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Case Report

Retrograde instillation of methylene blue for the localization of bronchopleural fistula

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Abstract Bronchopleural fistulas (BPF) resulting from pulmonary resection, radiation, or a direct tumor mass effect are associated with significant morbidity, reduced quality of life, and increased risk of mortality. Diagnosing BPF can be challenging, even with computed tomography, magnetic resonance imaging, and bronchoscopy. We report a case in which retrograde methylene blue instillation during bronchoscopy successfully confirmed the diagnosis of BPF.

Key words: Bronchoscopy, Methylene blue, Pleura, Respiratory tract fistula

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INTRODUCTION

Bronchopleural fistulas (BPFs) represent pathological communication between the bronchial tree and the pleural space.[1] These fistulas can arise as severe complications associated with various pulmonary conditions, particularly following lung surgery or invasive pulmonary procedures, such as bronchoscopy, lung biopsy, or pleural biopsy.[1] These fistulas are associated with significant morbidity, decline in the quality of life, and increased mortality risk. The incidence of BPFs following pneumonectomy ranges from 4% to 20%, with mortality rates ranging from 27% to 71%.[2]

The diagnosis and management of BPFs pose significant clinical challenges and often require the involvement of pulmonary specialists.[3] Several diagnostic modalities including bronchography, computed tomography (CT), and magnetic resonance imaging (MRI), have been used to identify BPFs. Bronchoscopy has emerged as a pivotal tool for the diagnosis and therapeutic management of BPFs.[3]

Various diagnostic methods have been used to localize BPFs. Bronchoscopy is used to identify mucosal defects in the proximal airways. Methylene blue (MB) instillation is effective for detecting the fistula.[4] MB instillation is an inexpensive and safe diagnostic tool. Anterograde instillation of MB through the surgical stump, followed by detection in the chest tube, is well-documented in the literature.[3] Another approach involves retrograde instillation of the MB into the pleural drainage catheter while simultaneously observing the bronchi via bronchoscopy to localize the origin of the BPF.[4] We describe a case in which BPF was effectively diagnosed through the retrograde instillation of MB during bronchoscopy.

CASE REPORT

An 84-year-old man with a left-sided complicated parapneumonic effusion (PPE) was initially managed using a pigtail catheter and intrapleural lytic therapy. The patient was discharged with instructions to complete a 4-week course of amoxicillin-clavulanate. However, he was readmitted 2 months later with a chief complaint of fever and purulent sputum. On initial assessment, his vital signs showed a blood pressure of 134/88 mmHg, pulse rate of 98 beats/min, body temperature of 38.2℃, respiratory rate of 18/min, and an oxygen saturation of 94%. Physical examination revealed mild tachycardia and reduced breathing sounds at the base of the left lung, although the patient's respiratory effort appeared normal. Laboratory results indicated leukocytosis (15.4×10^9 cells/L) and an elevated C-reactive protein level (18.3 mg/dL), with other findings being unremarkable. Chest radiography revealed a pleural effusion in the left lower lobe. A subsequent chest CT scan showed a moderate amount of loculated effusion along with diffuse pleural thickening in the left lower thorax, suggestive of PPE recurrence (Fig. 1). A 12-Fr pigtail catheter was then inserted for the diagnosis and treatment of pleural effusion. The pleural fluid examination supported the diagnosis of empyema complicated PPE. The fluid had a pH of 4.863, white blood cell count of 589,272/mm³, and consisted of 90% neutrophils and 5% lymphocytes. The lactate dehydrogenase was 22,164 IU/L, protein was 2.1 g/dL, glucose was 22 mg/dL, and adenosine deaminase was 368 U/L. Sample cultures showed no bacterial growth after 48 hours.

Given the recurrence of complicated PPE, we suspected the potential presence of a BPF. Flexible bronchoscopy (FBS) was performed and no fistula was detected. We diluted 5 mL (10 mg/mL) of MB in 50 mL of normal saline and slowly administered it into the pleural cavity via a small catheter using a three-way valve. The dye was detected in the posterior bronchial segment of the left lower lobe, confirming the presence of BPF (Fig. 2). Surgical intervention was considered; however, because of the patient's old age and poor performance status, we decided to proceed with conservative treatment. Considering the stability of the patient's clinical condition in the days after the procedure, he was discharged with plans for follow-up. During the 1-year follow-up period, the patient remained stable without any signs of worsening PPE. This study was approved by the Institutional Review Board (IRB) of Jeju National University Hospital (IRB No. 2024-08-005). The requirement for

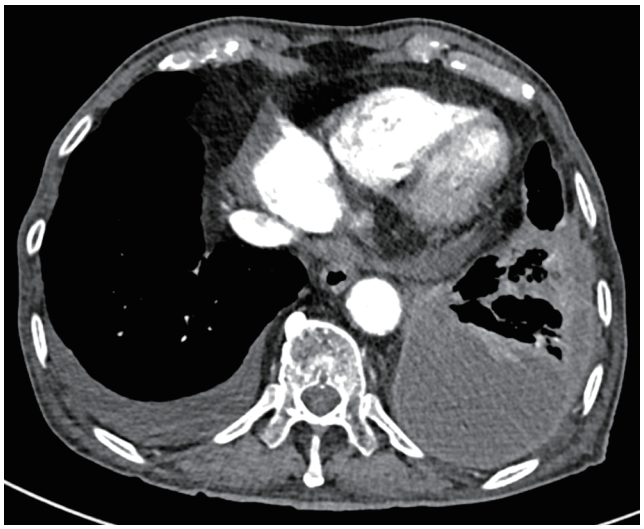

Figure 1. Chest computed tomography scan revealing moderate amount of pleural effusion in left hemithorax.

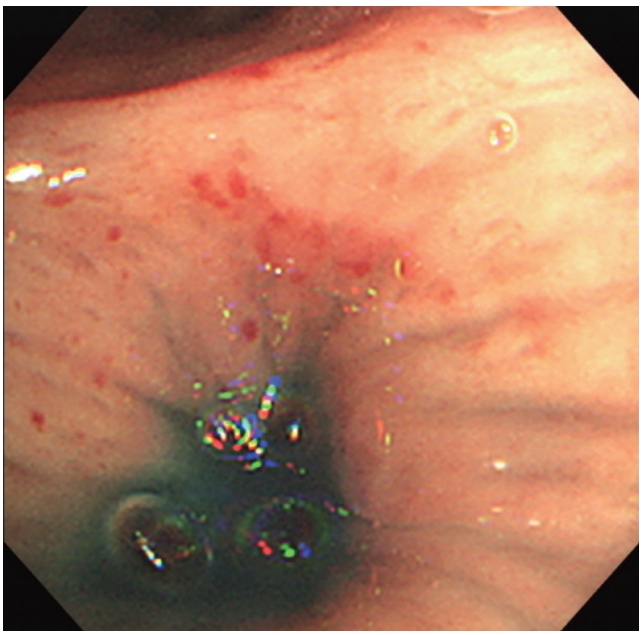

Figure 2. Bronchopleural fistula after retrograde instillation of methylene blue.

informed consent was waived by the IRB. We followed ethical guidelines, ensuring that all data was fully anonymized to protect privacy. No personally identifiable information was used in our reports.

DISCUSSION

BPFs are serious complications that can develop due to various conditions, including infections, Barrett's esophagus, cancers, thoracic injuries, acute respiratory distress syndrome (ARDS), and necrotizing lung disease associated with radiation or chemotherapy. In some cases, BPFs occur without a known cause. Among patients with lung cancer, BPFs are particularly associated with advanced disease stages, residual tumor presence in the bronchial stump post-surgery, and intrathoracic administration of chemotherapy. The incidence of BPFs after pulmonary resection varies widely, ranging from 1.5% to 28.0%, with the highest rates observed in patients who have undergone right pneumonectomy or right lower lobectomy.[2] These fistulas are frequently accompanied by complicated PPE and empyema, a severe complication of thoracic surgery, and associated with high morbidity and mortality rates.[2] Various other complications such as ARDS, aspiration pneumonia, respiratory dysfunction, infections, or vascular stump damage can further compromise survival. BPFs often lead to multiple hospitalizations and comorbid conditions, significantly increasing healthcare costs.[5]

Identifying BPFs can be difficult and presents a significant challenge for pulmonologists, radiologists, and other physicians managing symptoms. A precise diagnosis is essential to ensure that the patient receives appropriate treatment, which can reduce the risk of infection and improve quality of life. Several diagnostic approaches have proven effective in detecting BPFs, and chest imaging is often cited as a successful method. However, there are cases in which CT and MRI are not reliable for making a diagnosis. Moreover, FBS often fails to detect fistulas, especially when they are small or situated beyond the reach of the bronchoscope. Gas instillation and ventilation scintigraphy with gases, such as ^{133}Xe, ^{81m}Kr, and ^{99m}Tc diethylenetriamine pentaacetic acid, can be useful for diagnosing BPFs, although accurately identifying the precise location of the fistula can be difficult. In contrast, combining FBS with the instillation of a liquid dye not only allows for a precise diagnosis of BPF, but also helps identify the exact location of the fistula, making it easier to assess the success of subsequent treatment.[4]

The use of MB instillation into the affected pleural cavity through a needle or existing chest tube was introduced in 1972 for the evaluation of BPFs.[6] It is readily accessible, cost-effective, and relatively simple to apply in various surgical and non-surgical procedures, making it a vital resource in most hospitals. MB is an organic compound that appears as dark green crystals or crystalline powder with a bronze-like sheen.[4] When dissolved in water or alcohol, a deep blue solution is produced.[4] This compound is used as a bacteriological stain and indicator. It functions by inhibiting guanylate cyclase and has applications in the treatment of cyanide poisoning and reduction of methemoglobin levels.[4] It has a long history in clinical practice, especially during bronchoscopy, and is effective in diagnosing BPF. When prepared at a concentration of 10 mg/mL, MB can be diluted 50-250 times in saline, producing a solution that retains its vivid blue color and is practical for injection. An anterograde injection of MB can aid in diagnosing BPF, but does not reveal the exact site of the fistula. In contrast, retrograde instillation of MB through a drain in the abdomen or thoracic wall allows the bronchoscopist to identify the precise location of the BPF within the bronchial tree. Once identified, the leak can be sealed using various devices such as histoacryl glue, valves, stents, or other tools designed to stop air and fluid leaks.

We suggest that retrograde instillation of the MB be regarded as a primary diagnostic approach when BPF is suspected, especially if a drain is positioned on the thoracic or abdominal wall to allow for effusion drainage.

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REFERENCES

1. Leivaditis V, Braun-Lambur H, Windmüller V, Papatriantafyllou A, Huwe C, Lang D, et al. Bronchopleural fistula management with an amplatzer duct occluder: a comprehensive case report and literature review. Cureus 2023;15:e49431.
2. Abramian O, Rosenheck J, Taddeo-Kolman D, Bowen F, Boujaoude Z, Abouzgheib W. Therapeutic closure of bronchopleural fistulas using ethanol. Ther Adv Respir Dis 2021;15:17534666211044411.
3. Lois M, Noppen M. Bronchopleural fistulas: an overview of the problem with special focus on endoscopic management. Chest 2005;128:3955-65.
4. Ravenna F, Feo C, Calia N, Avoscan C, Barbetta C, Cavallesco GN. Retrograde instillation of methylene blue in the difficult diagnosis of BPF. Case Rep Med 2012;2012:714746.
5. Schneiter D, Grodzki T, Lardinois D, Kestenholz PB, Wojcik J, Kubisa B, et al. Accelerated treatment of postpneumonectomy empyema: a binational long-term study. J Thorac Cardiovasc Surg 2008;136:179-85.
6. Hsu JT, Bennett GM, Wolff E. Radiologic assessment of bronchopleural fistula with empyema. Radiology 1972;103:41-5.