



Case report

Management of esophagopulmonary fistula after gunshot – case report and literature review

**Kajetan Kielbowski¹ , Estera Bakinowska¹, Michał J. Kubisa², Janusz Wójcik²,
Bartosz Kubisa² **

¹ Student Scientific Society, Pomeranian Medical University, Szczecin, Poland

² Department of Thoracic Surgery and Transplantation, Pomeranian Medical University, Szczecin, Poland

ARTICLE INFO

Article history

Received: May 13, 2021

Accepted: November 18, 2021

Available online: December 27, 2021

Keywords

Esophagopulmonary fistula

Aerodigestive fistula

Benign esophageal fistula

Doi

<https://doi.org/10.29089/2021.21.00207>

User license

This work is licensed under a
Creative Commons Attribution –
NonCommercial – NoDerivatives
4.0 International License.



ABSTRACT

Introduction: An esophagorespiratory fistula is a pathological communication between the esophagus and respiratory tract. The most common type is a communication with the trachea, while the least common is with lung parenchyma. These fistulas are classified as congenital or acquired while etiology is benign or malignant.

Aim: We present a case report of a patient who developed esophagopulmonary fistula several years after gunshot in the right side of the chest. Additionally, we discuss the treatment methods and compare the outcomes with other case studies and analyses from world literature.

Case study: A 48-year-old male patient was admitted to the Department of Thoracic Surgery and Transplantation due to bleeding from the respiratory tract. Radiological images revealed a fistula between the esophagus and right lung parenchyma. Furthermore, bronchiectasis in the right lung was found. Tissues of the fistula, right middle and lower lobes were resected. The patient required renewed hospitalization due to pleural empyema. Furthermore, recurrence of the fistula was observed.

Results and discussion: Treatment of esophagorespiratory fistula depends on the etiology and location of the pathological communication. Surgery provides the best possible outcomes in patients with a benign fistula. Proximal location requires cervicotomy, while distal location a thoracotomy. In addition, a distal fistula may damage lung parenchyma.

Conclusions: A long-term distal fistula may require pulmonary resection, but early diagnosis would help to avoid more invasive procedures. As symptoms are non-specific and benign etiology is not frequent, thorough examination in search of malignancy is required.

1. INTRODUCTION

An aerodigestive fistula is a rare pathological communication between the respiratory and gastrointestinal tracts. There are three general types that can be distinguished: esophagotracheal (ETF), which is the most common,¹ esophagobronchial (EBF) and esophagopulmonary (EPF). These communications are usually neoplastic, traumatic or congenital in origin.² Malignant cause of the fistula seems to be the most frequent.³ Direct communications between the esophagus and lung parenchyma are rarely reported.

2. AIM

We present the case of a patient who developed esophagopulmonary fistula due to gunshot in the right side of the chest. Management of the fistula is described with review of analyses from world literature.

3. CASE REPORT

In 1990, an 18-year-old male was shot in the right side of the chest. Wedge resection of the damaged lung parenchyma was performed. Since 2013, the patient had been experiencing hemoptysis and choking in recumbency; however, lying on the right side would provide improvement. The patient was admitted to the Department of Thoracic Surgery and Transplantation on October 5, 2020 due to bleeding from the respiratory tract. Bronchofiberscopy revealed active bleeding from the right middle lobe. On October 6, a chest X-ray with esophageal contrast was performed (Figure 1) and EPF in the lateral wall of lower $\frac{1}{3}$ of the esophagus was found. Furthermore, a chest CT revealed that the fistula had expanded to the X segment of the right lung (Figure 1). Above the fistula, the esophagus was broadened by 2 cm, whereas in the IX and X segments peribronchial infiltration connecting the lung hilum and thickened pleura of the IX and X segments were found. Enlarged lymph nodes in the right hilum, fibrosis and bronchiectasis in the middle lobe and apex were observed as well. Thus, on October 8, 2020, a right lower bilobectomy with suturing of the esophagus was performed, resecting the right VII rib. Additionally, the intermedial bronchus, lower pulmonary vein, middle lobular vein and middle lobe artery were dissected using a stapler, and the lower and middle lobes were removed. Diverticulum of the esophagus was dissected in the basis; the esophagus was sewn using absorbable vicryl sutures. Then, tissues of the fistula were removed and a lymphadenectomy of the mediastinum was performed. Furthermore, decortication of the upper right lobe was performed. Lastly, the pleural cavity was cleansed. In the histopathological examination, features of pneumoconiosis were found in the lymph nodes. In addition, fibrosis of the pleura was revealed. There were no postoperative complications and the patient was discharged from hospital in a good general condition. There were no

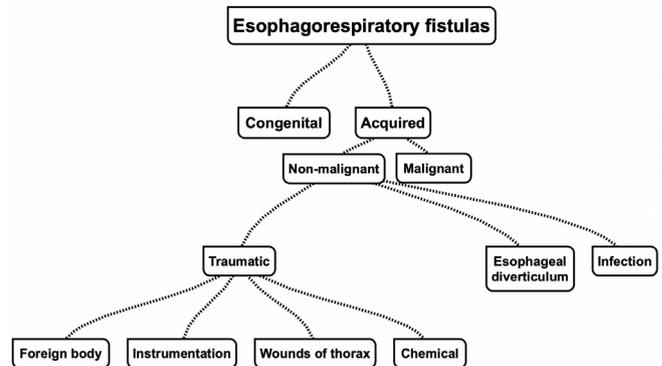


Figure 1. Etiologies of esophago-tracheobronchial fistulas. incidents of hemoptysis and the patient was again able to lie on his back without choking.

On November 3, 2020, the patient was admitted at the hospital once again, as he had been experiencing weakness and fever for four days. Elevation of C-reactive protein and fibrinogen was observed. A chest X-ray revealed fluid and gas bubbles in the right pleural cavity (Figure 2). Consequently, on November 5, 2020, a right thoracotomy (10 cm length) above the VIII rib was carried out, removing 8 cm of the mentioned rib. Also, pleural empyema with features of pulmonary-pleural fistula was found. Subsequently, the pleural cavity was cleansed and drainage was applied. After several weeks, a recurrence of the esophageal fistula was observed (Figure 3), but it closed spontaneously. According to the patient, his condition largely improved following the procedure. The unpleasant symptoms, including cough and choking, were no longer present.

4. RESULTS AND DISCUSSION

It is said that the first report of pathological communication between the respiratory and digestive tracts was presented in 1696 by Gibson.^{4,5} The first report of acquired ETF was published in 1844 by McDonnell.⁶ However, in 1737, a report of EBF with unknown etiology was published.⁷ Etiologies of this pathological communication are presented on Scheme 1, and they are based on the classification developed by Coleman and Bunch in 1950.⁸ In 1954, HS Abrams broadened this list with broncholithiasis, aneurysm and esophagomalacia.⁹ In the presented case, the fistula developed between the esophagus and lung parenchyma as a result of a thorax wound (gunshot). Surgery is considered the best treatment strategy for benign esophagorespiratory fistula (ERF), but these pathological communications still represent curative problems.

Treatment of the fistula depends on many factors. Firstly, it is crucial to verify if the etiology of the fistula is malignant or benign. Surgery is the typical treatment for benign ERF, while treatment of malignant fistulas depends on the patient's condition and often involves esophageal stents.¹⁰ Secondly, the location of the pathological communication between the esophagus and respiratory tract is significant,

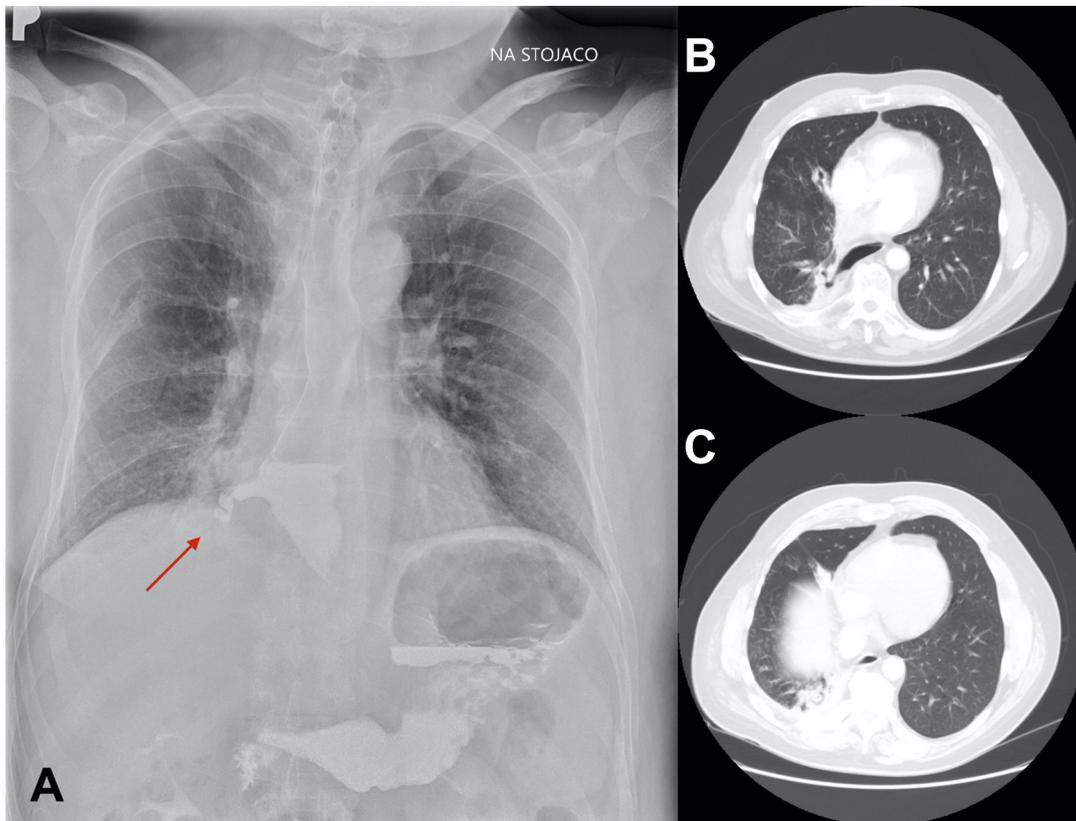


Figure 2. Radiological images of the patient before first operation (October 6, 2020): (A) Chest X-ray with contrast. Diverticulum of the esophagus with esophagopulmonary fistula (red arrow); (B) CT-broadened esophagus above the fistula; (C) CT - bronchiectasis and inflammation in the right lung.

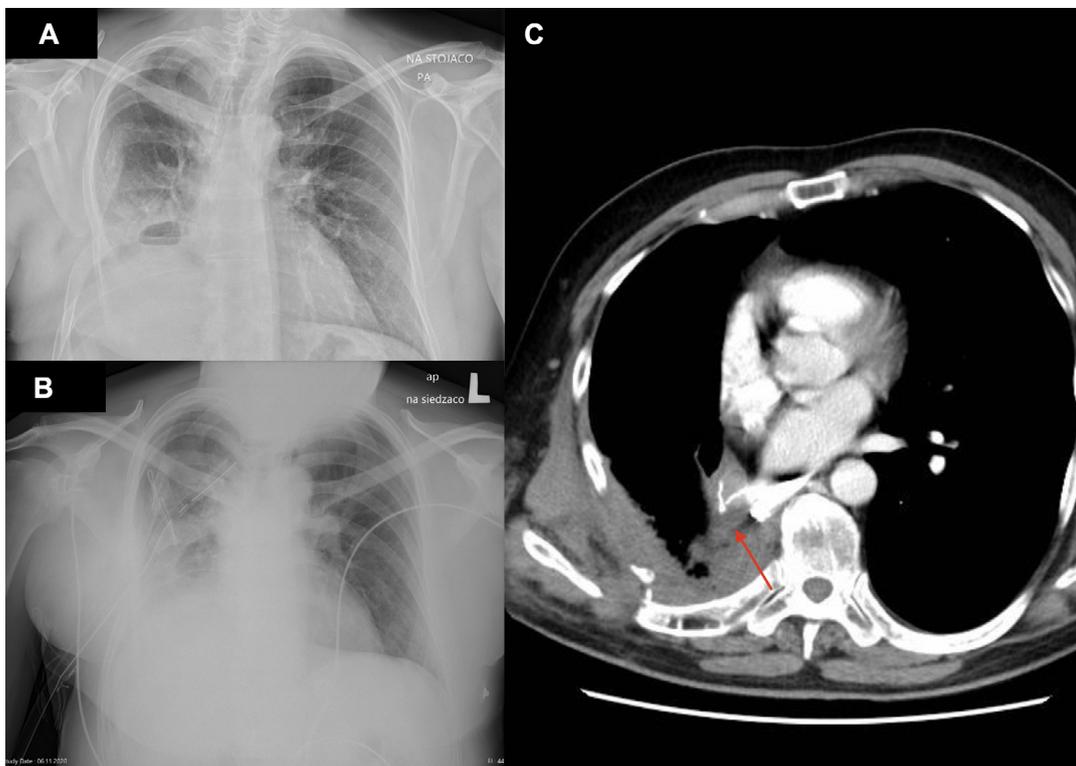


Figure 3. Radiological images of the patient: (A) Chest X-ray before second surgery (November 3, 2020); (B) Chest X-ray after second surgery (November 6, 2020); (C) CT with contrast – recurred fistula (red arrow, January 14, 2021).

as the fistula in the upper part of the esophagus requires a cervical approach (cervicotomy), while a thoracotomy is necessary for distal fistulas.¹¹ However, the latter type might damage lung parenchyma. Consequently, as a result of bronchiectasis, pneumonitis or abscesses, together with fistulectomy, a lung parenchyma resection would be required.^{8,11} Depending on the size of damage, segments, lobes or a whole lung may be resected.^{12,13} In the presented case, the two right lower lobes with tissues of the fistula were removed. Apart from fistulectomy, there are two other surgical methods commonly used. Fistula repair includes usage of mediastinal or pleural tissues to cover both endings of the divided fistula. Interposition of the muscle flaps is not frequently described, as it is more often used to treat bronchopleural fistulas.¹⁴ Exclusion and bypass of fistulous fragments of the esophagus is another strategy.¹¹ Non-surgical treatment of benign ERFs is not popular, but possible in the case of poor condition of the patient.¹⁵ Furthermore, spontaneous closure of the fistula might also be observed during treatment of an underlying disease.¹⁶ In general, symptoms are not characteristic. The intensity of symptoms depends on the size of the fistula as well. Thus, the fistula might be present for months or years before diagnosis is made. However, Ono's sign is commonly seen in patients with esophagobronchial fistulas. It involves paroxysmal cough, especially after liquid intake.¹⁷ Heller's myotomy is usually performed in patients with EPF due to achalasia, but these cases are not frequently reported. This treatment involves longitudinal incision of the anterior wall of esophagus.¹⁸ In the presented case, renewed hospitalization one month after primary procedure was required due to pleural empyema. It is caused by infection of pleural cavity, and etiology involves pneumonia, thoracic trauma and surgery (e.g., postpneumonectomy empyema). Treatment is focused on infection control, and drainage of pus and fluid.¹⁹

In an observational study performed by Sersar SI et al, out of 27 cases of aerodigestive fistulas, there were only 2 patients with EPF. The etiology of these pathological communications was malignant (squamous cell carcinoma of the esophagus). The fistulas were resected with left lower lobectomies.²⁰ In an analysis carried out by Mangi AA et al., out of 228 cases of EBF, 215 were malignant. Thirteen benign cases developed due to prior esophageal surgery, infection, foreign body ingestion and esophageal diverticulum. The majority of patients underwent fistula repair with interposition of intercostal muscle flap, omentum, thickened pleura or pericardial fat.²¹ In an analysis performed in 2007 by Kim et al., 14 patients underwent surgery for benign EBF. Fistulas were primarily repaired with parietal pleura interposition. In one patient, a leakage at the repair site caused empyema.²²

5. CONCLUSIONS

Benign esophagopulmonary fistulas are exceptionally rare communications between the esophagus and lung parenchyma. Thus, thorough examination in search of malignancy is required.

A long-term distal fistula may damage lung parenchyma. Consequently, early diagnosis is crucial in order to avoid pulmonary resection.

Conflict of interest

None declared.

Funding

None declared.

References

- 1 Qureshi YA, Muntzer Mughal M, Fragkos KC et al. Acquired Adult Aerodigestive Fistula: Classification and Management. *J Gastrointest Surg.* 2018;22(10):1785–1794. <https://doi.org/10.1007/s11605-018-3811-0>.
- 2 Herbella FA, Del Grande JC. Benign esophagopulmonary fistula through an epiphrenic diverticulum and asymptomatic achalasia. *Dig Dis Sci.* 2010;55(4):1177–1178. <https://doi.org/10.1007/s10620-010-1136-2>.
- 3 Shin JH, Kim JH, Song HY. Interventional management of esophagorespiratory fistula. *Korean J Radiol.* 2010;11(2):133–140. <https://doi.org/10.3348/kjr.2010.11.2.133>
- 4 Dumont A, Duprez A, Wittek F. Acquired and congenital oesophago-bronchial fistulas. *Thorax.* 1956;11(3):249–256. <http://dx.doi.org/10.1136/thx.11.3.249>.
- 5 Lansden FT, Falor WH. Congenital esophagorespiratory fistula in the adult. *J Thorac Cardiovasc Surg.* 1960;39:246–251.
- 6 Gupta RL, Banerjee T. Traumatic esophagotracheal fistula. *Am J Surg.* 1961;101:227–229. [https://doi.org/10.1016/0002-9610\(61\)90758-9](https://doi.org/10.1016/0002-9610(61)90758-9).
- 7 Chervenikov A, Tzekov C, Grigorov GE, Chervenikov P. Acquired benign esophago-airway fistulas. *Eur J Cardiothorac Surg.* 1996;10(9):713–716. [https://doi.org/10.1016/S1010-7940\(96\)80329-0](https://doi.org/10.1016/S1010-7940(96)80329-0).
- 8 Coleman FP, Bunch GH Jr. Acquired non-malignant esophago-tracheobronchial communication. *Dis Chest.* 1950;18(1):31–48. <https://doi.org/10.1378/chest.18.1.31>
- 9 Abrams HS. Esophagorespiratory fistulae. *AMA Arch Otolaryngol.* 1954;60(3):371–374. <https://doi.org/10.1001/archotol.1954.00720010380011>.
- 10 Zechowicz M, Pietroczyk D, Roszkowski M, Zechowicz T. Treatment difficulties of malignant esophagorespiratory fistula: Case report of a 56-year-old patient with esophageal cancer. *Pol Ann Med.* 2016;23(2):177–181. <https://doi.org/10.1016/j.poamed.2016.04.004>.
- 11 Qureshi YA, Muntzer Mughal M, Markar SR et al. The surgical management of non-malignant aerodigestive fistula. *J Cardiothorac Surg.* 2018;13(1):113–121. <https://doi.org/10.1186/s13019-018-0799-1>.
- 12 Braimbridge MV, Keith HI. Oesophago-bronchial fistula in the adult. *Thorax.* 1965;20(3):226–233. <http://dx.doi.org/10.1136/thx.20.3.226>.
- 13 Issaka A, Ermerak NO, Kara VH, Lerut T, Batirel HF. Two unusual cases of adult onset congenital bronchoesophageal fistulas treated with fistula division. *Ann Thorac Surg.* 2014;97(2):685–687. <https://doi.org/10.1016/j.athoracsurg.2013.06.111>.

- ¹⁴ Hammoudeh ZS, Gursel E, Baciewicz FA Jr. Split latissimus dorsi muscle flap repair of acquired, nonmalignant, intrathoracic tracheoesophageal and bronchoesophageal fistulas. *Heart Lung Circ.* 2015;24(6):75–78. <https://doi.org/10.1016/j.hlc.2014.12.166>.
- ¹⁵ Marco C, Doncel F, Veloso E, Viver JM, Vidal J. Non-surgical closure of a benign oesophagobronchial fistula. *Br J Surg.* 1987;74(5):415. <https://doi.org/10.1002/bjs.1800740532>.
- ¹⁶ Wigley FM, Murray HW, Mann RB, Saba GP, Kashima H, Mann JJ. Unusual manifestation of tuberculosis: TE fistula. *Am J Med.* 1976;60(2):310–314. [https://doi.org/10.1016/0002-9343\(76\)90443-5](https://doi.org/10.1016/0002-9343(76)90443-5).
- ¹⁷ Patel S, et al., Acquired spontaneous bronchoesophageal fistula in an adult, Egypt. *J Chest Dis Tuberc.* 2014;64(1):209–211. <https://doi.org/10.1016/j.ejcdt.2014.09.007>.
- ¹⁸ Zhu J, Ni Y, Lu Q, Li X, Wang W, Wang H. Benign esophago-pulmonary fistula complicating achalasia: case report and literature review. *J Thorac Dis.* 2015;7(4):92–96. <https://doi.org/10.3978/j.issn.2072-1439.2015.04.03>.
- ¹⁹ Reichert M, Hecker M, Witte B et al. Stage-directed therapy of pleural empyema. *Langenbecks Arch Surg.* 2017;402(1):15–26. <http://doi.org/10.1007/s00423-016-1498-9>.
- ²⁰ Sersar SI, Maghrabi LA. Respiratory-digestive tract fistula: two-center retrospective observational study. *Asian Cardiovasc Thorac Ann.* 2018;26(3):218–223. <https://doi.org/10.1177/0218492318755013>.
- ²¹ Mangi AA, Gaissert HA, Wright CD et al. Benign broncho-esophageal fistula in the adult. *Ann Thorac Surg.* 2002;73(3):911–915. [https://doi.org/10.1016/s0003-4975\(01\)03582-2](https://doi.org/10.1016/s0003-4975(01)03582-2).
- ²² Kim HK, Choi YS, Kim K, Kim J, Shim YM. Long-term results of surgical treatment in benign bronchoesophageal fistula. *J Thorac Cardiovasc Surg.* 2007;134(2):411–414. <https://doi.org/10.1016/j.jtcvs.2007.04.030>.