

Received: 2020.03.12  
 Accepted: 2020.05.08  
 Available online: 2020.06.08  
 Published: 2020.07.27

e-ISSN 1941-5923  
 © Am J Case Rep, 2020; 21: e924245  
 DOI: 10.12659/AJCR.924245

# Development of Bronchopleural Fistula Complicated by Empyema Fifteen Years After Right Lower Lobe Lobectomy: A Case Report

Authors' Contribution:  
 Study Design A  
 Data Collection B  
 Statistical Analysis C  
 Data Interpretation D  
 Manuscript Preparation E  
 Literature Search F  
 Funds Collection G





ABCDEF 1 **Luc M. Fortier\***  
 ABCDEF 2 **Vaishnavi Raman\***  
 ADE 2 **Daniel A. Grove**

1 Georgetown University School of Medicine, Washington, DC, U.S.A.  
 2 MedStar Medical Group Internal Medicine, MedStar Good Samaritan Hospital, Baltimore, MD, U.S.A.

\* Luc M. Fortier and Vaishnavi Raman contributed equally

**Corresponding Author:** Luc M. Fortier, e-mail: [lucmfortier@gmail.com](mailto:lucmfortier@gmail.com)  
**Conflict of interest:** None declared

<b>Patient:</b>	<b>Male, 55-year-old</b>
<b>Final Diagnosis:</b>	<b>Bronchopleural fistula • empyema</b>
<b>Symptoms:</b>	<b>Back pain • productive cough • shortness of breath</b>
<b>Medication:</b>	—
<b>Clinical Procedure:</b>	<b>Bronchoscopy • chest wall resection • laparotomy • omentectomy • thoracentesis • thoracotomy</b>
<b>Specialty:</b>	<b>Microbiology and Virology • Pulmonology • Surgery</b>
<b>Objective:</b>	<b>Unusual clinical course</b>
<b>Background:</b>	Bronchopleural fistula formation is a rare complication of lobectomy surgery, with a frequency reported ranging from 0.5% to 1%. A post-lobectomy bronchopleural fistula usually presents within 14 days of surgery. To our knowledge, it is extremely rare for a bronchopleural fistula to develop many years after an operation.
<b>Case Report:</b>	We present the case of a 55-year-old male smoker with history of a right lower lobe lobectomy 15 years prior who presented to the Emergency Department with complaints of worsening back pain, shortness of breath, and cough productive of sputum. He was found to have a right bronchopleural fistula with right-sided empyema. He was taken to the operating room a few days after initial admission for right thoracoscopic chest exploration, right chest debridement, right chest wall resection, and window procedure with creation of pleurocutaneous fistula. Ultimately, he required a right completion pneumonectomy and buttress of bronchial stump with transdiaphragmatic omental flap.
<b>Conclusions:</b>	We diagnosed a rare case of post-lobectomy bronchopleural fistula complicated by an empyema that demonstrates bronchopleural fistulas can appear 15 years postoperatively and present with subacute clinical signs and symptoms.
<b>MeSH Keywords:</b>	<b>Bronchial Fistula • Empyema • Postoperative Complications</b>
<b>Full-text PDF:</b>	<a href="https://www.amjcaserep.com/abstract/index/idArt/924245">https://www.amjcaserep.com/abstract/index/idArt/924245</a>

 1576  —  —  22



## Background

Bronchopleural fistula formation is a rare complication of lobectomy surgery, with a frequency reported ranging from 0.5% to 1% [1]. A post-lobectomy bronchopleural fistula usually presents within 14 days of surgery. Generally, a fistula is suspected when the patient develops acute symptoms of dyspnea, chest pain, hemodynamic instability, and subcutaneous emphysema recently after surgery [2]. To our knowledge, it is rare for a bronchopleural fistula to develop many years after an operation. We describe a case of a bronchopleural fistula complicated by an empyema that developed approximately 15 years after a right lower lobe lobectomy.

## Case Report

A 55-year-old male smoker with a history of recurrent lung abscesses status post right lower lobe lobectomy 15 years prior presented to our hospital 3 days after an approximately 11-hour drive with complaints of worsening back pain, shortness of breath, and cough productive of sputum. The pain was predominantly over the right lower back, dull and achy in nature, worsened by activity, and relieved with bed rest. There was no trauma, radiation, or associated tingling/numbness.

He endorsed some degree of shortness of breath at baseline, but it had increased substantially over the 3 days prior to admission and limited his exertion after walking 20 feet. He had a cough productive of white phlegm but no blood. He endorsed night sweats over the prior 1–2 months and a 4.54 kg (10-pound) weight loss within the prior 2 weeks. He denied fevers or chills.

In the Emergency Department, he was afebrile but hypotensive with mean arterial pressures (MAPs) between 50 to 60 mmHg. He was saturating 92% to 95% on room air. Laboratory tests were notable for a leukocytosis of 20 300/mm<sup>3</sup> with neutrophil predominance and a left shift. High resolution computed tomography (CT) showed a large right-sided hydropneumothorax, several nodular opacities throughout left lung, scattered inflammatory sequelae in the left lung, and aspirated content within the central airways.

He received morphine for pain control and a 2 L bolus of normal saline. Empiric treatment with vancomycin and piperacillin/tazobactam were initiated. He was admitted to the comprehensive cardiopulmonary care unit for further workup and management based on the radiographic CT findings.

On hospital day 2, thoracentesis showed fluid consistent with a right-sided empyema. Pleural fluid analysis showed a white blood cell count of 170 000/mm<sup>3</sup> (neutrophils 80%),

pH 6.8, and lactate dehydrogenase (LDH) >200 U/L. An intra-thoracic pigtail was placed and later upsized to a 24-French chest tube. Cultures from the fluid grew methicillin-sensitive *Staphylococcus aureus* (MSSA) and antibiotics were changed to ampicillin/sulbactam. He had a persistent air leak in the collection chamber. A bronchoscopy with barium fluoroscopy was performed and demonstrated a bronchopleural fistula into the right main stem bronchus.

A few days after initial admission, he was taken to the operating room for right thoracoscopic chest exploration. Approximately 0.5 L of frank pus was aspirated from the chest cavity when it was first entered. In addition, inspection of the chest cavity revealed frankly necrotic tissue on the chest wall with multiple adhesions which would have precluded packing. There was no viable lung noted, though there was a limited view of the chest cavity given the extent of necrotic debris. Ultimately, a window procedure with creation of pleurocutaneous fistula was performed.

Over the following weeks, due to persistent air leaks, a video thoracoscopic exploration was performed and revealed necrosis of the entire right lung with a Swiss cheese-like consistency and air leaks throughout its entire surface. At this point, it was clear that the lung was a chronic nidus for infection and would ultimately lead to the patient's demise, with the only option being a completion pneumonectomy. Consequently, he underwent a right thoraco-sternotomy, right extra-pleural completion pneumonectomy, partial parietal pleurectomy, laparotomy with jejunostomy tube placement to support nutrition, and harvest of pedicled omental flap to buttress bronchial stump with transdiaphragmatic omental flap. Unfortunately, during the post-operative period and rehabilitation phase, the patient developed recurrent chest wall infections and has been hospitalized multiple times requiring repeated debridements and continued use of the irrigating vac.

## Discussion

The most common occurrence of bronchopleural fistula is as a postoperative complication after a pneumonectomy or lobectomy [1]. By removing the entire lung or a lobe a conduit may be created between the end of the bronchus and the pleural space. This connection can be surgically repaired via multiple techniques including mechanical closure with staples or manual closure with sutures [3]. Rarely, this closure fails to heal and creates a fistula. This exposes the clean pleural space to the normal flora of the bronchus, which can lead to an empyema or aspiration pneumonia with significant morbidity [4].

Factors that increase the risk of fistula formation post-pneumonectomy or lobectomy include right-sided surgery, neoadjuvant

chemotherapy and radiation therapy, diabetes mellitus, heavy smoking, COPD, low nutritional status, poor wound healing, previous ipsilateral thoracotomy, residual tumor at the bronchial stump, extensive lymph node dissection, age greater than 60 years, and prolonged postoperative mechanical ventilation [4–8]. This patient's history was significant for right-sided surgery, smoking exposure, and COPD, which put him at a higher risk of developing this complication. However, the current literature describes that postoperative bronchopleural fistulas occur most frequently within 8 to 12 days after surgery and almost always within 14 days [2]. This patient's presentation 15 years after his surgery is quite unusual and likely secondary to a rarer etiology.

Rarer causes of bronchopleural fistula documented in the literature include complications of chemotherapy or radiotherapy for lung cancer treatment, pulmonary infection (bacterial, tuberculous, or fungal), or inflammatory reactions in the lung parenchyma that extend into the pleura [9,10]. This patient denied any history of chemotherapy or radiation therapy. The fluid culture drained from this patient's pleura grew MSSA, consistent with a bacterial infection that we suspect either necrotized the bronchus to create a bronchopleural fistula or was introduced into the pleura secondary to a bronchopleural fistula that was already formed.

There is a wide range of clinical presentations for a bronchopleural fistula, depending on the etiology. Patients developing a bronchopleural fistula within 2 weeks of resection surgery typically present with acute symptoms of dyspnea, chest pain, hemodynamic instability, and subcutaneous emphysema. Conversely, patients who develop a bronchopleural fistula greater than 2 weeks postoperatively or secondary to an infectious process usually present with subacute symptoms of fever, malaise, muscle wasting, and productive cough [2].

This patient's presentation in the delayed postoperative period (15 years postoperatively) with productive cough, leukocytosis, and new air fluid levels on imaging suggests an infectious process likely lead to the development of a bronchopleural fistula. The most common microbes responsible for pleural infections are viridans streptococci (25%) and *Staphylococcus aureus* (18%) [11]. Pleural fluid analysis yielding cultures growing MSSA, which is consistent with our suspicion that an initial infectious etiology resulted in a fistula.

The initial management of any bronchopleural fistula, early or delayed, is drainage of any pleural air or fluid with a chest tube [12]. Pleural fluid should be sent to the laboratory for complete blood count (CBC), pH, total protein, LDH, glucose, cytology, triglycerides, Gram stain, and culture. Patients should be started on broad-spectrum antibiotics to cover for

Gram-positive, Gram-negative, and anaerobic microorganisms until bacterial sensitivities are available [9]. Patients who present with delayed post-lobectomy bronchopleural fistula or those who develop a fistula secondary to infectious pleuropulmonary disease are managed initially with medical treatment. This includes dependent drainage, reduction of the pleural space, antibiotic treatment, nutritional supplementation, and, if needed, ventilator management [13]. Closure of the fistula is then performed with either airway stents, coils, occlusive agents, or Amplatzer devices [14,15]. Patients should then be monitored for clinical signs of fistula recurrence, air in the chest tube, and imaging of pleural space to ensure closure of the defect [12]. The options for patients who fail initial surgical or bronchoscopic repair include repeat surgery, an alternative bronchoscopic method, or an open window thoracostomy with either Eloesser flap or a Claggett window [16]. More recently, an accelerated treatment of a post-lobectomy or post-pneumectomy empyema that avoids an open chest window has gained popularity. First described in a case series in 2001, the procedure entails repeated open debridements, a negative pressure continuous suction wound therapy of the temporarily closed chest cavity filled with povidone-iodine-soaked dressings, and systemic antimicrobial therapy [17]. Once the chest cavity is macroscopically clean, the pleural space is filled with antibiotic solution and the bronchopleural fistula is subsequently repaired with muscle flap or omentum. A study of 75 patients undergoing accelerated therapy showed clinical success and safety with this technique, with 94.6% of patients' chests definitively closed within 8 days [18]. Nonetheless, experts agree that this procedure should not be attempted in cases with chronic empyema or complicated by a bronchopleural fistula, in which case an open window thoracostomy procedure is recommended [18–22].

During surgery, our patient's chest was entered with a video thoracoscope and revealed approximately 0.5 L of pus with clear evidence of necrotic chest wall tissue and no viable lung tissue. Given these findings, closure of the bronchopleural fistula was deemed insufficient. Due to the possible chronicity of the empyema and the clear presence of a bronchopleural fistula, the surgeon felt that an open window thoracostomy with a Claggett window was necessary to ensure a clean chest cavity and decreased risk of recurrence.

## Conclusions

We diagnosed a rare case of post-lobectomy bronchopleural fistula complicated by an empyema that demonstrates bronchopleural fistulas can appear 15 years postoperatively and present with subacute clinical signs and symptoms.

## References:

1. Alpert JB, Godoy MCB, deGroot PM et al: Imaging the post-thoracotomy patient. anatomic changes and postoperative complications. *Radiol Clin North Am*, 2014; 52(1): 85–103
2. Lois M, Noppen M: Bronchopleural fistulas: An overview of the problem with special focus on endoscopic management. *Chest*, 2005; 6(128): 3955–65
3. Uçvet A, Gursoy S, Sirzai S et al: Pulmonary bronchial closure methods and risks for bronchopleural fistula in pulmonary resections: How a surgeon may choose the optimum method? *Interact Cardiovasc Thorac Surg*, 2011; 12: 558–62
4. Okuda M, Go T, Yokomise H: Risk factor of bronchopleural fistula after general thoracic surgery: Review article. *Gen Thorac Cardiovasc Surg*, 2017; 65(12): 679–85
5. Wright CD, Wain JC, Mathisen DJ et al: Postpneumonectomy bronchopleural fistula after sutured bronchial closure: Incidence, risk factors, and management. *J Thorac Cardiovasc Surg*, 1996; 112(5): 1367–71
6. Farkas EA, Detterbeck FC: Airway complications after pulmonary resection. *Thorac Surg Clin*, 2006; 16(3): 243–51
7. Li SJ, Zhou XD, Huang J et al: A systematic review and meta-analysis-does chronic obstructive pulmonary disease predispose to bronchopleural fistula formation in patients undergoing lung cancer surgery? *J Thorac Dis*, 2016; 8(7): 1625–38
8. Toufektzian L, Patris V, Sepsas E, Konstantinou M: Does postoperative mechanical ventilation predispose to bronchopleural fistula formation in patients undergoing pneumonectomy? *Interact Cardiovasc Thorac Surg*, 2015; 21(3): 379–82
9. Mao R, Ying P-Q, Xie D et al: Conservative management of empyema-complicated post-lobectomy bronchopleural fistulas: Experience of consecutive 13 cases in 9 years. *J Thorac Dis*, 2016; 8(7): 1577–86
10. Duarte-Ribeiro F, Dias C, Mota M: Bronchopleural and pleurocutaneous fistula in HIV patient with pulmonary tuberculosis. *ID Cases*, 2017; 9: 82–84
11. Meyer CN, Rosenlund S, Nielsen J, Friis-Møller A: Bacteriological aetiology and antimicrobial treatment of pleural empyema. *Scand J Infect Dis*, 2011; 43(3): 165–69
12. Salik I, Abramowicz AE: Bronchopleural fistula. In: *StatPearls*. Treasure Island (FL): StatPearls Publishing, 2020
13. Baumann MH, Sahn SA: Medical management and therapy of bronchopleural fistulas in the mechanically ventilated patient. *Chest*, 1990; 97(3): 721–28
14. Fruchter O, El Raouf BA, Abdel-Rahman N et al: Efficacy of bronchoscopic closure of a bronchopleural fistula with Amplatzer devices: Long-term follow-up. *Respiration*, 2014; 87(3): 227–33
15. Battistoni P, Caterino U, Batzella S et al: The use of polyvinyl alcohol sponge and cyanoacrylate glue in the treatment of large and chronic bronchopleural fistulae following lung cancer resection. *Respiration*, 2017; 94(1): 58–61
16. West D, Togo A, Kirk AJB: Are bronchoscopic approaches to post-pneumonectomy bronchopleural fistula an effective alternative to repeat thoracotomy? *Interact Cardiovasc Thorac Surg*, 2007; 6(4): 547–50
17. Schneider D, Cassina P, Korom S et al: Accelerated treatment for early and late postpneumonectomy empyema. *Ann Thorac Surg*, 2001; 72(5): 1668–72
18. Schneider D, Grodzki T, Lardinois D et al: Accelerated treatment of postpneumonectomy empyema: A binational long-term study. *J Thorac Cardiovasc Surg*, 2008; 136(1): 179–85
19. Wain JC: Management of late postpneumonectomy empyema and bronchopleural fistula. *Chest Surg Clin N Am*, 1996; 6(3): 529–41
20. Zaheer S, Allen MS, Cassivi SD et al: Postpneumonectomy empyema: Results after the claggett procedure. *Ann Thorac Surg*, 2006; 82(1): 279–87
21. Deschamps C, Allen MS, Miller DL et al: Management of postpneumonectomy empyema and bronchopleural fistula. *Semin Thorac Cardiovasc Surg*, 2001; 13(1): 13–19
22. Wong PS, Goldstraw P: Post-pneumonectomy empyema. *Eur J Cardiothoracic Surg*, 1994; 8(7): 345–49