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Expeditious Resolution of Giant Bullae with Endobronchial Valves and Percutaneous Catheter Insertion

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As bullae contribute to decreased lung function in chronic obstructive pulmonary disease (COPD) patients, effective decompression of large bullae is important. Bronchoscopic lung volume reduction via endobronchial one-way valves is less invasive and has a lower mortality rate than lung volume reduction surgery. We report the case of a 48-year-old male who presented with giant bullae that were expeditiously resolved with endobronchial valves and percutaneous catheter insertion. Three days later, imaging revealed marked decreases in the extent of bullae and atelectasis of the contralateral lung without any complications, including air leakage or pneumothorax. Combination of endobronchial valves and percutaneous catheter insertion might be helpful to accelerate the release of large bullae and to achieve improved lung function and higher levels of physical activity in patients with COPD.

Key Words: Bullae, endobronchial valves, bronchoscopic lung volume reduction, chronic obstructive pulmonary disease

INTRODUCTION

A nonventilated air space in the lung measuring more than 1 cm in diameter is defined as a bulla, which is commonly associated with emphysema and chronic obstructive pulmonary disease (COPD).[1] Bullae contribute to decrease the forced expiratory volume in 1 sec (FEV_1), and increase the residual volume/total lung capacity ratio of lung, leading to chronic breathlessness and poor quality of life.[2,3] Thus, effective decompression of a large bullae is important to improve the lung function of COPD patients.

Various surgical methods have been proposed in the decompression of large bullae, from local excision of bullae to lobectomy of lung[4]; however, as postoperative mortality of bullectomy were up to 27.8% and not confer a survival advantage over medical therapy, the benefits and risks of surgical treatment of bullae in COPD patients have to be critically assessed.[5] Recently, there is increasing evidence that bronchoscopic lung volume reduction via endobronchial one-way valves (EBV) is less invasive and has a lower mortality rate with improved pulmonary function and patient-reported outcome than lung volume reduction surgery.[6,7]

Here, we report a 48-year-old male presented giant bullae, expeditiously resolved with endobronchial valves and percutaneous catheter insertion. Written informed consent was obtained from the patient for publication of this manuscript and any accompanying images.

CASE REPORT

A 48-year-old male with a 2-year history of progressive dyspnea was referred for the management of giant bullae. His spirometry showed severe COPD with a predicted FEV_1 of 40%, and the ratio of FEV_1 to forced vital capacity (FVC) was 67%. On chest X-ray and computer tomography (CT) scan (Fig. 1), multiple

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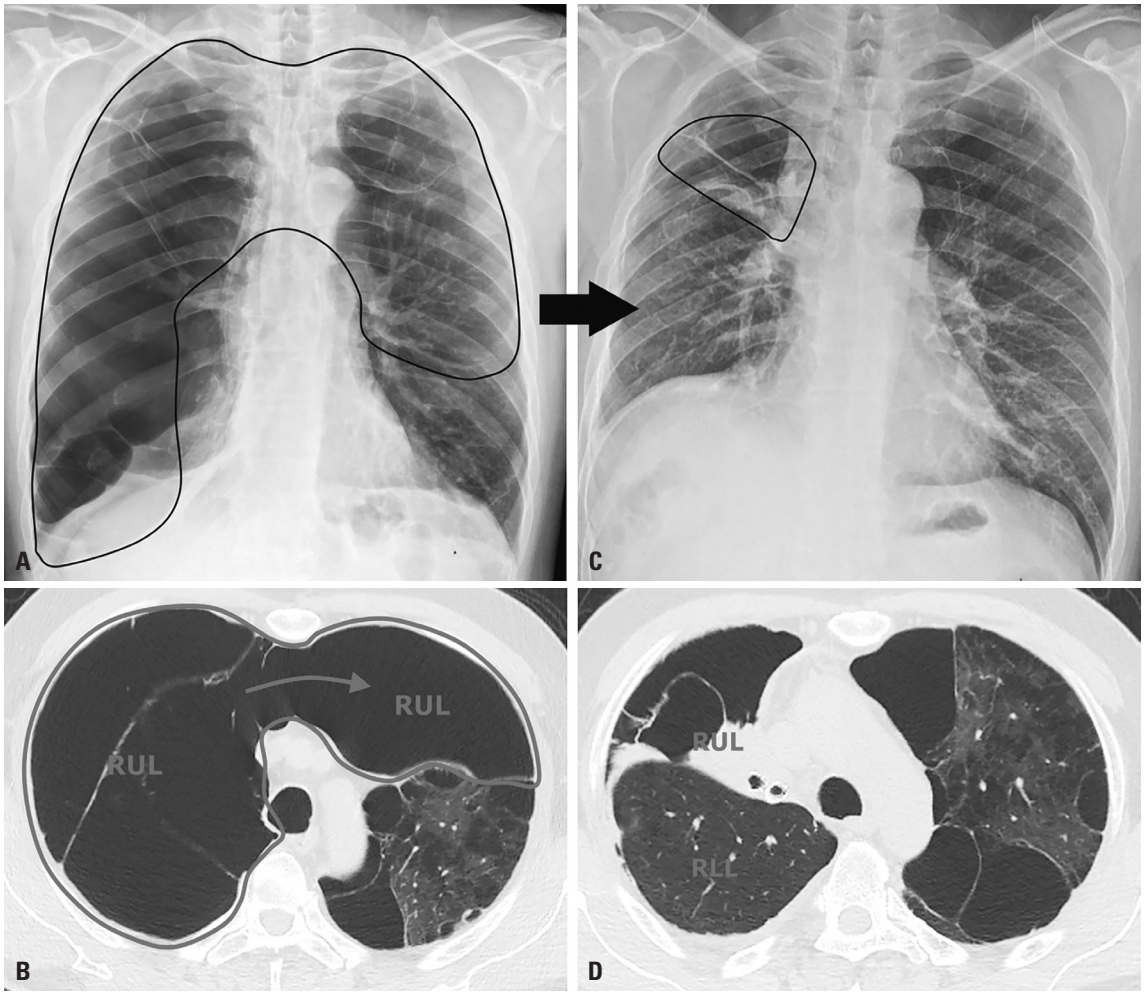

Fig. 1. Changes in radiologic findings before and after treatment. (A) Chest X-ray before treatment. (B) Chest computed tomography (CT) scan before treatment. (C) Chest X-ray and (D) chest CT scan after treatment. Chest X-ray in the posteroanterior projection and CT scan show an extensively hyperaerated RUL that is entirely replaced by multiple clustered giant bullae. Owing to the mass effect of the over-expanded RUL, the adjacent right middle and lower lobe were severely compressed within the right hemithorax. Moreover, the super-overinflated RUL encroaches upon even the contralateral left hemithorax across the anterior mediastinal junction line, occupying a large space in the left hemithorax, which was also detrimental to otherwise normal inflation of the left lung. After insertion of endobronchial valves and percutaneous drainage catheter, the volume of previously overinflated RUL markedly decreased, showing near-complete lobar atelectasis. RUL, right upper lobe; RLL, right lower lobe.

clustered giant bullae occupied his right upper lobe and encroached the left hemithorax across the anterior mediastinal junction line. The right middle and lower lobes were severely compressed by these giant bullae. The right interlobar fissures were complete at CT scan. Bronchoscopy under conscious sedation was performed for implantation of three EBVs (Zephyr, 4.0 mm). Postprocedural chest radiograph at 4 hours after bronchoscopy showed no acute complications, including pneumothorax. A percutaneous drainage catheter was inserted in the bullae for more rapid drainage of entrapped air within giant bullae (Fig. 2). Three days later, imaging exhibited remarkable decreases in the extent of bullae (Figs. 1 and 2) without any complications, including air leakage and pneumothorax. After 3 months, the patient recovered well with near-normal pulmonary function with a predicted FEV_1 of 79% and FEV_1/FVC of 74%. Six months later, the patient reported that he could exercise enough to sweat without breathlessness and had planned to return to work.

DISCUSSION

This report highlights an instance of expeditious resolution of giant bullae with a combination of EBVs and a percutaneous drainage catheter. EBV are implantable one-way valves, applicable to a patient with a flexible video-bronchoscopy under deep conscious sedation with spontaneous breathing, using short-acting benzodiazepine, such as midazolam and low dose propofol.[7,8] The most common acute complications of EBV insertion are pneumothorax and pneumonia, without major complications, such as death, and the effects of reduced lung volume usually remain stable for at least 5 years.[9,10] In the present

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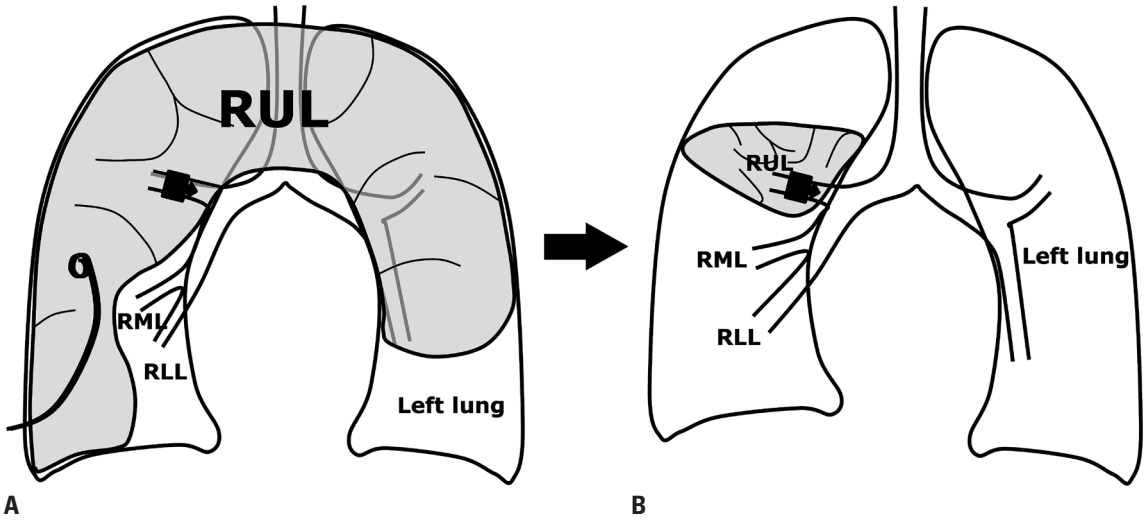

Fig. 2. Schematic drawing for illustration of the changes before and after endobronchial valve and pigtail catheter insertion. (A) Endobronchial valves (box with arrow) were inserted in the RUL segmental bronchi. Afterwards, a 5-French pigtail catheter was inserted in one of the clustered multiseptated giant bullae occupying the entire RUL (gray-colored area). (B) After catheter drainage of a huge quantity of entrapped air within the bullae, the volume of the previously overinflated RUL markedly decreased, showing near-complete lobar atelectasis (gray-colored area). RUL, right upper lobe; RLL, right lower lobe; RML, right middle lobe.

case, we drained a huge quantity of entrapped air within the bullae via a percutaneous catheter after EBV insertion. It is noteworthy to mention that the volume of the previously overinflated right upper lobe markedly decreased in just a few days and that the atelectasis of the right middle, right lower lobes, and even contralateral left lung, also completely re-expanded, completely relieving the mass effect of giant bullae in the right upper lobe.

Recently, several randomized controlled trials have set out to evaluate the role of EBV insertion in severe emphysema patients. These studies reported that emphysema patients without collateral ventilation who were treated with EBV showed better clinical improvements, assessed by lung function and exercise capacity, than controls.[11-14] To the best of our knowledge, clinical outcomes were assessed at 6–12 months after bronchoscopic lung volume reduction treatment using EBVs. However, without active intervention with closed thoracotomy drainage of bullous air in the correct upper lobe, it might take months for giant bullae to collapse spontaneously by very slow, natural gradual resorption of entrapped air, even after insertion of EBVs. In this regard, when complete interlobar fissures are confirmed on chest CT scan, thereby eliminating any risk of unexpected collateral air drift, closed thoracotomy of bulla puncture for rapid drainage of bullous air can help patients to recover instantaneously from persistent respiratory distress after insertion of EBVs.

In a study conducted in the Netherlands, patient-reported outcomes and goals were investigated 1 year after EBV insertion.[15] Most of the patients (77%) were able to walk, and only 25% of the patients could exercise. COPD patients with giant bullae were excluded from the largest clinical trial of EBV in treatment of COPD.[16] However, a couple of case series have reported the efficacy of EBV treatment for giant bullae, albeit with limited information regarding lung function, imaging, or physical activity.[3,17] One case series showed that out of five patients, only one patient whose bulla occupied the whole right middle lung experienced a long-term benefit. In our case, the giant bullae in right upper lobe showed sustained long-term improvement, leading to the patient being able to exercise with lung function improvement. Considering that the indication for EBV insertion in the LIBERATE study was 40 to 75 years of age,[7] higher goals in relation to quality of life and physical activity should be set. Our case suggests that resolution of giant bullae with EBVs and percutaneous catheter insertion might be useful to helping reach these higher goals. Further studies are, however, warranted to determine the role of additional percutaneous catheter drainage after EBV insertion in giant bullae in terms of improvement of pulmonary function and patient-reported goals.

A few potential limitations should be acknowledged. First, regarding the possibility that the presence of collateral ventilation is associated with iatrogenic bronchopleural fistula with percutaneous drainage after EBVs implantation, chest CT was evaluated for completeness of fissures before EBV insertion. However, chest CT may not be sufficient to assess complete fissure. Use of the Chartis system is necessary to verify negative collateral ventilation and to lower the risk of postprocedural complications, such as bronchopleural fistula, even though the Chartis system does not completely predict the risk of complications. Second, the patient in our case study was considered as a suitable candidate for expeditious resolution of giant bullae via sequential EBV and Pig-tail catheter drainage (PCD) insertion after careful evaluation by a multidisciplinary team comprised of expert pulmonologists and radiologists. There-

fore, the results of our study may not be generalized to other patients with giant bullae. Careful review of candidates is essential, focusing on other comorbidities of the patient and the size and location of bullae that may affect the potential benefits and risks from two consecutive interventions. Moreover, the risk of severe complications, such as bronchopleural fistula, pneumothorax due to rapid expansion of the affected lung, prolonged air leakage, and EBV removal, should be considered before the procedure. Third, future studies should be conducted to evaluate the long-term efficacy and safety of this technique with a larger number of patients and a well-designed study protocol that considers both ethical and scientific aspects, including the appropriate time point to insert PCD after EBV and treatment strategies for the management of severe complications.

In conclusion, EBV insertion is feasible and efficient in lung volume reduction, although the time to reduction may take up to a month.[7,10] A combination of EBV and percutaneous catheter insertion might be helpful to accelerate the release of large bullae and the achievement of improved lung function and higher levels of physical activity in patients with COPD. Notwithstanding, the benefits and risks of this technique should be considered carefully before application to patients.

AUTHOR CONTRIBUTIONS

Conceptualization: Yunjoo Im and Hojoong Kim. Data curation: Yunjoo Im and Tae Sung Kim. Formal analysis: Yunjoo Im, Byeong-Ho Jeong, and Hye Yun Park. Funding acquisition: Hojoong Kim. Investigation: Hojoong Kim. Methodology: Byeong-Ho Jeong and Hye Yun Park. Project administration: Hojoong Kim. Resources: all authors. Supervision: Hojoong Kim. Visualization: Tae Sung Kim. Writing—original draft: Yunjoo Im and Hye Yun Park. Writing—review & editing: all authors. Approval of final manuscript: all authors.

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