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Case report

Surgically intractable bronchopleural fistula treated with endobronchial valve insertion by isolating the tract with indigo carmine: A case report

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ABSTRACT

Bronchopleural fistula (BPF) is a communication between the pleural space and the bronchial tree. Herein, we report a patient with persistent air leak after necrotizing pneumonia, where tract of BPF was not apparent in computed tomography (CT). Despite control of infection, watchful observation, repetitive procedures, and surgery, air leak was not resolved. This led to consideration of the endobronchial valve (EBV) placement. After identifying the bronchial segment leading to the fistula with indigo carmine, precise occlusion of the lingular division of the left upper lobe (LUL) was performed using an EBV. Subsequently, the air leak was resolved. During 6 months of follow-up, neither recurrence of BPF nor any procedure-related complications were noted. We concluded that precise EBV placement by identifying bronchial segment leading to the BPF, could be an effective treatment for persistent air leak.

1. Introduction

Bronchopleural fistula (BPF) is a communication between the pleural space and the bronchial tree. BPF carries a high risk for morbidity and mortality and is associated with prolonged hospital stay and high resource utilization [1].

The treatment of BPF varies from conservative management to aggressive surgical procedures due to the lack of established guidelines or consensus. This suggests that no optimal therapy is available; rather, that current interventions are complementary and the treatment should be individualized [1]. The use of endobronchial valve (EBV) is one of the suggested therapeutic options.

In this case, we report bronchopleural fistula where tract was not evident and other treatment options such as observation, repetitive procedures, and surgery were not sufficient, and was eventually treated with endobronchial valve insertion by isolating the tract with indigo carmine.

2. Case report

A 64-year-old man without any significant past medical history presented to the emergency department with sputum and fever. Computed tomography (CT) revealed consolidation in both lungs, leading to the start of empirical therapy according as pneumonia. As sputum culture results revealed Klebsiella pneumoniae, the antibiotic coverage was narrowed to amoxicillin-clavulanic acid, based on an antibiotic sensitivity test. On day 9 of hospitalization, respiratory distress progressed, and a follow-up CT scan showed suspected ruptured necrotizing pneumonia with pleuritic and complicated pleural effusion. The patient was transferred to the intensive care unit (ICU) for close observation with the administration of high flow nasal cannula (HFNC). In addition, a left pleural percutaneous drainage was placed for effective drainage of the pleural effusion where K. pneumoniae growth was also found.

Although the oxygen requirement, symptoms, and parameters of infection improved over time, persistent air leak raised the suspicion of BPF. However, follow up CT scan only showed a decrease in the extent of his pneumonia, increased amount of left complicated hydro-pneumothorax but no definite tract of BPF. To manage consistent air

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Abbreviations

BPF	bronchopleural fistula
CT	computed tomography
EBV	endobronchial valve
ICU	intensive care unit
LLL:	left lower lobe
LUL:	left upper lobe
VATS	video-assisted thoracic surgery
HFNC	high flow nasal cannula

the instillation of indigo carmine through bronchoscopy to identify the bronchial segment leading to the fistula. Since injection of indigo carmine from the LUL result in expulsion into the LLL due to gravity and supine position of the patient, procedure started from segmental bronchus of LLL. When indigo carmine was injected to segmental bronchus of LLL and upper division segmental bronchi of LUL, only a small amount passed out through the chest tube. However, a large amount of indigo carmine was expelled instantly to the chest tube when instilled into the lingular division of the LUL. EBV (Zephyr) was placed into the lingular division of the LUL and air leak was resolved subsequently (Fig. 2). After removal of the chest tube, the patient was discharged. During the six months of follow-up, there were no EBV-associated complications or recurrence of air leak (see Fig. 3).

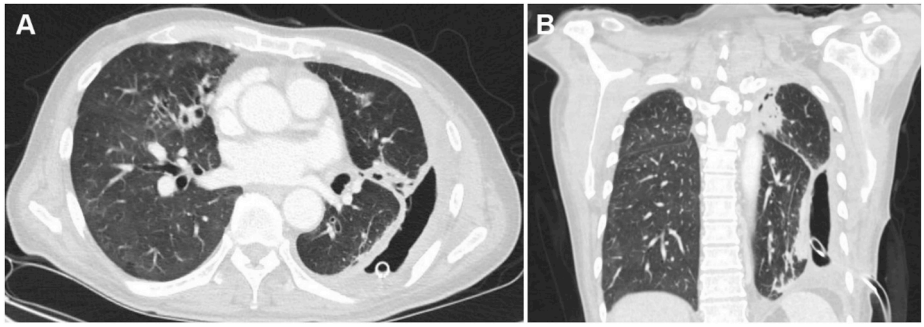

Fig. 1. Axial (A) and Coronal (B) thoracic computed tomography (CT) images showing complicated hydropneumothorax due to loculated empyema with chest tube. However, tract of Bronchopleural fistula (BPF) is not apparent.

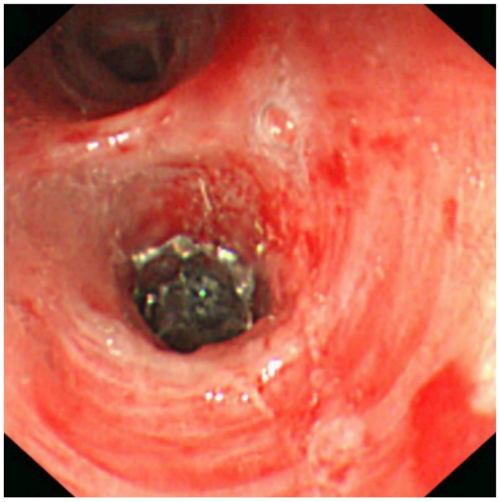

Fig. 2. Bronchoscopic view of the Endobronchial Valves (EBV) placement in the lingular division of the left upper lobe (LUL).

leak, the pleural percutaneous drainage was changed to chest tube, with Heimlich bag kept for a month, not resolving the problem (Fig. 1). This eventually led to pleural exploration and decortication via video-assisted thoracic surgery (VATS), followed by additional application of polyglycolic acid felt (Neoveil) and glue via VATS for three times.

As air leak continued despite repetitive surgery and procedures, the need for additional surgery was raised. However, due to extensive range of pneumonia and empyema from the left lower lobe (LLL) to the left upper lobe (LUL), pneumonectomy had to be considered. Weighing gains and losses, we decided to undertake an endoscopic approach. Because BPF was not apparent from CT scan, the procedure began with

3. Discussion

BPF is considered as a dreadful complication of several pulmonary disorders. The etiologies of BPF include postoperative complications of pulmonary resection, which is the most common cause, followed by necrotic lung complicating infection, chemotherapy or radiotherapy (for lung cancer), persistent spontaneous pneumothorax, and tuberculosis [1].

The diagnosis of BPF is often obvious from the clinical presentation, particularly with the chest tube. Radiological features including steady increase in intrapleural air space, appearance of a new air fluid level, changes in an already present air fluid level also suggest the presence or the development of a BPF [2]. On the other hand, demonstration of actual fistulous communication of BPF may not be possible. By using fiberoptic bronchoscopy (FOB), direct visualization of the fistula opening or return of continuous bubbles on bronchial wash may locate the tract. In some cases, placing a balloon-tip catheter into the selected airways and inflating it to occlude the tract could be another option.

In this case, selective instillation of indigo carmine into segmental bronchi with its subsequent appearance in the chest drainage was used to identify the tract of BPF. In our experience, small amount of indigo carmine (1ml) instillation has been effective in sectioning bronchus to the finest range to subsegmental bronchus, without additional use of balloon catheter.

The treatment options for BPF include conservative management; different bronchoscopy techniques, such as glues, coils, and sealants; use of the EBV; and surgery. Guidelines from the American College of Chest Physicians (2001) and the British Thoracic Society (2010) recommend surgical evaluation if an air leak does not resolve spontaneously after four or three to five days [3,4]. The success rate of surgical closure of BPF has been reported to be between 80% and 95% but is associated with a risk for open thoracotomy [5]. Considering the risk for surgery in high-risk patients and a lack of controlled studies comparing other therapeutic options, the application of endobronchial therapy has increased, including the insertion of EBV.

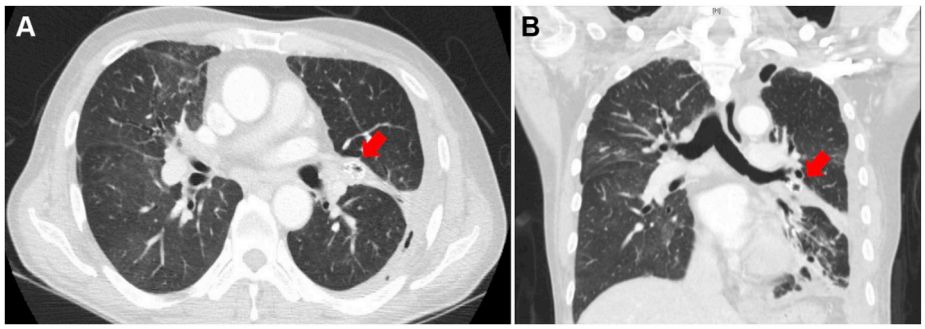

Fig. 3. Axial (A) and Coronal (B) images from thoracic computed tomography (CT) scan 1 month after insertion of EBV in the lingular division of LUL (red arrows). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

EBVs, which were originally developed for the endoscopic reduction of lung volume in emphysema, and offering a non-surgical minimally-invasive alternative, have been available since 2003 [6]. The first attempt to use EBVs for the treatment of BPF was in 2005, leading to approval as a humanitarian-use device for the treatment of BPF in 2006 [7]. EBVs are introduced through the working channel of a flexible bronchoscope utilizing a unidirectional valve to prevent airflow into the diseased area of the lung resulting in the collapse of the distal lung parenchyma and attenuation of an air leak [8]. Recently, Bermea et al. [9] present the largest case series for the use of EBVs as treatment for persistent air leaks, suggesting successful removal of thoracotomy tube in eighty percent of cases with only three percent of patients with device failure.

To best of our knowledge, this is the first report of successful EBV therapy in a patient with persistent air leak, without apparent tract of BPF, which was refractory despite effective control of infection, sufficient observation, repetitive surgery, and procedure. However, precise identification of the involved segment of the bronchus by using indigo carmine and placement of EBV eventually led to successful treatment.

Declaration of competing interest

There are no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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