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CASE REPORT

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A novel approach involving reversed placement of endobronchial valves combined with retrograde methylene blue instillation for the localization of multifocal bronchopleural fistula

Weihua Xu[1] | **Ming Chen**[1] | **Jiagan Xu**[1] | **Lei Wang**[2] | **Congbin Peng**[3] | **Guohong Mo**[4]

[1]Respiration Department, Tongde Hospital of Zhejiang Province, Hangzhou, China

[2]Cardiothoracic Surgery Department, Tongde Hospital of Zhejiang Province, Hangzhou, China

[3]Anesthesiology Department, Tongde Hospital of Zhejiang Province, Hangzhou, China

[4]Endoscopy Department, Tongde Hospital of Zhejiang Province, Hangzhou, China

Correspondence
Weihua Xu, Respiration Department, Tongde Hospital of Zhejiang Province, 234#, Gucui Road, Hangzhou, Zhejiang Province, 310012, China.
Email: xwhzju@163.com

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Abstract

The bronchopleural fistula (BPF) is a pathological communication between the bronchus and the pleural space. Diagnosing BPF poses a significant challenge for physicians, particularly when identifying multifocal BPFs. Traditionally, retrograde instillation of methylene blue (MB) into the pleural cavity with simultaneous observation with a bronchoscope has been used to locate a BPF. However, MB instillation is not effective in identifying multifocal BPFs. In this article, we report a new method for locating multifocal BPFs which involves placing the endobronchial valve (EBV) in reverse combined with retrograde MB instillation. First, the thoracic cavity is filled with MB solution. Then, using bronchoscopy, the location of a BPF can be identified as the MB solution flows into the bronchus. Secondly, an EBV is deployed in reverse in the bronchus where the identified BPF is located. Retrograde MB instillation is then repeated to locate any additional BPFs until no new ones are found. Two cases were reported using this novel method to identify multifocal BPFs, and each case was ultimately diagnosed with 2 BPFs. After precisely locating all the BPFs, the EBVs are then removed and placed forward in the target bronchi for treating the BPFs. During the follow-up period, no recurrence of BPFs was observed. We conclude that reversed placement of EBVs combined with retrograde MB instillation appears to be an effective approach for locating multifocal BPFs.

KEYWORDS
bronchopleural fistula, bronchoscopy, empyema, interventional pulmonology

INTRODUCTION

The bronchopleural fistula (BPF) is a pathological communication between the bronchus and the pleural space. The mortality rate of BPF ranges from 16% to 72% [1]. It can be a complication of pneumonectomy, pulmonary or segmental resection, with a reported incidence between 1.5% and 28% [1]. BPFs are associated with higher morbidity and mortality rates, as well as pulmonary and/or pleural infections, prolonged hospital stays, and increased resource utilization. Diagnosing BPFs poses great challenges for physicians. To date, several methods have been used to diagnose BPFs. These techniques can be classified into two categories: (a) identifying the presence of a BPF and (b) locating its site. Obviously, the latter is more important for pulmonologists who decide to treat BPFs using bronchoscopy.

Retrograde instillation of methylene blue (MB) into the pleural cavity with simultaneous observation with the bronchoscope is an alternative approach to locate a BPF [2]. However, MB instillation cannot effectively identify multifocal BPFs. This article reports a new method for locating multifocal BPFs, which involves reversed placement of an endobronchial valve (EBV) combined with retrograde MB instillation.

CASE 1

A 27-year-old woman underwent a wedge resection of the apicoposterior segment and anterior segment of left upper lung due to a pulmonary nodule. The postoperative pathology report revealed adenocarcinoma. Five months later, she was admitted for empyema with symptoms of fever, cough, and sputum. A 10-Fr pigtail catheter (Guangdong Baihe Medical Technology Co., Ltd. China) was placed in our hospital to treat the condition. Chest CT showed a left hydropneumothorax (as shown in Figure 1A). Aspergillus was identified from pleural fluid culture and the patient received voriconazole treatment until infection control was achieved. Subsequently, flexible bronchoscopy revealed that MB solution (20 mg/250 mL) instilled into the thoracic cavity entered LB4. After cleaning up MB solution inside bronchi, a version 4.0 EBV (Zephyr endobronchial valve, Pulmonx Inc., USA) was reversely placed in LB4 with its edge covered by EBV (as shown in Figure 1B). Then another 50 mL of diluted MB was injected into the pleural cavity via the pigtail catheter, and the dye entered LB1 + 2 and LB3's tracheobronchial tree. A version 5.5 EBV (Zephyr endobronchial valve, Pulmonx Inc., USA) was deployed inversely in the superior division bronchus. The instillation of 100 mL MB was repeated, but no new BPFs were found. Based on the precise location of multifocal BPFs, the EBVs inside the bronchi were removed and re-placed in a forward direction in LB4 and superior division bronchus (shown in Figure 1C). On the same day, pleural debridement was performed sequentially, and finally a 10-Fr pigtail catheter was placed for chest drainage. After placing the EBV, there was no air leak. One month later, a pedicled pectoralis major muscle tamponade operation was performed. During the 6-month follow-up period, neither BPF nor empyema recurred (shown in Figure 1D).

CASE 2

A 74-year-old man underwent a right lower lobectomy and wedge resection of the right upper lobe for squamous cell carcinoma, followed by neoadjuvant chemotherapy with albumin paclitaxel and carboplatin. One month after the

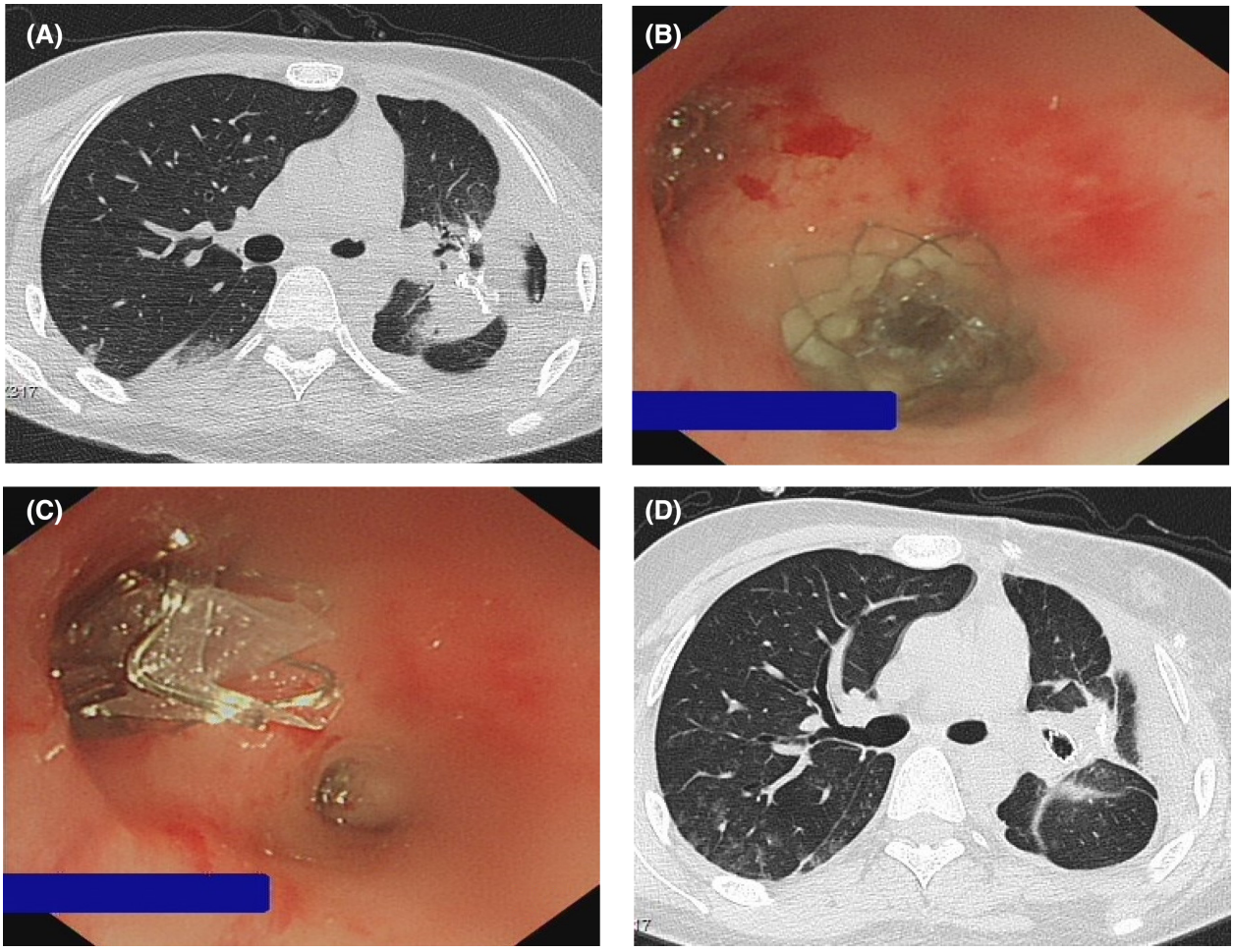

FIGURE 1 Case 1: (A) The computed tomography (CT) scan revealed a left hydropneumothorax. (B) A version 4.0 EBV was placed in LB4 in reverse, with the edge of the EBV covering LB5. The dye entered the intrinsic segment bronchus of the left upper lung after retrograde MB instillation. (C) The EBVs inside the bronchi were removed and replaced in a forward direction into LB4 and the left intrinsic segment bronchus. (D) During the follow-up period of 6 months, a CT scan showed neither pneumothorax nor empyema.

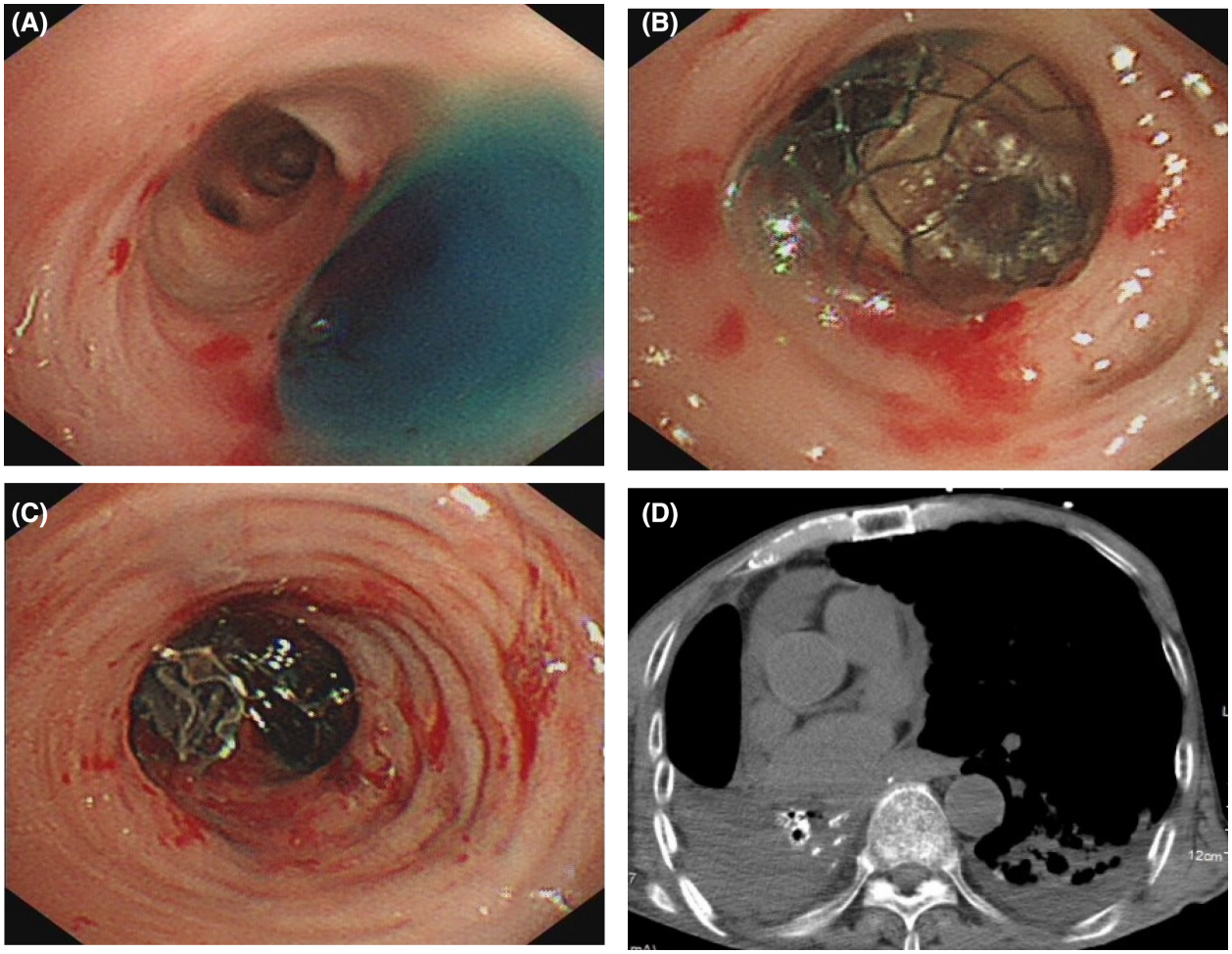

FIGURE 2 Case 2: (A) After retrograde instillation of 50 mL MB into the pleural cavity, the dye entered the airway from RB4. (B) A version 4.0 EBV was placed in reverse position in RB4 (right bronchus) with the edge of EBV covering RB5 (left bronchus). Another 50 mL MB solution was injected into the pleural cavity, and the dye entered the airway from RB5. (C) The reversely placed version 4.0 EBV was removed and repositioned forward into RB4 (right bronchus). Another EBV of version 5.5 was deployed into RB5 (left bronchus). (D) After deployment of EBV, a chest computed tomography scan showed pulmonary atelectasis in the middle lobe.

operation, the patient developed an empyema, and a 10-Fr pigtail catheter (Guangdong Baihe Medical Technology Co., Ltd. China) was placed for chest drainage. Flexible bronchoscopy was performed with simultaneous instillation of 50 mL MB into the pleural cavity. The dye entered the airway from RB4 (shown in Figure 2A). Then, a version 4.0 EBV (Zephyr endobronchial valve, Pulmonx Inc., USA) was reversely placed in RB4 with the edge of EBV covering RB5. Another 50 mL of diluted MB was injected into the pleural cavity through the pigtail catheter, and the dye entered the tracheobronchial tree via RB5 (shown in Figure 2B). The reversely placed EBV was removed and replaced in the forward direction in RB4. Another EBV of version 5.5 (Zephyr endobronchial valve, Pulmonx Inc., USA) was deployed to RB5 (shown in Figure 2C). Pleural debridement was performed sequentially, and another 10-Fr pigtail catheter was finally placed for chest drainage. After the deployment of the EBV, no air leak occurred. Twenty days later, a pulmonary atelectasis in the middle lobe was observed on chest CT (shown in Figure 2D). During the six-month follow-up period, neither BPF nor empyema recurred.

DISCUSSION

The precise location of BPF is a key premise for successful treatment. So far, several methods have been developed to diagnose BPF. These techniques involve direct bronchoscopic observation, retrograde MB instillation, bronchography, measurement of gas concentration changes in the pneumonectomy cavity after inhalation of different concentrations of oxygen and N_2O, balloon occlusion of the bronchi particularly for locating peripheral or segmental BPF that leads to an air leak, radiological techniques especially chest CT and three-dimensional reconstruction, capnography, inhalation of radiolabeled aerosol and sequential single-photon emission tomography (SPECT), and computed tomography bronchography (CTB).

The use of retrograde MB instillation was first described by Hsu et al [3] in 1972. This procedure is quick, inexpensive, effective, and easy to perform, especially when combined with bronchoscopy for locating the BPF. However, the retrograde MB instillation approach may not identify multifocal BPFs because the dye tends to follow

the path of least resistance. In other words, it passes through relatively 'easy' BPFs and avoids those that are relatively 'difficult' to pass. Therefore, pure retrograde MB instillation alone cannot locate multifocal BPFs in empyema patients who often have multiple BPFs due to lung tissue damage at several sites caused by pus immersion.

Each of the 2 patients in the present study received instillation of MB in the pleural cavity, and only one BPF was found. When using reversely placed EBV to occlude the identified BPF, another MB instillation diagnosed a second BPF. The results showed that the 2 patients had two BPFs.

Initially designed for endoscopic reduction of lung volume [4], EBVs interrupt air flow into the terminal bronchus during inspiration and allow exhalation of air and secretions. Subsequently, EBVs were also used for prolonged air leaks and BPFs [5]. When an EBV is placed in reverse, airflow or secretions from the terminal bronchus to the central airway are blocked. This feature opens up possibilities for exploring additional potential BPFs following retrograde MB instillation. Based on this principle, EBVs can be used to locate multifocal BPFs before being finally deployed to treat potential BPFs.

As showed above, we describe the successful use of reversed placement of EBVs, which, when combined with retrograde MB instillation, appears to be an effective approach for locating multifocal BPFs.

AUTHOR CONTRIBUTIONS

Weihua Xu: Designing the operation; final approval of the version to be published; methodology. **Ming Chen**: Revising the manuscript; investigation. **Jiagan Xu**: Writing the manuscript; methodology. **Lei Wang**: Collecting the relevant clinical data; investigation. **Congbin Peng**: Ensuring the accuracy or integrity of the operation. **Guohong Mo**: Investigation.

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CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

All data generated or analyzed during this study are included in this article. Further enquiries can be directed to the corresponding author.

ETHICS STATEMENT

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

ORCID

Weihua Xu https://orcid.org/0000-0002-9123-8748

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