subjective symptoms improved. The chest radiograph performed showed there was a residual cavity in the upper right thoracic cavity and the lung compressed about 30% (Figure 2A). Although an air-fluid level was reserved, the chest radiograph (Figure 2B) performed after 2 months showed the lung inflated well suggesting that the patient made a good recovery. Meanwhile, a lung function test showed: FEV₁ 1.33 L (45.20% predicted); FVC 3.09 L; FEV₁/FVC 43.04%;

MVV 39.85 L (35.60% predicted). Arterial blood gas analysis revealed that his PaO₂ was at 85 mmHg while PaCO₂ was 39 mmHg. The lung expanded completely at the last follow-up (*Figure 2C*), ten months after surgery, when he did general activities without restriction. He got SF-36 physical functioning scale scores of 75. But the lung function had no further improvement.

Discussion

Today's surgeons focus not only on operative mortality but also on the quality of daily life for patients after surgery. In almost all studies performed on patients with bullous lung disease, dyspnea is the most common complaint (7-9), so dyspnea becomes an important indicator of the life quality. Palla *et al.* investigated patients with GBE during a 5-year-follow-up period, and concluded the degree of dyspnea decreased markedly soon after surgery and kept diminishing until the fourth year of follow-up (4). In this case, the man had history of COPD over the previous 4 years. With progression of this disease, the severity of airways obstruction deteriorated and eventually became irreversible. The patient could do normal activities without restriction (MRC grade 4) postoperatively. Six-minute walk distance increased significantly postpulmonary rehabilitation. What's more, health related quality of life as measured by the SF-36 Physical Functioning Scale showed marked improvement from a baseline preoperative score of 0 to a 10-month postoperative score of 75. From our point of view, for patients with giant pulmonary bullae occurring in association with end-edge COPD, the decrease in dyspnea sensation with exercise, are important to operate on.

The selections of patients suffering GBE for surgery have been widely reported (10-12). It is accepted that patients who have nonfunctioning bullae that compresses normal lung and occupies space in the chest cavity will benefit most from a surgical procedure (13), but there is no clear guidelines on the severely impaired lung function. Gunstensen *et al.* have suggested on the basis of their own surgical results that the more severe the preoperative impairment of FEV₁ the less likely the chance of marked improvement after operation (14). Nakahara *et al.* concluded that those patients with an FEV₁% less than 35% did not show as much benefit from bullectomy (15). In this case, pre- and postoperative FEV₁% value was 31.3% and 45.2%, respectively. And FEV₁ improved more than 0.4 of a liter. Obviously, there was significant change after surgery. So FEV₁% less than 35% should not be considered

a contraindication to bullectomy. Patients with very low preoperative FEV₁ still have striking improvements.

Though the case of the spontaneous resolution of a giant pulmonary bulla have been reported (16), surgical intervention was the only alternative in our patient, given that he had giant bullous disease-associated end-edge COPD, which responded poorly to conventional treatment. Previous literature stressed the importance of resecting as little nonfunctioning lung as possible-performing bullectomy (13,17-19). There has been controversy on operation range of diffuse bullae. In the preoperative assessment, MVV% value was far less than 55%, which indicated that the man could not tolerate complete lung excision (20). As stated above, we removed about 95% of right pulmonary parenchyma. Delightfully, the 5% of residual lung compressed for 4 years gradually inflated, and occupied the whole hemithorax ten months after surgery. A few lung tissues (about 5%) is still able to well re-expand to occupy whole hemithorax, which can be considered in the controversy on operation range of diffuse bullae. Pneumonectomy should be avoided. Although the patient had a prolonged air leak postoperatively, which was the most common complication for bullectomy (21-23), it did not prevent compressed lung tissue from expanding.

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