

## Intractable Bronchopleural Fistula After Lung Resection in Mycobacterial Infection; Surgical and Endoscopic Treatment With Fibrin Glue Application Followed by Latissimus Dorsi Musculocutaneous Flap Coverage: A Case Report

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Postoperative bronchopleural fistula (BPF) is a challenging and complicated problem to cope with. Involving with multidisciplinary care team is essential for the best outcome. This report provides our experiences in intractable BPF after lung resection surgery which fail to completely heal after received surgical and endoscopic treatment. A 56-year-old female with no known underlying disease presented with nonmassive hemoptysis, productive cough, low-grade fever, and significant weight loss for 3 years. Her sputum consisted of *Mycobacterium abscessus* with multidrug resistant. Radiological examination revealed reticulonodular infiltration at middle lobe of the right lung and lingular lobe of the left lung, also a bronchiectatic change of both lungs. After 3 years of medical treatment, neither of her symptoms nor radiological findings improved. Therefore, a video-assisted thoracoscopic surgery (VATS) with middle lung lobectomy and lingulectomy was performed. After that, BPF at lingular stump occurred. Many surgical and endoscopic techniques followed by latissimus dorsi musculocutaneous flap along with vacuum dressing were introduced to encourage the complete healing of the BPF. One month later, the patient's clinical was improved and endoscopic findings showed nearly complete healing of the BPF. In conclusion, surgical and endoscopic treatments combined with postoperative vacuum dressing encourage patient's symptoms to be subsided.

**Keywords:** Bronchopleural fistula, Mycobacterial infection, Lung surgery, Latissimus dorsi flap, Vacuum dressing, Chest wall defect

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## Introduction

Bronchopleural fistula (BPF) is a sinus tract between the airway (trachea, main bronchus, and segmental bronchus) and the pleural space which resulted from many causes.<sup>1,2</sup> This condition is a rare condition but challenging problem for multidisciplinary health care team.<sup>3</sup> After BPF has occurred, it causes pneumothorax and contamination from the airway to pleural space which leads to chronic infection of pleural space and empyema thoracis.<sup>2</sup> One of the major causes of BPF results from unhealed bronchial stump after lung surgery (pneumonectomy, lobectomy and segmentectomy).<sup>4</sup> Risk factors for the occurrence of BPF comprise of diabetes mellitus, poor nutritional status, right pneumonectomy, long bronchial stump, radiation and postoperative mechanical ventilation.<sup>2,4</sup>

Morbidity and mortality rate of postoperative BPF are between 20% and 70%, respectively<sup>1,5</sup> which are very high but we can reduce it by early detection of the BPF and providing the proper management to the patient. Most of patients with the BPF suffer and die from aspiration pneumonia and acute respiratory distress syndrome.<sup>6</sup>

## Case Report

A 56-year-old Thai female with no known underlying disease presented with nonmassive hemoptysis, productive cough, low-grade fever, and significant weight loss for 3 years. In the first place, she went to private hospital then was examined by chest x-ray and computerized tomography (CT) chest which showed reticulonodular infiltration at the middle lobe of the right lung and lingular lobe of the left lung and bronchiectasis change of both lungs. Sputum was collected and showed *Mycobacterium abscessus* which resist all drug susceptibility test (multidrug resistant, MDR). Then the infectious specialist was consulted and prescribed clofazimine, moxifloxacin, clarithromycin, doxycycline, and amikacin to treat pulmonary nontuberculous mycobacterial infection. After that, symptoms were not improved and sputum still showed *Mycobacterium abscessus* whenever she came for follow-up. CT chest was serially

examined and showed no improvement of reticulonodular infiltration at the middle lobe of the right lung and lingular lobe of the left lung. After the diagnosis for 3 years, a video-assisted thoracoscopic surgery (VATS) with middle lung lobectomy and lingulectomy was done.

After postoperative day 3, she developed productive cough, and the next 4 months she developed dyspnea, weight loss, fever, and tachycardia, then CT chest was done to find the cause. Postsurgical empyema at left pleural cavity was considered then she was referred to Thammasat University Hospital for further management.

At Thammasat University Hospital, in the operation, the left thoracic cavity was explored under general anesthesia by VATS, we found entrapped remnant of the left upper lung surrounded by thick fibrous capsule, clot and fibrin, no pus was seen. Moreover, BPF-1 cm in diameter from the stump of lingular segment was identified. Therefore, standard thoracotomy was done through the 5th intercostal space and the 6th rib was resected. Lysis adhesion was performed by blunt dissection and all necrotic tissue was removed by tissue forceps. Decortication was completely done. BPF was closed by direct interrupted suture using PDS 3-0 and glue (Closeal). Intercostal muscle flap was mobilized to cover BPF. Air leak and bleeding were checked. Vacuum dressing was applied and continuously changed every 3 days. No organism was found from tissue culture. Tissue was sent to pathology unit and was reported that there was focal necrosis with few inflammation and extensive fibrosis.

On postoperative day 21, while changing vacuum dressing under intravenous sedation in the operative room, an air leak from BPF was detected. After that, bronchoscopy revealed recurrent BPF-5 mm in diameter at lingular stump. So, endobronchial occlusion was done with Fibringluraas.

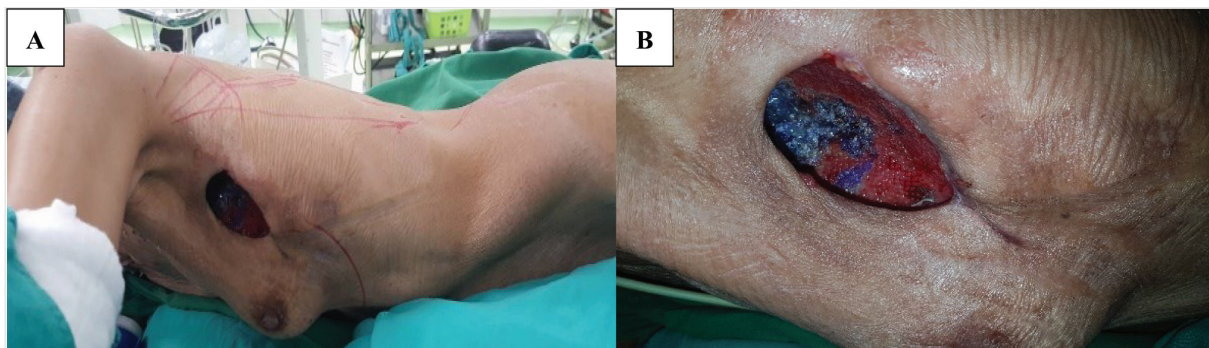
The air still persistently leaked from BPF, several procedures including endobronchial occlusion by ASD occlude (Amplazer), Histoacryl glue application, and direct suture by Prolene were done respectively. However, air leak from BFP stump remained. Before plastic surgeons was consulted for flap coverage 3 days, Histoacryl glue application was done. Finally, due to repetitively recurrent

air leakage, the musculocutaneous flap was performed to control the infection, promote BPF healing and cover a soft tissue defect (Figure 1).

The latissimus dorsi (LD) musculocutaneous flap was chosen in this operation. The LD flap was planned and designed (Figure 2A). Skin was incised along LD muscle longitudinally with a 5×10 cm center cutaneous island size. The LD muscle was dissected from its origin to

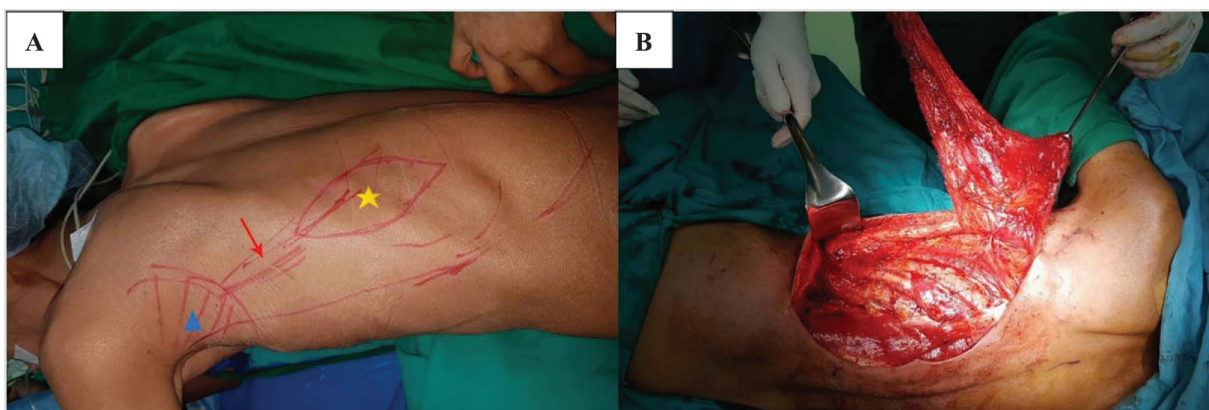
10 cm below its insertion (Figure 2B). The LD was elevated and mobilized with its cutaneous island (Figure 3A). Perforator arteries were preserved. The LD flap was rotated underneath the bridging skin to cover the left chest wall defect and BPF site then LD muscle was fixed with pleura to cover the BPF stump (Figure 3B). The recipient and donor site were closed layer by layer (Figure 3C-D).

**Figure 1. Bronchopleural Fistula and Soft Tissue Defect at the Left Chest Wall**



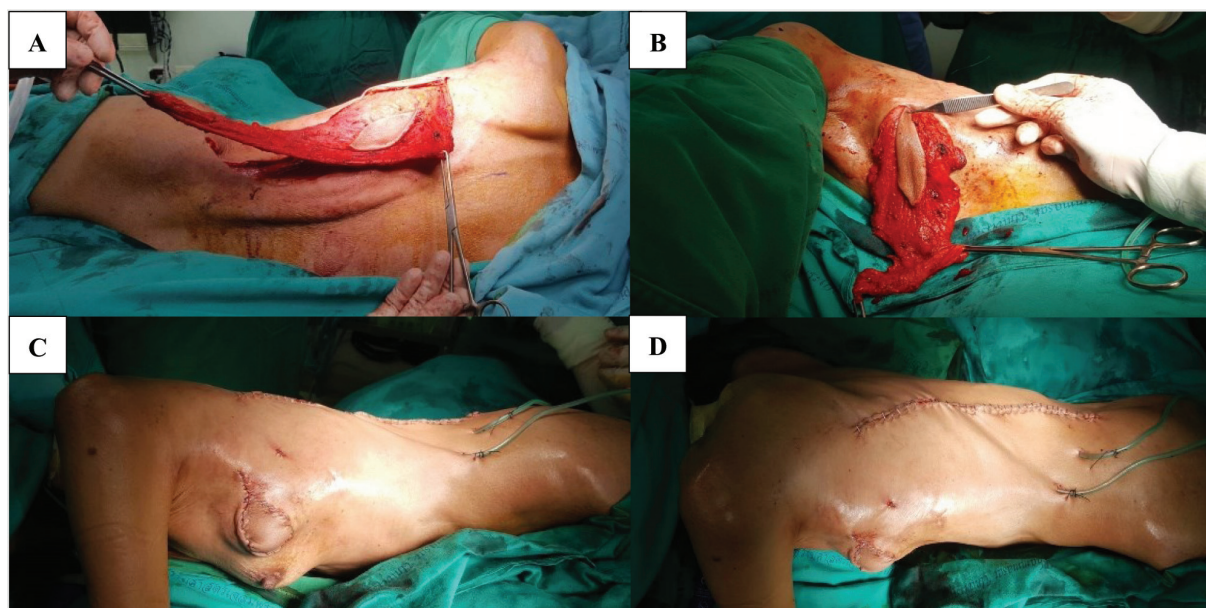
A, Patient was in right lateral decubitus position. The soft tissue defect located from anterior to posterior axillary fold, between the 2nd and 5th intercostal space. The defect was connected with left pleural space. B, The purple area shown suture material buried in Histoacryl glue from the prior surgery surrounded by good granulation tissue.

**Figure 2. Design and Dissection of Latissimus Dorsi Musculocutaneous Flap**



A, The skin pedicle (star), size of 5×10 cm, was designed for latissimus dorsi musculocutaneous flap. A triangle shows pivot point of the flap. An arrow shows accurately measured length which can be mobilized to cover the soft tissue defect. B, The latissimus dorsi muscle was dissected from its origin to 10 cm below its insertion.

**Figure 3. Mobilization and Closure of Latissimus Dorsi Musculocutaneous Flap**



A, The latissimus dorsi muscle was elevated and mobilized with its cutaneous island. B, The latissimus dorsi flap was rotated underneath the bridging skin to cover the left chest wall defect and bronchopleural fistula site. C, The recipient site at immediate postoperative period. D, The donor site at immediate postoperative period.

## Discussion

BPF is a pathologically connected sinus tract between the bronchus and the pleural space which resulted from many causes.<sup>1,2</sup> One of the common and serious causes originates from the incompletely heal of the bronchial stump after pulmonary resection surgery.<sup>4</sup> The incidence of BPF after lung resection surgery varies in many studies which ranges from 4.5% to 28% after pneumonectomy, and approximated 0.5% after lobectomy.<sup>2,4,7</sup>

The classification of the BPF, which depends on 2 factors: onset and size of BPF, reflects the prognosis and management.<sup>2</sup> Firstly, the time of its onset divided into “Early” (from 1 to 7 days after surgery), “Intermediate” (from 8 to 30 days after surgery) and “Late” (more than 30 days after surgery).<sup>2</sup> Secondly, its size which classified into small and large defects. The cutoff point of BPF size is different through each research, commonly use 5 mm<sup>2</sup> as a cutoff point but some use 8 mm.<sup>7</sup>

The early detection is crucially important since the postoperative BPF has high morbidity and mortality. It can be suspected from the patient’s history, physical examination, and operative procedures. In the early BPF, patients usually present with cough, respiratory distress, and signs of systemic inflammatory response syndrome (SIRS) within 7 days after the operation.<sup>2,8,9</sup> The most common causes of the early BPF are surgical technical problems intentionally to secure the stump and procedures that disturb the blood flow to the stump such as proximal dissection and/or extended lymphadenectomy which can cause ischemia and necrosis.<sup>2</sup>

Therefore, the early BPF especially the one that occurs within 3 days postoperative will have a good result after receiving proper treatments. Whereas, the intermediate and late BPFs presents differently because they are usually superimposed by infection. The presentation includes productive cough, SIRS, weight loss, and poor health status.<sup>2,8,9</sup> Patients who tend to develop the intermediate and late BPFs frequently

have some risk factors, for example, advanced age, malnutrition, ongoing pulmonary or pleural infection, and recurrence of malignancy.<sup>2,4,8,9</sup> The general causes are chronic ischemia and malignancy at the bronchial stump. The surgical management of these cases may have disappointing results.<sup>2</sup>

This patient's clinical corresponds to the late BPF with many risk factors including poor nutritional status and ongoing pulmonary infections. Due to a continuing of infectious process in the pleural cavity, left thoracotomy with decortication with open drainage was done, intended to eradicate the infection. Next step, BPF was identified and closed by a direct suture technique, glue, and intercostal muscle flap. We used multimodality of the closure techniques and thoroughly checked the air leak to verify that BPF will completely heal in the future.

The checking process was done orderly. At first, we filled the defect with normal saline solution (NSS) then observed air bubbles when the patient was being ventilated via an endotracheal tube. In consequently, vacuum dressing was applied for 3 weeks in order to taper and shallow up the defect while waiting for the BPF to heal. After that, the air leak was detected recurrently while changing vacuum dressing. Therefore, bronchoscopy detected a 5 mm BPF at the lingular stump. On account of surgical procedures failure in this patient, an endoscopic treatment was introduced as an alternative,<sup>4</sup> by using several varieties of materials such as Fibringluraas and ASD occlude. According to the prior case reports, the BPF was closed successfully by means of these materials and methods.<sup>4</sup> Due to the incessantly recurrent air leak, a Histoacryl glue was applied.

In operative period, after Histoacryl glue was applied, the air leak was disappeared, so the plastic surgeons were consulted for a defect coverage. After 3 days of Histoacryl glue application, LD musculocutaneous flap coverage was done. However, intraoperative air leakage was found, so we decided to use LD musculocutaneous flap because a vascularized muscular flap is able to vanish infections and promote tissue healing. Then soft tissue and skin was used to cover the defect.

Three days postoperatively, there are some mucous discharges permeate to the skin. Sutured materials were removed to evaluate and remove the discharge then debridement was done, and the vacuum dressing was applied and changed every 5 to 7 days. A month later, bronchoscopy was done and found that the size of BPF was smaller, so we introduced Histoacryl glue again to diminish the air leak. Patient health status is improved and symptoms, which are productive cough and dyspnea, are subside.

Postoperative BPF is the challenging and complicated problem to deal with. We have learned several crucial points from this case which we will consider and adjust for other patients' treatments, endeavor the best care in the future. Firstly, we have learned that an early detection of the BPF by using patient's complaint and physical examination is a significant factor that strongly impacts the morbidity and mortality. Secondly, we should use an optimal pressure of vacuum dressing in order to stabilize and maintain the BPF closure materials, in which support its comprehensive healing. However, there are limit studies about usage of vacuum dressing in the BPF in nowadays. Thirdly, after the closure of the BPF is secured and the air leak has not been detected, the vascularized muscular flap coverage should be performed simultaneously or as soon as possible.<sup>3</sup>

Thereby, we suggest that the coverage of the defect should be consecutively done in the same operation after the BPF closure. Finally, the postoperative BPF patients usually confront with other sophisticated medical problems. The intricacy of these problems requires cooperation between health care workers to achieve the best comprehensive care. Therefore, multidisciplinary team approach is indispensable for dealing with the postoperative BPF.

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## Conclusions

Postoperative BPF is the challenging and complicated problem to cope with. Involving with multidisciplinary care team is essential for the best outcome. The early detection

of the BPF and giving the proper management to the patient are essential points to improve the outcomes and quality of life of the patient. This report provides our experiences in intractable postoperative BPF management. Surgical and endoscopic treatments combined with postoperative vacuum dressing encourage patient's symptoms to be subsided.

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## รายงานผู้ป่วยติดเชื้อไมโครแบคทีเรียที่มีภาวะแผลรูเชื่อมระหว่างหลอดเลือดและช่องเยื่อหุ้มปอด หลังการผ่าตัดทรวงอกที่ต่อเนื่องการรักษา โดยใช้การรักษาร่วมกันระหว่างการผ่าตัดซ่อม การส่องกล้องผ่านทางหลอดเลือด การทำแผลด้วยระบบสุญญากาศ และใช้เนื้อเยื่อปะปลูก จากกล้ามเนื้อหลังส่วนบน

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ภาวะแผลรูเชื่อมระหว่างหลอดเลือดและช่องเยื่อหุ้มปอดเป็นภาวะที่มีทางเชื่อมผิดปกติระหว่างหลอดเลือดกับช่องเยื่อหุ้มปอด สาเหตุส่วนใหญ่ที่พบได้บ่อยนั้นเกิดมาจากการผ่าตัดทรวงอก จึงถือได้ว่าเป็นภาวะแทรกซ้อนที่สำคัญอย่างหนึ่ง ผู้ป่วยที่มีภาวะแผลรูเชื่อมระหว่างหลอดเลือดและช่องเยื่อหุ้มปอดนี้ มีอัตราเจ็บป่วยจนทำให้เกิดภาวะทุพพลภาพ และเสียชีวิตที่ค่อนข้างสูง ดังนั้น เมื่อภาวะนี้เกิดขึ้นแล้วจะส่งผลกระทบต่อคุณภาพชีวิตของผู้ป่วย และครอบครัวเป็นอย่างมาก การรักษาในปัจจุบันถือเป็นเรื่องที่ซับซ้อน ทำหาย และต้องอาศัยความร่วมมือในการดูแลผู้ป่วย จากสหสาขาวิชาชีพ ทั้งศัลยแพทย์ อายุรแพทย์ พยาบาล และนักกายภาพบำบัด เพื่อให้ผลการรักษาเป็นไปตามที่คาดหวัง บทความนี้นำเสนอรายงานผู้ป่วย เพื่อเป็นแนวทางการรักษาผู้ป่วยที่มีภาวะแผลรูเชื่อมระหว่างหลอดเลือดและช่องเยื่อหุ้มปอดที่เกิดขึ้นจากการผ่าตัดปอดที่ล้มเหลวจากการรักษาด้วยวิธีการผ่าตัดซ่อม และการรักษาด้วยวิธีการส่องกล้องผ่านทางหลอดเลือดเพื่อแก้ไขบริเวณที่มีแผลรูเชื่อม ตัวอย่างผู้ป่วยรายนี้เป็นผู้ป่วยหญิง อายุ 56 ปี มาโรงพยาบาลด้วยอาการไอเป็นเลือดสดจำนวนไม่มากแต่บ่อยครั้งร่วมกับมีไข้ต่ำๆ เป็นครั้งคราว และน้ำหนักตัวลดลงอย่างมีนัยสำคัญ ได้รับการวินิจฉัยว่าพบการติดเชื้อไมโครแบคทีเรียที่ปอดชนิดคอตีบ เริ่มต้นได้รับการรักษาด้วยยาพบว่า อาการและผลตรวจรังสีทรวงอกไม่ดีขึ้น ต่อมาจึงได้รับการรักษาโดยการผ่าตัดส่องกล้อง เพื่อตัดเนื้อปอดส่วนที่ติดเชื้อ หลังการผ่าตัดตรวจพบภาวะแผลรูเชื่อมระหว่างหลอดเลือดและช่องเยื่อหุ้มปอด ผู้ป่วยได้รับการรักษาต่อด้วยวิธีการผ่าตัดซ่อมและการส่องกล้องผ่านทางหลอดเลือดแต่ยังพบภาวะแผลรูเชื่อมระหว่างหลอดเลือดและช่องเยื่อหุ้มปอดกลับมาเกิดซ้ำ จึงได้มีการใช้วิธีการรักษาหลากหลายแบบร่วมกันในคราวเดียว ทั้งการผ่าตัดซ่อม การรักษาด้วยวิธีการส่องกล้องผ่านทางหลอดเลือด การใช้เนื้อเยื่อปะปลูกจากกล้ามเนื้อหลังส่วนบน รวมถึงการทำแผลด้วยระบบสุญญากาศ หลังการรักษา 1 เดือน อาการของผู้ป่วยดีขึ้นมาก ตรวจไม่พบแผลรูเชื่อมระหว่างหลอดเลือดและช่องเยื่อหุ้มปอด โดยสรุปแล้วการใช้การรักษาร่วมกันข้างต้นช่วยให้อาการผิดปกติหายไปมากที่สุด

**คำสำคัญ:** ภาวะแผลรูเชื่อมระหว่างหลอดเลือดและช่องเยื่อหุ้มปอด ภาวะติดเชื้อไมโครแบคทีเรีย การผ่าตัดทรวงอก เนื้อเยื่อปะปลูก การทำแผลด้วยระบบสุญญากาศ ความผิดปกติของทรวงอก

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