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Original Article

CT-guided drainage for patient with giant emphysematous bulla who was invalid 40 days after placement of EBV: a case report

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Abstract: Background: Bullectomy is preferred for the treatment of giant emphysematous bulla (GEB). For the surgically unfit candidates, bronchoscopic placement of one-way endobronchial valve (EBV) and intracavitary drainage of bulla have been considered as alternative strategies. Case presentation: In this report, we present a surgery unfit patient with giant emphysematous bulla in the left lower lobe who was not effective 40 days after EBV placement. As the patient was reluctant to EBV placement again, CT-guided bulla drainage was performed together with bronchial occlusion by bronchoscopy and instilling of autologous blood into the bulla. The patient had immediate and sustained symptomatic relief with significant improvement in lung function. Conclusion: CT-guided bulla drainage is a simple, minimally invasive treatment method for inoperable GEB patients.

Keywords: Giant emphysematous bulla, endobronchial valve, CT-guided drainage, intracavitary drainage

Introduction

Giant emphysematous bulla (GEB) is defined as a large bulla occupying at least one third of a hemithorax, which causes collapse of adjacent lung parenchyma and also impairs the function of the inspiratory muscles, especially the diaphragm [1]. To date, surgical bullectomy is the preferred option for GEB, and has been approved to improve symptoms, exercise tolerance and respiratory reserve [2, 3]. Despite surgical and anesthesiological technical progress, there are still many cases considered to be high risk for surgery [4]. For the patients unfit for surgery and/or those reluctant to the surgery, further studies are needed to develop appropriate treatment strategies. Among these strategies, endobronchial valve (EBV) placement and intracavitary drainage have been considered as alternative approaches [1, 5]. In the present report, we describe a simple, speedy and minimally invasive method using computed tomography (CT)-guided drainage to treat a GEB patient who was unfit for surgery and was not effective 40 days after EBV placement.

Case presentation

A 69-year-old male patient presented to our department due to chronic obstructive pulmonary disease (COPD) plus emphysema, which was manifested by obvious dyspnea, shortness of breath and poor exercise tolerance. The patient received regular administration of salmeterol-fluticasone (50/500 μg, b.i.d.), and tiotropium bromide (18 μg, q.d.) in the recent 12 months. Chest computed tomography scanning indicated a giant emphysematous bulla in left lower lung with a size of 17.5 × 9.1cm^2 (**Figure 1A**). Arterial blood gas parameters were pH 7.40, pressure of carbon dioxide ($PaCO_2$) 39.2 mmHg, partial pressure of oxygen (PaO_2) 89.2 mmHg, and fraction of inspired oxygen (FiO_2) 29.0%. The patient was considered as a poor surgical candidate as his pulmonary function was severely damaged. Thus, placement of EBV was performed after written informed consent was obtained. A Zephyr 4.0 EBV (Emphasys Medical, Inc. Redwood City, CA, USA) was placed in the lower lobe bronchi (B7+8, B9, B10) through a flexible bronchoscope (Olympus, Inc. Tokyo, Japan) (**Figure 1B**, **1C**). Prior to EBV

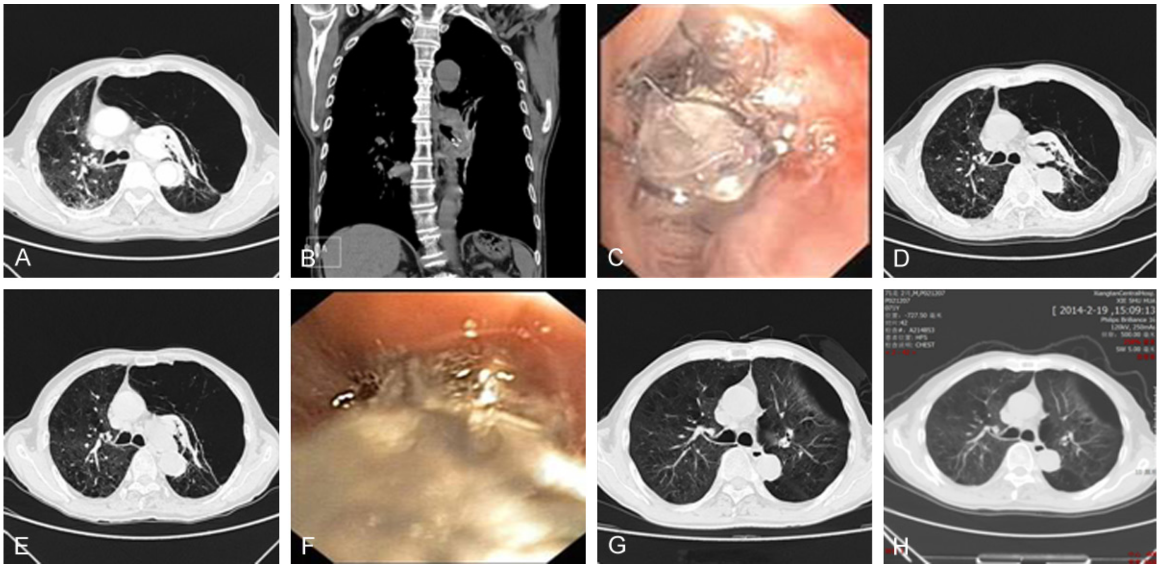

Figure 1. A: Chest CT showed a GBE occupying almost the left chest, together with compression atelectasis in adjacent pulmonary parenchyma. B and C: Chest CT and bronchoscopy showed that the Zephyr 4.0 EBV was positioned in the left lower lobe segmental bronchi; D and E: Chest CT showed partial volume reduction of the left lobe bullae with the mitigation of the mediastinal shift on d5 and d30 after EBV placement. F: Bronchoscopy displayed the ineffective EBV and abundant viscous secretion. G and H: Chest CT showed a marked volume reduction of the left lobe bullae with no mediastinal shift 7 months and 14 months after intracavitary drainage.

insertion, no collateral ventilation was noted between the targeted lobe in correspondence to the bulla and the neighboring lung lobes by Chartis system. The whole procedures were performed under intravenous anesthetic regimen using midazolam and fentanyl. The procedure lasted for 30 minutes with no unexpected events. Subjective improvement was noted in the dyspnea 1~7 days after the procedure, and obvious improvement of dyspnea was noticed on d19, whereas, no significant improvement was noted in the dyspnea on d30 compared with the baseline level. CT scanning was performed on d5 and d30 after the procedure, which revealed the volume of the GEB was significantly reduced, and the shifting of mediastinum was significantly relieved (**Figure 1D**, **1E**). The pre- and post-operative pulmonary function, 6-minute walk distance (6MWD) and St George's respiratory questionnaire (SGRQ) score were listed in **Table 1**.

After that, the shortness of breath was deteriorated, and excessive phlegm was generated. Bronchoscopy performed on d40 after the procedure revealed excessive white sticky secretions within the EBV and the targeted lobe bronchus. The EBV was then washed using normal saline, but it was still not effective (**Figure 1F**). Acute deterioration of the symptoms was reported by the patient three times within 4 months. On d127, the EBV was removed under the bronchoscope. The patient was reluctant to the placement of EBV due to financial reasons.

Twelve months after the procedure, the patient reported gradual deterioration of shortness of breath. He was too ill to perform pulmonary function test. Arterial blood gas values were pH 7.42, PaCO2 36.4 mmHg, PaO2 63.4 mmHg, FiO_2 21.0%, and a SGRQ score of 67. Thus, CT-guided GEB drainage was performed after informing the benefits and risks to the patient (the patient signed the informed consent). The site of puncture was determined under CT scan. A 16-gauge puncture needle was inserted into the bulla under local anesthesia. Afterwards, a guide wire was introduced into the bulla through the needle. A central venous catheter with a diameter of 1.7 mm was then placed in the bulla along the guide wire. Subsequently, a three-way connector attached to the proximal hub in order to aspirate air from the bulla. The pressure within the GEB was measured by a pressure gauge (Mallinckrodt Medical GmbH, Hennef, Germany). A syringe (50 ml) was used to aspirate the air via the three-way connector. After air aspiration (approximately 1,200 mL)

Table 1. Comparison of pre- and post-operative pulmonary function, 6-minute walk distance and St George's respiratory questionnaire score

	Before EBV	19 d after EBV	30 d after EBV	7 months after ID	14 months after ID
FEV1 (L)	0.77	1.04	0.92	1.64	1.68
FEV1 (%)	32	43	38	67	68.5
FVC (L)	1.61	2.02	1.82	3.03	3.26
RV (L)	4.20	4.19	4.34	2.72	2.66
TLC (L)	6.03	6.29	6.41	4.74	4.70
6MWT (M)	240	260	246	406.5	412
SGRQ	67	58	77	26	25

EBV: endobronchial valve; ID: intracavitary drainage; FEV1: forced expiratory volume in one second; FVC: forced vital capacity; RV: residual volume; TLC: total lung capacity; 6MWT: 6-minute walk distance; SGRQ: St George's respiratory questionnaire.

from the bulla, the shortness of breath was significantly alleviated. However, the shortness of breath was deteriorated with the continuation of the aspiration, which may be related with the circulatory instability caused by fast aspiration. Thus, the aspiration was terminated, and the pressure within the GEB was reduced from 22 cm H_2O to 10 cm H_2O. Immediately, CT scanning was performed, which showed a significant reduction in the volume of the bulla with no adverse events. Subsequently, a closed drainage bottle was connected to three-way connector for the constant drainage and the negative pressure drainage was performed with a pressure range of -6~-4 cm H_2O one day after the procedure. Autologous blood (25 ml) and thrombin (1000 U) were instilled into the bulla on d2 and d7, respectively. In addition, erythromycin lactobionate (0.5 g diluted in 20 ml 50% glucose) was injected in the targeted lobe bronchus in correspondence to the bulla by bronchoscope. On d20, intermittent drainage was performed instead of constant drainage. CT scanning was performed on d30, which revealed the volume of the GEB was remarkably reduced. On this basis, the drainage tube was removed. Chest CT scanning was performed in the 7th month and 14th month after the procedure, and the outcome was satisfactory (**Figure 1G**, **1H**). During the 14 months follow up, significant improvement was noticed in the postoperative pulmonary function, 6MWD and SGRQ score compared with the baseline levels (**Table 1**).

Discussion

Placement of EBV and intracavity drainage has been mainly used for the treatment of GEB in surgically unfit patients. In 2006, Noppen et al reported a poor surgical candidate underwent bronchoscopic placement of one-way endobronchial valves for treating chronic obstructive pulmonary disease (COPD) concurrent with GEB, and the GEB was almost complete disappeared within 1 month after the procedure [1]. Subsequently, the feasibility and efficacy of EBV placement was confirmed by a prospective analysis carried out in 9 surgically unfit patients with GBE. During the 6 months follow-up, a significant improvement of respiration and life quality was noted in these patients [6]. Although it is simple and reversible, it still faces postoperative complications. For example, development of transient pneumothorax was noticed in some patients after EBV insertion for volume reduction [7]. In addition, elective removal of valves was needed in some patients due to adverse events (e.g. pulmonary infection) [8]. In this case, the valve was removed as it was valid 40 days after placement. Furthermore, high cost of EBV is a great challenge for its application in developing countries.

Intracavity drainage, the technique involving insertion of drainage tube and filling of iodised talc in the pleural cavity and the bulla [5], has been reported to contribute to the the fibration of bulla wall, and prevention of pleural adhesions and bronchopleural fistula [9]. However, many patients were not tolerable to the wound and complications triggered by conventional Brompton technique as a Foley catheter (32 FG in size) was inserted into the cavity via a separate stab incision in the chest wall [5]. In a previous study, Macarthur et al reported 2 operative deaths (6.5%) and 1 (3.25%) treatment failure in the patients using Foley catheter for intracavity drainage [10]. Subsequently, a modified technique based on Brompton drainage using a mushroom catheter in combination with low pressure aspiration was developed, however, it still was invasive as thoracotomy was required [11].

To overcome the potential disadvantages of drainage using a thick catheter (e.g. thoracotomy and wound), Bhattacharyya performed bronchoscopic transbronchial decompression in a 50-year-old patient presented emphysematous bullae using a transbronchial aspiration needle, followed by instilling of autologous blood into the bulla before the withdraw of the needle to prevent re-expansion of the decompressed bulla. However, the procedure was not applicable to the peripheral GBE. In 2004, Takizawa developed CT-guided drainage for two poor surgical candidates with large pulmonary bullae. Meanwhile, OK-432 and minocycline (as adhesive agents) were injected into the bulla to promote fibrosis and shrinkage of the bullae, and avoid the bronchopleural fistula. Further, to prevent air leak, infusion of fibrin glue was performed bronchoscopically. The surgery was simple, speedy and mini-invasive treatment choice for GEB patients [12]. Infusion of adhesive agents into the GBE contributes to the fibration of bulla wall and collapse of bulla, and prevention of bronchopleural fistula. While infusion of fibrin glue contributes to the close of bronchus [12, 13]. In this study, significant right-shifting was noticed in the mediastinum caused by GBE, and the internal pressure was up to 22 cm H_2O, demonstrating the patient suffered from tension GBE. On this condition, CT-guided drainage of GBE under local anesthesia, which showed immediate improvement of shortness of breath together with decrease of bulla volume, and attenuation of mediastinal shifting. This indicated that CT-guided drainage is effective for the decompression of tension GBE.

In this case, drainage of GBE was performed in an accurate and safe manner under CT monitoring. A 1.7 mm catheter was used, therefore, the approach was considered to be simple and less-invasive, and could avoid the pneumothorax. However, much attention should be paid to avoid the pneumothorax during and/or after the GBE decompression, and immediate CT recheck was needed after insertion of catheter. As the drainage may often end with failure due to prolonged air leakage following drainage, erythromycin was injected into the targeted lobe bronchus by bronchoscope that was considered responsible for the air leakage. Combination of intracavitary drainage, autologous blood and thrombin and endobronchial occlusion can contribute to the earlier recovery [7]. Sufficient collapse of the bulla was observed and the reduction persisted 14 months after drainage. During the follow up, significant improvement was noticed in the subjective symptoms, life quality and pulmonary function compared with the baseline levels.

Special care should be taken to monitor the potential risks induced by unstable circulation and re-expansion of pulmonary edema during the transbronchial decompression of GEB [14, 15]. In this case, deteriorated shortness of breath was noticed due to excessive aspiration, and the internal pressure of GBE decreased from 22 cm H_2O from 10 cm H_2O, which may be associated with the unstable circulation. On this basis, we recommended the drainage should be slow and the aspiration volume should not be too much. Continuous negative pressure drainage is beneficial for the closure of GBE, but a low pressure is required to prevent the complications due to fast drainage. In this study, we developed a low pressure suction, which proved equally effective and minimized the hazard of circulatory instability in critical practices.

Conclusions

We developed a CT-guided bulla drainage method for inoperable patients with GEB. The method was approved equally effective and could minimize the hazard of circulatory instability in critical practices. In future, we will focus on large sample studies to valid its clinical efficiency.

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Disclosure of conflict of interest

None.

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