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Case Report

Pulmonary mucoepidermoid carcinoma with endobronchial involvement: A case report

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ABSTRACT

Although mucoepidermoid carcinomas (MEC) commonly occur in the salivary glands, they are rarely encountered in the lungs. In this case report, we present a rare case of pulmonary mucoepidermoid carcinoma with endobronchial involvement in a 36-year-old Indian man who presented to our pulmonary outpatient clinic with a 10-year history of recurrent lower respiratory tract infections. A comprehensive workup was performed in his home country, but did not lead to a definitive diagnosis. At our hospital, high-resolution chest computed tomography was repeated, confirming a well-demarcated soft tissue mass measuring 1.7 x 1.8 x 1.1 cm in the right lower lobe bronchus. This was followed by positron emission tomography (PET) showing intense fluorodeoxyglucose (FDG) uptake in the mass with a standardized uptake value (SUV) of 7.9. The final diagnosis was made by histopathological examination of a resected right lower lobe, which confirmed the diagnosis of low-grade mucoepidermoid carcinoma, with no metastases found in the lymph nodes. The patient was then referred to a multidisciplinary oncology team, which opted for regular surveillance and follow-up.

1. Introduction

Mucoepidermoid carcinomas (MECs) are the most common malignant tumors of the salivary glands, typically involving the parotid and minor salivary glands, but may rarely occur in organs such as the pancreas, liver, esophagus, stomach, and lungs.[1] Pulmonary MECs account for 0.1–0.2% of all malignant lung neoplasms, with equal incidence in males and females, and a median age of presentation around 40 years.[2] These tumors arise from the glandular tissue of the tracheal and bronchial submucosa and consist of mucin-secreting cells, glandular cells, and squamous epithelial cells. Clinical manifestations are nonspecific, but often present with symptoms of bronchial obstruction and atelectasis.[3]

According to the WHO classification, MECs fall under the category of salivary gland-type carcinomas, along with adenoid cystic carcinoma and epimyoepithelial lung carcinomas.[4] Surgical resection is the main modality of management of pulmonary MECs, While low-grade MEC is associated with a favorable prognosis and a promising 5-year survival rate, the prognosis for high-grade MEC is poor, potentially comparable to that of other types of non-small cell lung cancer (NSCLC).[5] Endobronchial lesions as a presentation of pulmonary MECs are uncommon. In this report, we describe an uncommon instance of an MEC endobronchial lesion.

2. Case Presentation

A 36-year-old Indian male with no prior medical history presented to our pulmonary outpatient clinic with

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a 10-year history of recurrent lower respiratory tract infections. Initially, these episodes occurred annually but had progressively increased in frequency to every 3-4 months over the past two years. The patient's symptoms typically included a productive cough and intermittent febrile episodes, both of which responded to antibiotic therapy. The patient denied any associated weight loss, night sweats, hemoptysis, or history of recurrent skin, sinus, or gastrointestinal infections. He also reported no history of foreign body aspiration. The patient is a never-smoker, does not consume alcohol, and reported no exposure to pets or environmental triggers. There was no significant family history of respiratory diseases, malignancies, or autoimmune conditions

Three months before his presentation, the patient had undergone extensive investigations at another institution in his home country, including a chest CT scan, flexible bronchoscope (FOB) and biopsy. Unfortunately, the patient did not bring the imaging films or pathology slides, but a review of his medical reports indicated that the CT scan had revealed an endobronchial multilobulated irregular nodule in the right lower lobe, causing complete bronchial obstruction. Flexible bronchoscopy performed at that time showed a mucoid plug and a polypoid growth in the posterior segment of the right lower lobe bronchus. The lesion was debulked using snare electrocautery and a flexible cryoprobe. And the pathology report from the endobronchial biopsy indicated a neoplasm composed of clear to eosinophilic cells arranged in a cribriform pattern with mucinous material. These histological features raised the possibility of a salivary gland-type tumor or a neuroendocrine tumor, though immunohistochemistry results were inconclusive.

Upon presentation to our pulmonary clinic, the patient was in no apparent distress, with vital signs showing a blood pressure of 131/71 mmHg, a heart rate of 80 beats per minute, a temperature of 36.8°C, and an oxygen saturation of 99% on room air. Cardiopulmonary examination was unremarkable, with vesicular breath sounds bilaterally and no audible wheezes, rales, or rhonchi. Baseline blood investigations were within normal limits: hemoglobin 14.1 g/dL, white blood cell count 7100/L, and platelets 308,000/L. Renal function tests and electrolyte levels were unremarkable, including serum creatinine of 71 mol/L, urea 5 mmol/L, potassium 4.5 mmol/L, sodium 135 mmol/L, and bicarbonate 24 mEq/L. C-reactive protein (CRP) was mildly elevated at 17 mg/L. Liver enzymes were within normal range, with alanine transaminase (ALT) at 11 U/L and aspartate transaminase (AST) at 13 U/L.

High resolution Chest CT scan was repeated in our institution and confirmed a well-defined soft tissue mass measuring 1.7 x 1.8 x 1.1 cm within the right lower lobe bronchus (Figure 1). This followed by Positron emission tomography (PET) which demonstrated intense fluorodeoxyglucose (FDG) uptake within the mass, with a standardized uptake value (SUV) of 7.9. No additional FDG-avid pulmonary lesions or nodal involvement were identified (Figure 2). FOB is performed and revealed a fleshy endobronchial lesion in the lateral segment of the right lower lobe bronchial tree (Figure 3). An endobronchial biopsy was obtained, and the histopathology results showed minute fragments of poorly differentiated carcinoma. Immunohistochemistry was inconclusive, and the amount of biopsied tumor was minute, insufficient for further subtyping of the carcinoma or for conducting molecular studies. Based on the radiological and histopathology findings, the patient was referred to thoracic surgery. Where he subsequently underwent a video-assisted thoracoscopic surgery (VATS) right lower lobectomy. Histopathological analysis of the resected specimen confirmed the diagnosis of low-grade mucoepidermoid carcinoma, with no metastasis found in the lymph nodes (Figure 4). The patient was then referred to an oncology multidisciplinary team, which opted for regular surveillance and follow-up..

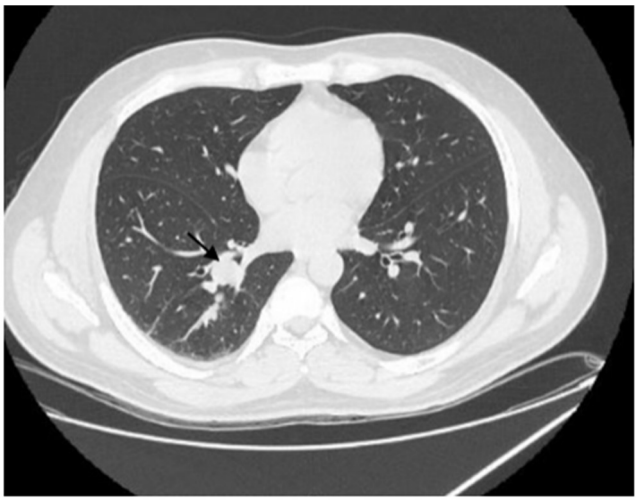

Figure 1: Axial section of chest ct scan showed a 1.7 x 1.8 x 1.1 cm soft tissue mass in the right lower lobe bronchus (Black arrow)

3. Discussion

Primary salivary gland-type tumors of the lung, including mucoepidermoid carcinoma (MEC), adenoid cystic carcinoma (ACC), and epithelial-myoepithelial carcinoma (EMEC), are rare neoplasms. Unlike more common lung cancers such as adenocarcinoma, squamous cell carcinoma, and small cell lung cancer, these tumors predominantly present in younger patients, and generally exhibit a more indolent clinical course.[6] Pulmonary mucoepidermoid carcinoma (PMEC) was first characterized by Smetana et al. in 1952 as a rare malignant neoplasm of the lungs, constituting approximately 0.1–0.2% of all adult lung malignancies.[7] There is limited literature on PMEC,

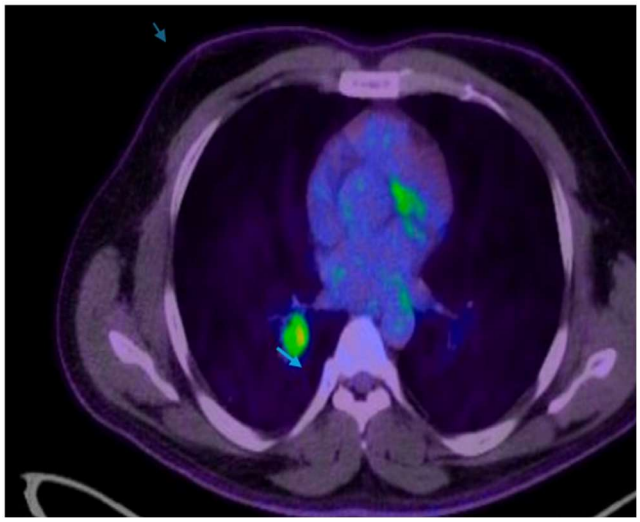

Figure 2: Intense FDG tracer uptake with SUV 7.9 seen in the right lower lobe nodule (Blue arrow).

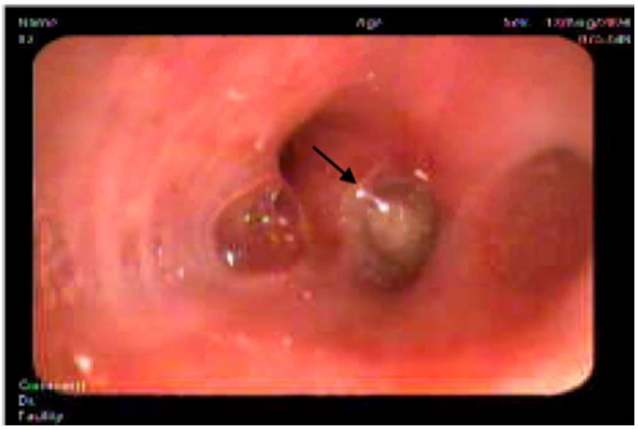

Figure 3: Right bronchial tree examination showing fleshy lesion far in the lateral segment of the right lower lobe (Black arrow).

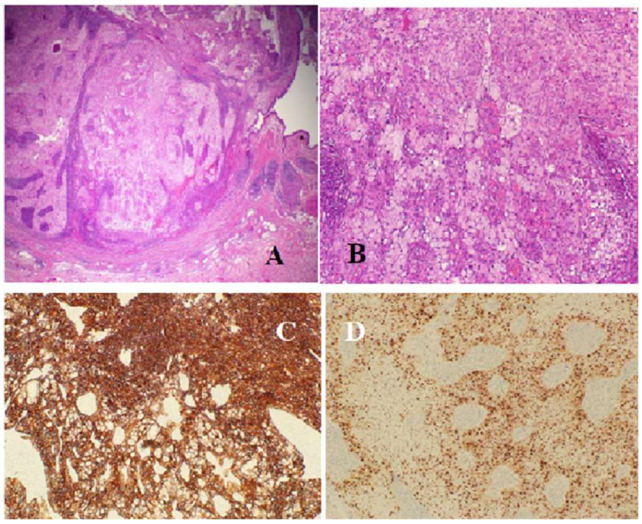

Figure 4: Microscopically, the tumor was located under bronchus epithelium where salivary gland normally present. It was composed of squamoid cells, intermediate cells and mucous secreting cells (Fig 4A-4B), growing in nested and solid pattern with focal cystic formation. There were no mitotic figures, necrosis of significant atypia. By immunohistochemistry, CK7 and CyclinD1 were positive (Fig 4C-4D). CK5/6, P40, P63 and Mucin special stain were focally positive, while TTF1 and Napsin were negative. All the above concluded an intermediate grade mucoepidermoid carcinoma.

with most reports being case studies. PMEC can arise across a wide age spectrum (3–78 years), with peak incidence observed in the third and fourth decades of life. The clinical manifestations of bronchial MEC are often nonspecific and may include cough, hemoptysis, bronchitis, wheezing, fever, chest pain, digital clubbing,[8] and signs of obstructive pneumonia.[9] Due to symptom overlap, patients may be erroneously diagnosed with bronchial asthma, as the symptoms of airway obstruction and irritation closely resemble those associated with bronchial asthma.[10] Although PMEC typically follows an indolent course, there have been reports of aggressive presentations with brain metastases.[11]

Radiologically, PMEC is often visualized as marked heterogeneous contrast enhancement on HRCT images. The presence of abundant microvessels, detected by microscopic examination, may affect the enhancement pattern on HRCT.[12] The tumor size can vary significantly, ranging from several millimeters to as large as 6 cm in diameter in some cases,[13] with some cases exhibiting calcification, particularly in low-grade malignancies.[14] The adjacent lung parenchyma often shows signs of atelectasis or pneumonia secondary to airway obstruction.[15]

During bronchoscopy, PMEC typically presents as an exophytic luminal mass, which may be sessile, polypoid with a broad base attached to the bronchial wall, or pedunculated with a well-defined stalk predominantly occurs in the major airways, particularly within the segmental bronchi, main bronchus.[16]

Histologically, MEC consists of a mixture of cell types, including mucus-secreting glandular cells, squamous cells, and intermediate cells. Based on morphology and cytology, MEC is categorized into low-grade and high-grade types. Low-grade MEC typically exhibits an indolent behavior with a low risk of metastasis, whereas high-grade MEC is associated with a more aggressive course, characterized by local invasion and early metastasis.[16] CRTC1/CRTC3-MAML2 fusion transcripts are detected in approximately 33.7% to 69.7% of MEC cases,[17] and epidermal growth factor receptor (EGFR) is overexpressed in about two-thirds of MEC cases, with less than 5% of cases exhibiting overexpression of human epidermal growth factor receptor 2 (HER2).[18] Although testing for MAML2 gene translocation

is recommended, this was unfortunately not performed in our case due to unavailability.

Surgical resection remains the primary treatment modality for tracheobronchial mucoepidermoid carcinoma (MEC). The main objective is to achieve complete tumor excision with lymph node dissection while preserving as much functional lung parenchyma as possible. Surgical techniques, including pneumonectomy, lobectomy, and sleeve lobectomy, have been shown to facilitate complete resection, particularly for low-grade tumors, which are associated with favorable outcomes. In contrast, high-grade tumors are linked to poorer prognoses.[19] The role of neoadjuvant chemotherapy or radiotherapy remains poorly defined in these cases. Patients with EGFR mutations have shown favorable responses to Gefitinib, a tyrosine kinase inhibitor. Additionally, bronchoscopic neodymium-yttrium-aluminum-garnet (Nd) laser therapy has been utilized in selected cases of MEC.[9] Long-term survival rates following surgical resection are promising, with 87% survival at both 5 and 10 years.[20] Prognosis in PMEC patients is significantly influenced by factors such as TNM stage, histological grade, lymph node involvement, and MAML2 gene translocation.[14]

4. Conclusion

Endobronchial mucoepidermoid carcinoma (MEC) is a rare subtype of salivary gland tumor. Fiber-optic bronchoscopy plays a key role in identifying endobronchial lesions, especially in cases of non-resolving pneumonia. Clinicians should maintain a high index of suspicion for MEC when assessing unusual or persistent pulmonary lesions to ensure timely diagnosis and management.

5. Patient Consent

Written informed consent was obtained from the patient for publication of this case report.

6. Authors' Contribution

All authors contributed to the completion of this work. The final manuscript was read and approved by all authors.

7. Source of Funding

None

8. Conflict of Interest

None

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