

DOI: 10.7759/cureus.59145

Received 04/02/2024 Review began 04/18/2024 Review ended 04/24/2024 Published 04/27/2024

© Copyright 2024

Sindhu et al. This is an open access article distributed under the terms of the Creative Commons Attribution License CC-BY 4.0., which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Unmasking the Invisible: A Case Study of Aspiration Pneumonia Unveiling a Bronchoesophageal Fistula

Arman Sindhu ¹, Ulhas Jadhav ¹, Babaji Ghewade ¹, Jay Bhanushali ¹, Pallavi Yadav ²

1. Respiratory Medicine, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research, Wardha, IND 2. Obstetrics and Gynecology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research. Wardha, IND

Corresponding author: Arman Sindhu, armansindhu1198@gmail.com

Abstract

Bronchoesophageal fistula (BEF) is a rare, yet clinically significant, condition characterized by an abnormal connection between the bronchial tree and the esophagus. We present the case of a 25-year-old female who initially presented with symptoms of aspiration pneumonitis and was subsequently diagnosed with BEF, attributed to poorly differentiated squamous cell carcinoma. Despite initial attempts at palliative intervention through esophageal stent placement, persistent symptoms prompted further investigation, revealing the underlying malignancy. This case underscores the diagnostic challenges associated with BEF, particularly when malignancy is involved, and emphasizes the importance of a multidisciplinary approach in optimizing patient outcomes. Early recognition, thorough evaluation, and comprehensive oncological management are essential in addressing the clinical complexities posed by BEF. Further research is warranted to better understand the pathophysiology and optimal management strategies for this rare but clinically significant condition.

Categories: Pulmonology

Keywords: bronchoesophageal fistula, diagnostic challenges, multidisciplinary approach, endoscopic intervention, squamous cell carcinoma, aspiration pneumonitis

Introduction

Bronchoesophageal fistula (BEF) is a rare pathological condition characterized by an abnormal connection between the bronchial tree and the esophagus. It can result from various etiologies, including congenital malformations, trauma, inflammatory diseases, and malignancies [1]. Although BEF is uncommon, its clinical significance lies in its potential to cause severe respiratory complications such as recurrent pneumonia, aspiration pneumonitis, and respiratory distress, which can lead to significant morbidity and mortality if left untreated [2]. The clinical presentation of BEF often includes symptoms such as coughing, dyspnea, recurrent pneumonia, hemoptysis, and gastrointestinal complaints like dysphagia or regurgitation [3]. Diagnosis typically involves a combination of imaging modalities such as chest X-ray, CT scans, and endoscopic procedures like bronchoscopy and esophagoscopy. Confirmation of the diagnosis is crucial for appropriate management, including endoscopic interventions, surgical repair, or treatment of underlying conditions such as malignancy [4].

Endoscopic interventions such as stent placement have emerged as a viable option for the palliative management of BEF, offering symptom relief and improving quality of life in select cases [5]. However, definitive treatment often depends on addressing the underlying cause, especially when malignancy is involved. Multidisciplinary management involving gastroenterologists, pulmonologists, thoracic surgeons, and oncologists is essential for optimizing outcomes in patients with BEF [6]. Here, we present a case of aspiration pneumonitis secondary to BEF in a young female ultimately diagnosed with poorly differentiated squamous cell carcinoma. This case highlights the importance of considering BEF in the differential diagnosis of patients presenting with recurrent respiratory symptoms. It underscores the need for prompt diagnosis and multidisciplinary management for optimal outcomes.

Case Presentation

A 25-year-old female presented to the emergency department with a 15-day history of cough with expectoration, fever, and breathlessness on exertion. She also reported weight loss and decreased appetite over the same duration. Before this presentation, she had been admitted to a rural hospital with similar complaints but was later referred to our center for further management.

The patient reported coughing episodes after ingesting water or semi-solid meals. There was no significant past medical history or history of trauma. On initial examination, she was lethargic with tachycardia and reduced breath sounds in the right mammary area on lung auscultation. Abdominal examination was unremarkable. A radiological examination of the chest X-ray on admission showed homogenous opacity in



the right lower lobe (Figure 1).

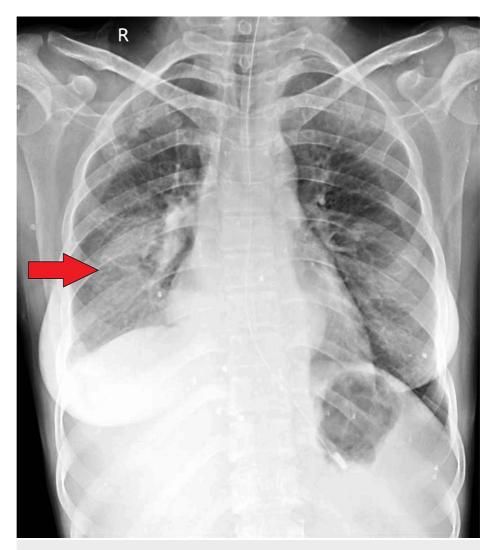


FIGURE 1: A chest X-ray done on admission showing homogenous opacity in the right lower lobe

Her initial vital signs were notable for a pulse of 138 beats per minute, a blood pressure of 110/70 mm Hg, a respiratory rate of 24 breaths per minute, an oxygen saturation of 96% on room air, and an oral temperature of 38°C. High-resolution CT of the thorax revealed a heterogeneous opacity in the right lower lobe with communication between the lower one-third of the esophagus and the lower lobe of the lung, consistent with a BEF (Figure 2A-2B).







FIGURE 2: (A) An axial section of the CT of the thorax showing a communication between the esophagus and the right bronchus. (B) An axial section of the CT of the thorax showing a fluid-filled cavity in the right posterior segment of the lower lobe

Flexible optic bronchoscopy confirmed the fistulous connection between the segmental bronchus and the esophagus. Methylene blue injected into the right lower lobe showed a decreased return on suction, and subsequent aspiration from the nasogastric tube revealed food particles mixed with methylene blue, further confirming the diagnosis. The patient underwent an endoscopy, which revealed a fistulous opening in the esophagus, 30 cm from the incisor teeth. An esophageal-covered metal stent with hemoclip placement was performed to manage the fistula. However, the patient did not experience relief from symptoms post-stent placement (Figure 3). A contrast study demonstrated contrast-filled tracts between the esophagus and the right bronchus, confirming the persistence of the fistula (Figure 4A-4C).



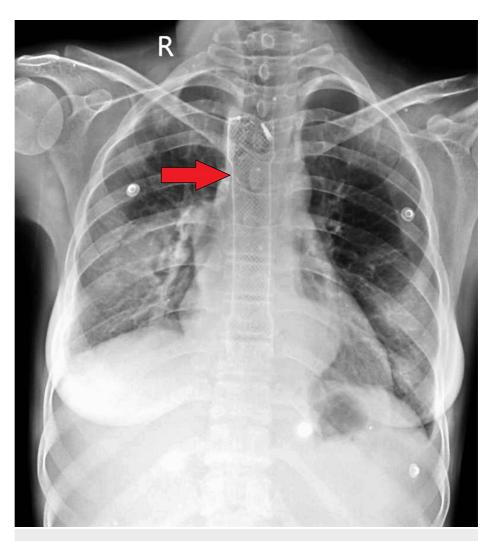


FIGURE 3: A chest X-ray posteroanterior view, showing the placement of the esophageal stent

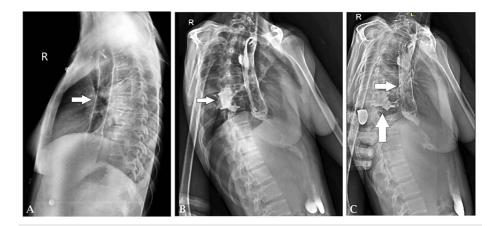


FIGURE 4: (A) A lateral chest X-ray showing the placement of the esophageal stent. (B) Contrast study showing leak of contrast from the esophagus to the right bronchus with a tract filled with contrast. (C) Contrast study showing leak of contrast in the cavity in the right lower lobe



Histopathological examination of biopsies taken during endoscopy revealed poorly differentiated squamous cell carcinoma underlying the fistula. Nasogastric tube aspirate shows methylene blue injected during bronchoscopy in the right lower lobe suggesting a communication between the bronchus and the esophagus (Figure *5*). The patient was referred for further oncological management.



FIGURE 5: Nasogastric tube aspirate shows methylene blue injected during bronchoscopy in the right lower lobe suggesting a communication between the bronchus and the esophagus

Discussion

BEF is a rare yet clinically significant condition requiring prompt recognition and appropriate management. In this case, the patient presented with symptoms consistent with aspiration pneumonitis, which led to the discovery of a BEF ultimately attributed to poorly differentiated squamous cell carcinoma. BEF can arise from various etiologies, including malignancy, inflammation, trauma, or iatrogenic causes [7]. Malignancy is a well-recognized cause of BEF, with squamous cell carcinoma being the most common histological type associated with this condition [8]. Our case adds to the existing literature by highlighting the diagnostic challenges posed by BEF and emphasizing the importance of considering underlying malignancy, particularly in cases where symptoms persist despite initial interventions.

Endoscopic evaluation, including flexible bronchoscopy and esophagoscopy, is crucial in diagnosing BEF [9]. In our case, bronchoscopy revealed a fistulous connection between the bronchus and esophagus, further



confirmed by contrast studies and histopathological examination. These findings align with previous studies demonstrating the utility of endoscopic modalities in diagnosing BEF and guiding subsequent management [10]. Management of BEF often involves a multidisciplinary approach, with interventions aimed at both symptom palliation and addressing the underlying cause [11]. In our case, placement of an esophageal stent was initially attempted to alleviate symptoms; however, the persistence of symptoms prompted further investigation, leading to the diagnosis of squamous cell carcinoma. This underscores the importance of thorough evaluation and consideration of underlying malignancy in patients with suspected BEF, as palliative measures alone may not provide definitive relief.

The prognosis of BEF depends largely on the underlying etiology and the stage of the disease at presentation. While benign causes of BEF may respond well to conservative management or endoscopic interventions, malignant BEF often carries a poorer prognosis and requires comprehensive oncological treatment [12]. In our case, the presence of squamous cell carcinoma necessitated further oncological evaluation and treatment planning.

Conclusions

This case underscores the diagnostic challenges and complexities associated with BEF, particularly when malignancy is involved. Despite initial attempts at palliative intervention through esophageal stent placement, the persistence of symptoms prompted further investigation, ultimately leading to the diagnosis of poorly differentiated squamous cell carcinoma. This highlights the importance of considering underlying malignancy in patients with suspected BEF and emphasizes the need for a multidisciplinary approach to optimize patient outcomes. Early recognition, thorough evaluation, and comprehensive oncological management remain paramount in addressing the clinical complexities posed by BEF. Further research and clinical studies are warranted to better understand the pathophysiology and optimal management strategies for this rare but clinically significant condition.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Arman Sindhu, Ulhas Jadhav, Babaji Ghewade, Jay Bhanushali

Acquisition, analysis, or interpretation of data: Arman Sindhu, Ulhas Jadhav, Babaji Ghewade, Pallavi Yadav, Jay Bhanushali

Drafting of the manuscript: Arman Sindhu, Pallavi Yadav, Jay Bhanushali

Critical review of the manuscript for important intellectual content: Arman Sindhu, Ulhas Jadhav, Babaji Ghewade, Pallavi Yadav, Jay Bhanushali

Supervision: Ulhas Jadhav

Disclosures

Human subjects: Consent was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

Acknowledgements

I would like to express my deep appreciation for the integral role of artificial intelligence (AI) like Grammarly and ChatGPT in completing this research paper. Incorporating AI technologies has significantly elevated the efficiency and depth of our analysis, adding a layer of sophistication to our study. By utilizing advanced algorithms and machine learning techniques, we were able to process extensive datasets with unprecedented speed, allowing for a more comprehensive exploration of intricate patterns and trends.

References

- Zhang BS, Zhou NK, Yu CH: Congenital bronchoesophageal fistula in adults. World J Gastroenterol. 2011, 17:1358-61. 10.3748/wjg.v17.i10.1358
- Aggarwal D, Mohapatra PR, Malhotra B: Acquired bronchoesophageal fistula. Lung India. 2009, 26:24-5. 10.4103/0970-2113.45201



- Kim HS, Khemasuwan D, Diaz-Mendoza J, Mehta AC: Management of tracheo-oesophageal fistula in adults. Eur Respir Rev. 2020, 29:200094. 10.1183/16000617.0094-2020
- Colt HG, Janssen JP, Dumon JF, Noirclerc MJ: Endoscopic management of bronchial stenosis after double lung transplantation. Chest. 1992, 102:10-6. 10.1378/chest.102.1.10
- Shin JH, Song HY, Ko GY, Lim JO, Yoon HK, Sung KB: Esophagorespiratory fistula: long-term results of palliative treatment with covered expandable metallic stents in 61 patients. Radiology. 2004, 232:252-9. 10.1148/radiol.2321030733
- Smith C, Bannon M, Ashraf A, Kaushik P, Marak C: Bronchoesophageal fistula: an unusual manifestation of lung cancer. Respir Med Case Rep. 2022, 37:101634. 10.1016/j.rmcr.2022.101634
- Sersar SI, Maghrabi LA: Respiratory-digestive tract fistula: two-center retrospective observational study.
 Asian Cardiovasc Thorac Ann. 2018, 26:218-23. 10.1177/0218492318755013
- Gschossmann JM, Bonner JA, Foote RL, Shaw EG, Martenson JA Jr, Su J: Malignant tracheoesophageal fistula in patients with esophageal cancer. Cancer. 1993, 72:1513-21. 10.1002/1097-0142(19930901)72:5<1513::aid-cncr2820720504>3.0.co;2-g
- 9. Appelqvist P, Mattila S, Jyrälä A, Tala P: Surgical treatment of carcinoma of the oesophagus and cardia . Scand J Thorac Cardiovasc Surg. 1977, 11:278-82.
- Iwagami H, Ishihara R, Yamamoto S, et al.: Esophageal metal stent for malignant obstruction after prior radiotherapy. Sci Rep. 2021, 11:2134. 10.1038/s41598-021-81763-x
- Reed MF, Mathisen DJ: Tracheoesophageal fistula. Chest Surg Clin N Am. 2003, 13:271-89. 10.1016/s1052-3359(03)00030-9
- 12. Daly M, Zarate-Lopez N: Functional gastrointestinal disorders: history taking skills in practice . Clin Med (Lond). 2021, 21:e480-6. 10.7861/clinmed.2021-0189