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Case Report

Tracheal adenoid cystic carcinoma: A case report

A rare case of ACC

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Abstract

Introduction: Adenoid cystic carcinoma (ACC) of the trachea is a rare malignancy that often leads to airway obstruction and misdiagnosis as chronic respiratory conditions such as asthma. Due to its slow-growing nature and submucosal infiltration, ACC presents diagnostic and therapeutic challenges.

Case Presentation: Here, we report the case of a 46-year-old female who was misdiagnosed with bronchial asthma for seven years before being accurately diagnosed with tracheal ACC. The patient underwent a rigid bronchoscopy for airway stabilization, followed by transcarinal tracheal resection and lymph node dissection. Postoperatively, the patient exhibited significant improvement in pulmonary function and was referred for adjuvant radiotherapy.

Conclusion: This case highlights the importance of considering alternative diagnoses in patients with persistent respiratory symptoms and underscores the necessity of a multidisciplinary approach for optimal management of tracheal ACC.

Keywords

adenoid cystic carcinoma, tracheal tumor, airway obstruction, bronchoscopy, surgical resection

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Introduction

Tracheal tumors constitute approximately 1% of all respiratory system malignancies. The majority of these tumors are squamous cell carcinomas, while only about 10% are diagnosed as adenoid cystic carcinoma (ACC).[1,2] ACC can partially or completely obstruct the airway due to its involvement of the trachea and main bronchi, often leading to misdiagnosis as bronchial asthma and prolonged incorrect management. The optimal treatment for primary tracheal ACC involves surgical resection combined with radiotherapy.[1,2] In this case report, we present a rare case of tracheal ACC, its diagnostic challenges, and the treatment approach.

Case Presentation

A 46-year-old female patient was admitted with complaints of wheezing and dyspnea. Her medical history revealed that she had been followed for seven years with a misdiagnosis of bronchial asthma. Over the past six months, her dyspnea had progressively worsened, leading to the use of combination inhaler bronchodilator therapy. On physical examination, bilateral biphasic diffuse rhonchi were noted. Laboratory tests revealed leukocytosis and an elevated erythrocyte sedimentation rate. The posteroanterior chest radiograph was unremarkable (Figure 1). Pulmonary function tests (PFT) showed: FVC: 110% (3.27 L), FEV_1: 38% (0.96 L), and FEV_1/FVC: 79%, with a negative bronchodilator reversibility test. The flow-volume curve demonstrated flattening of both inspiratory and expiratory limbs (Figure 2). Fiberoptic bronchoscopy revealed a tumoral mass occluding three-quarters of the tracheal lumen at its distal end. The patient had an oxygen saturation of 78% while receiving 5 L/min of oxygen, was unable to lie supine, and exhibited stridor. To secure the airway, the patient was intubated in the operating room and underwent rigid bronchoscopy with argon plasma coagulation (40 W) for vaporization of the mass. A biopsy taken during the procedure confirmed the diagnosis of adenoid cystic carcinoma.

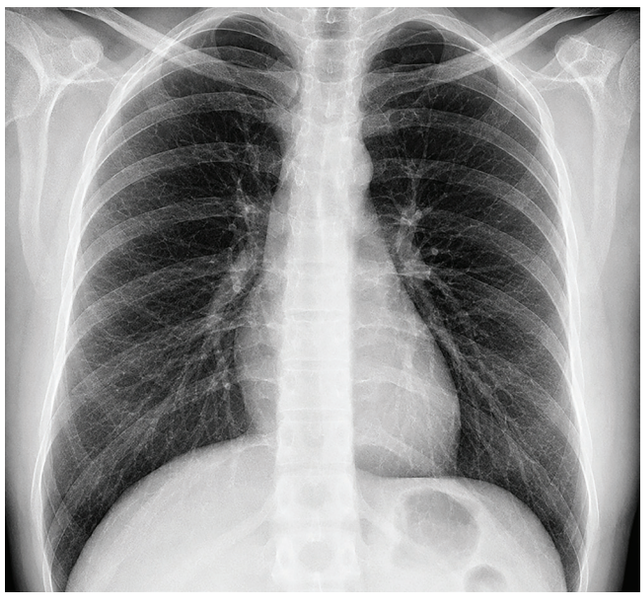

Figure 1. *Posteroanterior chest radiograph showed no apparent abnormalities*

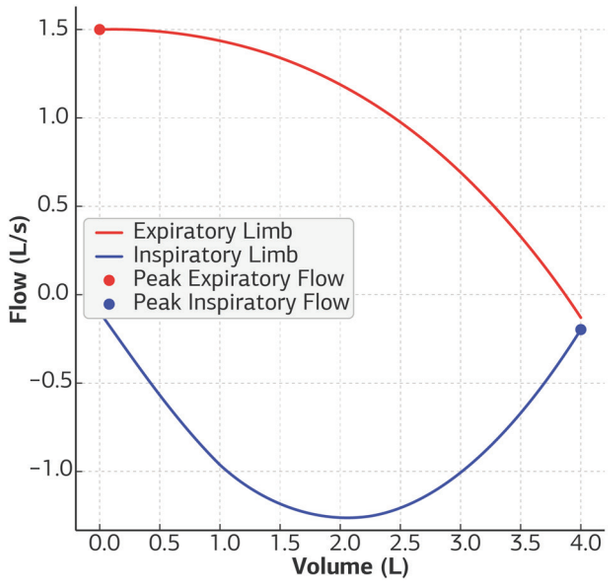

Figure 2. *Pulmonary function test flow-volume curve demonstrating inspiratory and expiratory flattening*

Chest computed tomography (CT) demonstrated a soft tissue density extending from the distal trachea to the level of the right main bronchus, narrowing the carina from the right anterolateral side. Given the known submucosal spread characteristic of tracheal ACC, endobronchial treatment alone is not curative. Positron emission tomography-computed tomography (PET-CT) revealed pathological metabolic activity within the lesion and mildly increased metabolic activity in bilateral cervical, left jugular, and hilar lymph nodes (Figure 3). The patient was deemed suitable for surgical intervention and underwent right thoracotomy with transcarinal tracheal resection (3.5 cm) and lymph node dissection. Histopathological examination confirmed adenoid cystic carcinoma with tumor-free surgical margins and reactive lymph nodes. Postoperative PFT results showed improvement: FVC: 80% (2.34 L), FEV_1: 73% (1.83 L), FEV1/FVC: 79%. The patient was referred to the radiation oncology department for adjuvant radiotherapy.

Due to its rarity, we present this case to highlight the diagnostic and therapeutic approach to tracheal adenoid cystic carcinoma.

Ethical Approval

This study was approved by the Ethics Committee of Chest Diseases and Thoracic Surgery Research and Education Hospital, Ankara, Türkiye (Date: 12.01.2022, Decision No: 25).

This case report was prepared in accordance with the CARE guideline.

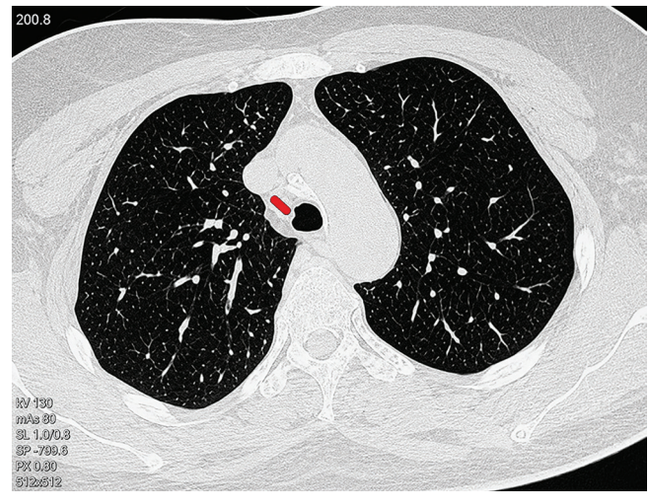

Figure 3. *PET-CT scan showed pathological metabolic activity in the lesion and mildly increased metabolic activity in cervical and hilar lymph nodes*

Discussion

Adenoid cystic carcinoma of the trachea is a rare malignancy with a slow but persistent growth pattern and a high tendency for perineural invasion. Due to its submucosal spread, it can be challenging to diagnose early, often leading to misclassification as asthma or chronic bronchitis.[3-5] In this case, the patient had been treated for asthma for several years before the correct diagnosis was made. This highlights the importance of considering alternative diagnoses in patients with refractory respiratory symptoms. Bronchoscopy and imaging techniques, particularly CT and PET-CT, play a crucial role in the diagnosis of tracheal ACC.[3,6-8]] The management of tracheal ACC involves a multidisciplinary approach, with surgical resection being the cornerstone of treatment. However, given the tumor's tendency for submucosal extension, achieving clear margins can be challenging, and adjuvant radiotherapy is often recommended to reduce recurrence risk.[3,7] In this case, the patient underwent transcarinal tracheal resection with negative margins, followed by adjuvant radiotherapy. The postoperative improvement in pulmonary function parameters indicated a successful intervention. Long-term follow-up is necessary, as tracheal ACC has a high potential for late recurrence.

Limitations

This case report is limited by its single-patient design, which restricts the generalizability of findings. Additionally, the long-term prognosis of the patient remains uncertain, necessitating extended follow-up. While imaging and histopathological findings support the diagnosis, molecular and genetic analyses were not performed, which could have provided further insights into the tumor's characteristics and potential targeted therapies.

Conclusion

Tracheal adenoid cystic carcinoma is a rare but significant airway malignancy that can be misdiagnosed as asthma for an extended period, delaying appropriate treatment. This case underscores the importance of thorough clinical evaluation, advanced imaging, and histopathological confirmation in patients with persistent respiratory symptoms. Surgical resection remains the preferred treatment, with adjuvant radiotherapy playing a key role in reducing recurrence risk. Early diagnosis and a multidisciplinary treatment approach are essential for improving patient outcomes.

Declarations

Animal and Human Rights Statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed Consent

Written informed consent was obtained from all participants using appropriate patient consent forms.

Data Availability

The datasets used and/or analyzed during the current study are not publicly available due to patient privacy reasons but are available from the corresponding author on reasonable request.

Conflict of Interest

The authors declare that there is no conflict of interest.

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None.

Abbreviations

ACC: Adenoid cystic carcinoma
CT: Computed tomography
FEV_1: Forced expiratory volume in 1 second
FVC: Forced vital capacity
PET-CT: Positron emission tomography-computed tomography
PFT: Pulmonary function test

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