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CASE REPORT

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Tracheal Tumor Fistula: an Unusual Complication of Endobronchial Ultrasound-Guided Transbronchial Needle Aspiration

Fístula traqueo-tumoral: una complicación excepcional de la punción-aspiración con aguja transbronquial guiada por ecografía endobronquial

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ABSTRACT

The endobronchial ultrasound-guided transbronchial needle aspiration (EBUS-TBNA) is a safe, minimally invasive technique used for the diagnosis of mediastinal and hilar adenopathy, especially lung cancer. Even though complications are rare (around 1%), they may include severe bleeding, pneumomediastinum and tracheomediastinal fistulas. We present the case of a patient with lung adenocarcinoma diagnosed through EBUS-TBNA who developed a fistula between the trachea and the tumor after the procedure. No previously described cases were found in the consulted scientific literature, as the patient did not have the main risk factors for the development of this type of complication. The patient did not develop any subsequent infectious symptoms, possibly thanks to the early use of antibiotic therapy.

Key words: Fístula; Tumor; Transbronchial needle aspiration; Endobronchial ultrasound

RESUMEN

La punción con aguja transbronquial guiada por ultrasonido endobronquial (EBUS-TBNA) es una técnica segura y mínimamente invasiva utilizada para el diagnóstico de adenopatías mediastínicas e hiliares, especialmente en el cáncer de pulmón. Aunque las complicaciones son raras (alrededor del 1%), pueden incluir sangrado grave, neumomediastino y fístulas traqueomediastínicas. Presentamos un caso clínico de un paciente con adenocarcinoma de pulmón diagnosticado mediante EBUS-TBNA, el cual desarrolló una fístula entre tráquea y tumor tras la realización de la técnica, no encontrando casos descritos previamente en la literatura científica consultada al no presentar los principales factores de riesgo para el desarrollo de este tipo de complicaciones. El paciente no desarrolló clínica infecciosa posterior, posiblemente gracias al uso de antibioterapia de forma precoz.

Palabras clave: Fístula; Tumor; Punción-aspiración con aguja transbronquial; Ecografía endobronquial

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INTRODUCTION

The endobronchial ultrasound-guided transbronchial needle aspiration (EBUS-TBNA) is a minimally invasive diagnostic bronchoscopic technique performed with the assistance of an ultrasound convex mini-probe at the tip of the bronchoscope. It is used for the study of hilar and mediastinal adenopathy, being particularly useful for staging lung cancer. It is a safe technique because ultrasound allows for the recognition of pulmonary, pleural, and vascular structures, resulting in a low complication rate, typically around 1%.[1] Complications are generally of minor severity and often resolve with conservative treatment.[2] However, within the spectrum of complications, there is a small percentage, estimated at 0.26%[3], that includes significant bleeding, pneumomediastinum, tracheomediastinal fistulas, and their infectious complications. These complications can lead to prolonged hospitalization and, in the case of lung cancer, a delay in the start of oncological treatment.[4]

Clinical observation

Recently, we attended a 54-year-old male patient with a significant history of tobacco use (cumulative smoking index of 72 packs-years), who initially consulted the Otorhinolaryngology Service due to dysphonia. During the fiberoptic examination, right vocal cord paralysis was identified. For further investigation, a chest computed tomography (CT) was requested, revealing right paratracheal and mediastinal adenopathies. The patient was then referred to a pulmonology consultation to be assessed.

During the initial consultation, the patient reported a 6-month history of dysphonia associated with constitutional symptoms and unintentional weight loss of 20 kg over the past 6 months, without any additional symptoms. Physical examination was unremarkable. The chest CT images were reviewed, showing conglomerate lymph node masses in the right paratracheal region (4.8×4.2×5.5 cm in diameter) and subcarinal region (1.5×2.7×5 cm in diameter) with certain suggestive signs of necrosis. Additionally, a right hilar adenopathy (3.2×2.7 cm) was observed (Image 1).

A bronchoscopy was performed, revealing mucosal thickening in the middle and distal third of the trachea without clear signs of infiltration. Furthermore, an EBUS-TBNA procedure was conducted in the right paratracheal station without immediate complications. The histologic diagnosis was consistent with metastasis from non-small cell lung cancer, although the sample was insufficient to complete the immunohistochemical study.

Therefore, a new EBUS-TBNA of the same station was performed three weeks later, without immediate complications. This second procedure confirmed the final diagnosis of metastatic lung adenocarcinoma, allowing for further relevant studies (EGFR [epidermal growth factor receptor], ALK [anaplastic lymphoma kinase gene], ROS1 [c-ros oncogene 1], and PDL1 [programmed death ligand-1]).

Two weeks later, upon completion of staging with PET-CT (positron emission tomography - computed tomography), an incidental finding was observed, indicating a contained rupture of the right tracheal wall with air leak towards the right paratracheal tumor (Figure 1).

The following week, a new diagnostic fiberoptic bronchoscopy was performed, revealing grade III mucosal infiltration of the middle and distal third of the trachea, along with a fistula in the middle third of the trachea, of approximately 5 mm, surrounded by tumor tissue (Figure 2). The

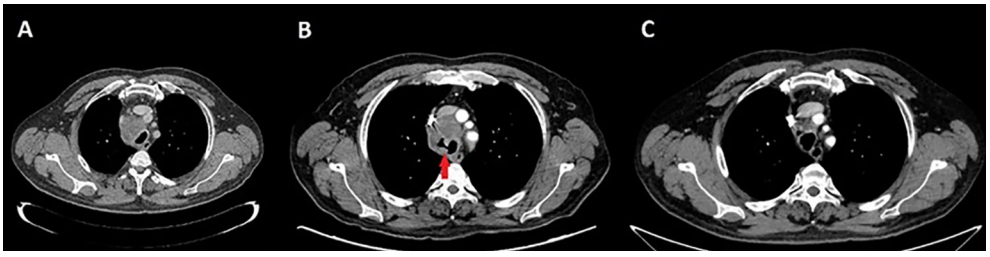

Figure 1. A) Initial chest CT showing conglomerate in the right paratracheal region. B) Chest PET-CT – tracheal tumor fistulization in the right paratracheal conglomerate (date). C) Chest CT post-chemotherapy treatment – significant reduction of right paratracheal adenopathic conglomerate with persistence of fistula.

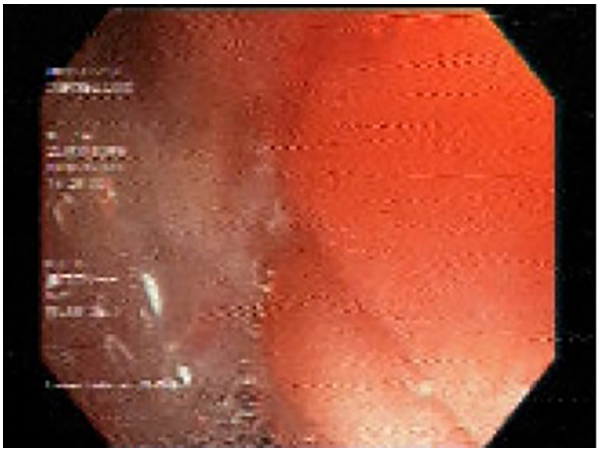

Figure 2. Endoscopic image of tracheal tumor fistula

patient received prophylactic treatment with oral amoxicillin-clavulanic acid for one week, and no other complications developed during follow-up.

Given the diagnosis of stage T×N3M1 lung adenocarcinoma, the patient began oncological treatment with chemotherapy. However, radiation therapy was not included due to the high risk of mediastinitis associated with the tracheal fistula.

DISCUSSION

EBUS-TBNA is a safe bronchoscopic technique for the study of hilar-mediastinal adenopathy and staging of lung cancer.[1-4]

Despite the fact that serious complications from this procedure account for a very low percentage (0.26%) and represent exceptional cases,[3] given the great usefulness and increasing number of procedures performed with EBUS-TBNA, we must take it into account to minimize their occurrence as much as possible.[4]

After reviewing the available literature on serious complications following EBUS-TBNA related to the occurrence of fistulas, we have found a case of bronchomediastinal fistula with development of pneumomediastinum following the EBUS-TBNA procedure after a mediastinoscopy[5]; a case of hemoptysis due to the development of an aortopulmonary fistula following EBUS-TBNA in a patient who was previously receiving antiangiogenic treatment with bevacizumab[6]; and even the development of a tracheomediastinal fistula, without clinical consequences, following the initiation of radiotherapy in a patient who had recently undergone EBUS-TBNA[3]. However, we have not found any description of tumor fistulization into the trachea following the EBUS-TBNA procedure, as in the case we present. Furthermore, this is a patient without apparent risk factors, as he had not previously undergone any mediastinoscopy or received antiangiogenic treatment (bevacizumab), or radiotherapy, which also differentiates him from the rest of the cases of the consulted literature. Although the possible presence of necrosis in the conglomerate lymph node masses could have been a risk factor in our patient's case.

Currently, there is no clear evidence of the efficacy of the use of prophylactic antibiotics to prevent infectious complications following EBUS-TBNA.[7] However, our case would support the thesis of Jang et al, who already described that prophylactic antibiotic therapy should be considered in cases of cystic or necrotic lesions, with the intention of covering the most common microorganisms in the oral cavity and preventing the development of infectious complications.[4]

In conclusion, we present a case that shows the development of a fistula between the trachea and tumor following the EBUS-TBNA procedure, not previously described in the consulted scientific literature, as it does not have the main risk factors for the development of fistulous complications nor subsequent infectious symptoms, possibly due to the early use of antibiotic therapy.

Conflict of interest

The authors have no conflict of interest to declare that are relevant to the presented case.

REFERENCES

1. Asano F, Aoe M, Ohsaki Y, et al. Complications associated with endobronchial ultrasound-guided transbronchial needle aspiration: a nationwide survey by the Japan Society for Respiratory Endoscopy. Respir Res. 2013;14:50. https://doi.org/10.1186/1465-9921-14-50
2. Eapen GA, Shah AM, Lei X, Jimenez CA, et al. American College of Chest Physicians Quality Improvement Registry, Education, and Evaluation (AQuIRE) Participants. Complications, consequences, and practice patterns of endobron-

chial ultrasound-guided transbronchial needle aspiration: Results of the AQuIRE registry. Chest. 2013;143:1044-53. https://doi.org/10.1378/chest.12-0350
3. Holty JE, Kuschner WG, Gould MK. Accuracy of transbronchial needle aspiration for mediastinal staging of non-small cell lung cancer: a meta-analysis. Thorax. 2005;60:949-55. https://doi.org/10.1136/thx.2005.041525
4. Jang JG, Ahn JH, Lee SS. Delayed onset of mediastinitis with tracheomediastinal fistula following endobronchial ultrasound-guided transbronchial needle aspiration; A case report. Thorac Cancer. 2021;12:1134-6. https://doi.org/10.1111/1759-7714.13888
5. Bougioukas I, Seipelt R, Huwer H. Bronchial Fistula and Pneumomediastinum after EBUS-TBNA Following Mediastinoscopy. Thorac Cardiovasc Surg Rep. 2019;8:e11-e13. https://doi.org/10.1055/s-0039-1688476
6. Wong J, Gutierrez C, Shannon VR, Eapen GA, Faiz SA. Bronchomediastinal Fistula after Endobronchial Ultrasound-guided Transbronchial Needle Aspiration. Am J Respir Crit Care Med. 2016;194:114-5. https://doi.org/10.1164/rccm.201601-0144IM
7. Takagi H, Nagaoka T, Ando K, et al. Efficacy of antibiotic prophylaxis after endobronchial ultrasound-guided transbronchial needle aspiration: a preliminary prospective study. J Pulm Respir Med. 2017;7:416. https://doi.org/10.4172/2161-105X.1000416