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A Rare Tracheopleural Fistula Secondary to Chemoradiotherapy for Non-Small Cell Lung Cancer

Mansoor Alam[1], Pravachan Hegde[2], William Silveira[3], Amna Ali[4], Megan Trang[5], Emaan Baqai[6], Kyle McCullough[7], Habiba Hashimi[8], John Lin[9].

1. Oncology, Premier Cancer Care, Fresno, CA
2. Advanced Interventional Thoracic Endoscopy, UCSF Fresno, CA
3. Radiation oncology, Community cancer institute, Clovis, ca
4. General Surgery, UCSF Fresno, CA
5. University of California at Los Angeles, CA
6. California Health Sciences University, Clovis, CA
7. Baylor Scott and White Research Institute, TX
8. Plastic Surgery, Virginia Commonwealth University, VA
9. Thoracic Surgery, Central California Chest Surgery, CA

*Correspondence: John Lin Linj2011@gmail.com

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ABSTRACT

The literature regarding tracheopleural fistulae is limited: very little is known regarding the development of these fistulae secondary to non-small cell lung cancer (NSCLC), for it is an exceedingly rare occurrence. However, given the high associated morbidity and mortality, analysis is needed. We report the case of a tracheopleural fistula in a 42-year-old man with clinical stage IV non-small cell lung cancer treated with chemoradiation. He presented with worsening shortness of breath and new-onset hemoptysis, and he was found to have a 2 cm tracheopleural fistula. Despite multiple interventions including rigid bronchoscopy and Y stent placement, he continued to deteriorate, and one month after diagnosis of the tracheopleural fistula, he died as a result of respiratory failure and septic shock.

Introduction

Lung cancer is the number one cause of oncological mortality in both men and women in the United States [1]. Only about 22% of lung cancer patients survive 5 or more years after their diagnosis, including non-small cell lung cancer (NSCLC) and small cell lung cancer [2]. More than half of patients die within one year of diagnosis. NSCLC

represents the majority of lung cancer cases, comprising 80-85%, with cigarette smoking as the leading risk factor [3]. Pancoast tumors, accounting for 3-5% of all lung cancers, primarily manifest as NSCLC and originate from the apical pleuropulmonary groove superior to the first rib [4].

While lung cancer is a frequently discussed topic in literature, the development of tracheopleural fistulae in cancer patients is a rare occurrence. We present a patient who developed a tracheopleural fistula following chemotherapy and radiation for his clinically stage IV NSCLC Pancoast tumor.

Case Report

The patient is a 42-year-old man with a history of tobacco use and stage IV poorly differentiated, non-small cell lung cancer metastatic to the mediastinum. CT chest showed a 7.8 cm x 6.2 cm right apical lung mass consistent with Pancoast tumor. An 8.9 cm x 4.5 cm right paratracheal mass, radiologically consistent with metastatic lesion, compressing the superior vena cava was also noted (Figure 1). He was treated with chemotherapy (1 cycle carboplatin/etoposide, 3 cycles carboplatin/taxol/tecenteriq/avastin) and radiation (total dose 6000cGy in 30 fractions). While the patient had good response to chemotherapy and radiation, post-treatment imaging revealed the right apical lung mass with right metastatic paratracheal mass had expanded into a 10 cm x 11 cm x 10 cm complex cavitary lesion involving the entirety of the right upper lobe (Figure 2). The presence of gas within the mass and proximity to the trachea were indicative of tracheal necrosis. A repeat computerized tomography one week later revealed a large right hydropneumothorax with mediastinal shift to the left and a 2 cm tracheopleural fistula associated with recurrent right upper lobe infections and soilage of the right middle and right lower lobes.

Figure 1: Pretreatment paratracheal metastasis

Figure 2: Post treatment of tracheopleural fistula

He then presented to the local emergency department with worsening shortness of breath and new-onset hemoptysis. On presentation, the patient was tachycardic to the 130s, tachypneic to the 30s, normotensive and afebrile. A chest x-ray demonstrated a large right-sided hydropneumothorax and a large right lung mass. During his admission, he underwent a rigid bronchoscopy with Y stent and right chest tube placement (Figure 3). The Y stent

was placed to both exclude the fistula and to improve ventilation. The chest tube was for source control of the patient's infected hydropneumothorax. He also required a pericardial window for tamponade. The patient tolerated the procedure well and was extubated the following day. His bronchoscopy had raised concern for invasive fungal infection (Figure 4), and he was placed on amphotericin, in addition to cefepime and metronidazole for his necrotizing pneumonia.

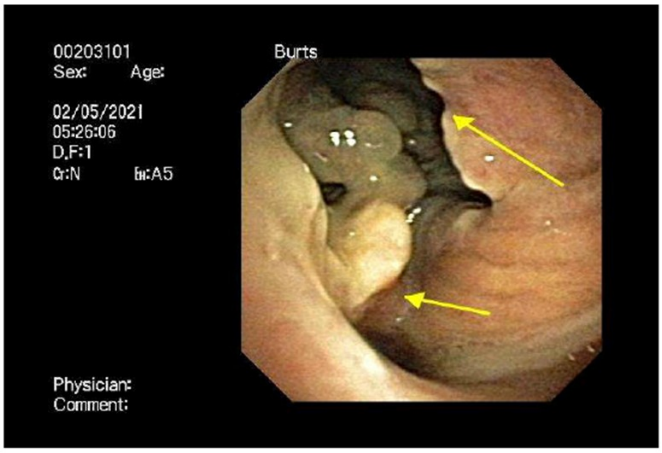

Figure 3: lower end of trachea with fistula (upper arrow) and carina (lower arrow)

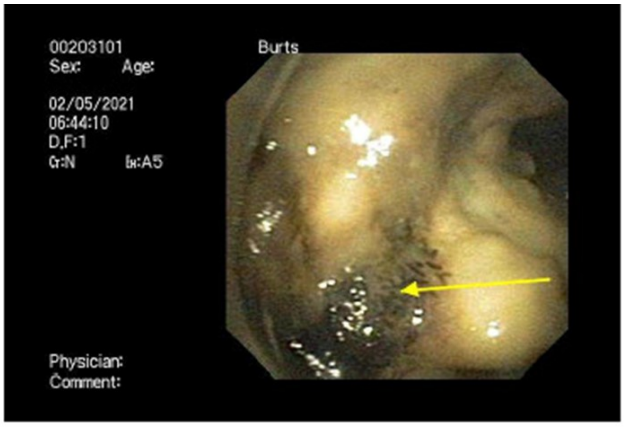

Figure 4: black eschar-like material of the right upper lobe; invasive fungus

One week after his admission, the patient was re-intubated for respiratory distress. He eventually required maximal ventilator support as well as multiple vasopressors indicative of septic shock. Imaging revealed worsening hydropneumothorax. Repeat bronchoscopy showed the Y stent was in an appropriate position in the lower trachea, but mucus plugging and moderate mucopurulent secretions were observed in the right bronchial tree.

Due to his declining status and metastatic cancer, the patient's family chose to transition the patient to comfort care. The patient died from acute respiratory failure and septic shock one month after the tracheopleural fistula diagnosis. He was a poor candidate for surgery due to the large tracheal defect, advanced cancer staging, and septic shock. Had he survived his infection, he would have potentially been a candidate for a right pneumonectomy with tracheal reconstruction.

Discussion

Tracheopleural fistulae are rare occurrences [5] and rare complications of lung cancer treatment. Reported literature is limited, mainly consisting of case reports and studies. Malignant tumors associated with tracheal or bronchial-mediastinal fistulae include adenocarcinoma [6], lymphoma [7], and squamous cell carcinoma [8] of the lung. Other case reports have described instances of such fistulae following endobronchial and tracheal radiation [9], and potentially stemming from airway perforation and necrosis after the chemotherapy and radiation [10].

In the reviewed literature, the following two cases have demonstrated mortality from a tracheopleural fistula. These instances highlight the significant tumor-related morbidity and mortality of patients with advanced cancers. The treatment strategy involves endoscopic stent placement. Although endoscopic treatments of fistulae have shown success, the procedure's role is palliative. Surgery is typically not an option, given infection in the field of

tracheal resection/plasty and the lack of material substitute for trachea.

Machuzak et al. reported a patient who developed a tracheopleural fistula after undergoing chemoradiation for his unresectable NSCLC [11]. The patient, who initially presented with recurrent episodes of "bronchitis," developed a severe, productive cough with marked dyspnea six weeks after starting bevacizumab. Chest computed tomography was positive for a tracheopleural fistula. Bronchoscopic examination revealed malignant-related airway wall necrosis and a defect along the entire right lateral wall of the trachea. To recreate normal anatomy, a self-expanding metallic stent (SEMS) was placed to support the trachea, and a Dumon Y-stent was deployed inside the SEMS to exclude the eroded right tracheal wall. With significant symptom improvement and no supplemental oxygen, the patient was discharged 48 hours after the procedure. The patient was stable for 18 months after stent placement, but after restarting palliative radiation, eventually died from massive hemoptysis.

Ferrari et al. reported a case of a patient with a history of heavy tobacco use who developed sudden onset of dyspnea after undergoing concomitant chemoradiotherapy (CRT) for pharyngeal squamous cell carcinoma [12]. CT confirmed a tracheopleural fistula permitting enteral reflux to the upper airway and pleura. The fistula was treated with endoscopic insertion of a silicone stent. Three months later, CT showed resolution of the fistula and no evidence of malignant disease. However, two months later, the patient unexpectedly died. Autopsy revealed sparse tumor cells in the oropharyngeal and tracheal mucosa, minimal residual tracheopleural fistula, and extensive myocardial and epicardial metastases from the oropharyngeal squamous cell carcinoma.

Tracheopleural fistula have been sparsely documented in the literature; very few instances of these fistulae as secondary to non-small cell lung cancers have been reported, despite their high association with morbidity and mortality.

Endoscopic treatments, while temporarily successful, serve a mainly palliative role, highlighting significant risk of mortality in patients with advanced cancers and tracheopleural fistulae.

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