Delayed diagnosis of endobronchial mucoepidermoid carcinoma in a 29-year-old male

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ABSTRACT

Mucoepidermoid carcinoma (MEC) is an uncommon primary lung tumor. It usually involves large airways and presents clinically and radiologically with nonspecific features. Because of nonspecific presentation diagnosis is frequently delayed. We report the case of a 29-year-old male patient wherein a clinico-radiological consideration of tuberculosis (TB) led to a prolonged treatment with anti-TB medications without response. Flexible bronchoscopic biopsy confirmed the diagnosis of MEC following that the patient underwent curative surgical resection.

KEY WORDS: Bronchoscopy, endobronchial biopsy, lung tumors, mucoepidermoid carcinoma

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INTRODUCTION

Mucoepidermoid carcinomas (MECs) usually occur in the major and minor salivary glands. Pulmonary MECs are uncommon, and primary endobronchial MECs (EMEC) account for 0.1-0.2% of all pulmonary neoplasms.[1] In the lung, the tumor arises from sub-mucosal glands of the tracheobronchial tree. It is slow growing and usually presents with symptoms related to bronchial obstruction. Symptoms may include cough, hemoptysis, wheezing, and signs of post obstructive pneumonia.^[2] The nonspecific clinical and radiological findings can lead to a diagnostic dilemma and therefore, a considerable delay before correct diagnosis is not uncommon.[3] In tuberculosis (TB) endemic countries, such a clinico-radiological profile may lead an inappropriate consideration of TB as the underlying diagnosis and initiation of anti-TB drugs, further compounding the diagnostic delay.

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CASE REPORT

A 29-year-old never smoker male patient was referred to the Pulmonary Medicine Outpatient Clinic for evaluation of possible drug resistant TB. The patient had received more than 1 year of anti-TB medications with no symptomatic improvement. Patient history and records were reviewed.

Two years ago, the patient developed insidious onset right sided pleuritic chest pain associated with a dry cough. It was associated with on and off fever with evening rise of temperature. There was a history of intermittent wheezing without any seasonal worsening and patient had noted 3–4 episodes of streaky hemoptysis. There was no history of anorexia or weight loss. The patient was evaluated at a local health care facility wherein based on the findings of a right sided chest radiographic opacity and thoracic ultrasound demonstrating mild right side

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pleural effusion, a possibility of pleuro-pulmonary TB was considered, and four-drug anti-TB treatment (directly observed treatment regimen) with first-line drugs was initiated. The patient was reassessed after initial regimen, but there was no radiological response. Sputum smear examination for acid-fast bacilli was negative on multiple occasions. Despite 12 months of regular treatment, there was no improvement in symptoms and cough persisted. The patient was initiated on treatment with an intensified anti-TB regimen (Cat II drugs administered on daily basis for 9 months) along with streptomycin and referred to our center.

General physical examination was normal. On chest examination, crackles were audible in the right infrascapular area, and breath sounds were reduced in the lower right hemithorax. Routine blood investigations were normal. Sputum cytology was negative for malignant cells. A thoracic computed tomography (CT) scan followed by flexible bronchoscopy was planned.

Contrast-enhanced CT thorax demonstrated a mass lesion measuring 5 cm \times 7.5 cm \times 4.5 cm in relation to the bronchus intermedius causing its significant luminal compromise with resultant atelectasis and bronchiectatic changes in middle lobe and lower lobe of the right lung [Figure 1a and b]. No significant mediastinal/axillary lymphadenopathy or pleural effusion was noted. Findings were suggestive of a primary endobronchial mass with parenchymal extension with post obstructive pneumonitis changes. Flexible fiberoptic bronchoscopy showed a lobulated endobronchial growth in proximal right intermediate bronchus completely occluding the middle and lower lobe [Figure 2a]. Endobronchial biopsy showed a malignant neoplasm comprising squamoid cells and mucin-secreting cells showing mild to moderate cytological atypia. Few mucin-secreting cells were seen lining glandular structures and foci of extracellular mucin were noted [Figure 2b]. Occasional mitotic figure was seen, and necrosis was absent. Immunohistochemistry revealed CK5 and CK7 positivity, whereas, CK-20, S-100, and SMA were negative. A diagnosis of MEC (low grade) was given. Positron emission tomography-CT scan showed no significant uptake apart from the right lung mass. The patient underwent

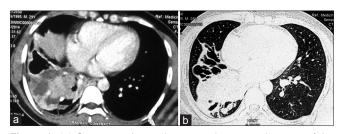


Figure 1: (a) Contrast-enhanced computed tomography scan of the thorax (mediastinal window section) demonstrating a mass in relation to the bronchus intermedius with distal lung parenchyma showing volume loss and mucoid impaction. (b) Computed tomography scan of the thorax (lung window section) demonstrating bronchiectatic changes in the right lower lobe distal to the obstruction

right middle and lower lobectomy and his postoperative period was uneventful. The final histopathology report was consistent with the result of the bronchoscopic biopsy. Follow-up bronchoscopy performed after 1 year showed an intact suture line with no tumor recurrence.

DISCUSSION

MEC is a common tumor of salivary glands, but pulmonary MEC is rare. [4,5] It is considered as a benign tumor with malignant potential. EMEC arises in large airways of the tracheobronchial tree and affects males and females equally with a median age of presentation around 40 years.[6] The patient may present with symptoms directly related to endobronchial involvement such as a cough, wheezing, hemoptysis or those related to post-obstructive pneumonia-like, fever and chest pain. [7] Clinical findings may mimic pulmonary TB. Chest X-ray shows nonspecific features. Bronchoscopy and CT are required for diagnosis, assessment of the extent of involvement and for differentiation from other conditions.[8] On bronchoscopy, EMEC usually appears as an exophytic polypoid luminal mass. Distal to the lesion, bronchus is usually dilated, filled with abundant mucoid material and adjacent lung parenchyma generally demonstrates atelectasis or features of pneumonia.[9] Differential diagnoses of endobronchial masses include pulmonary hamartoma, leiomyoma, endobronchial lipoma, squamous cell papilloma, pleomorphic adenoma, adenoid cystic carcinoma, granular cell tumor, bronchogenic adenocarcinoma, squamous cell carcinoma, small cell carcinoma, and endobronchial carcinoid.[5]

Confirmatory diagnosis is made on histopathology. EMEC is morphologically similar to MEC of major salivary glands. It comprises mucus-secreting, squamous, and intermediate cells that can be organized into different patterns and are classified as low-grade and high-grade lesions. [1,2] High-grade lesions usually demonstrate necrosis, mitosis, and nuclear pleomorphism, while low-grade lesions lack these features as was seen in the present case. [6]

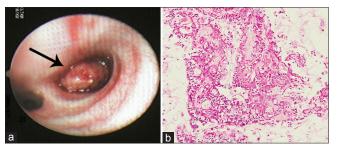


Figure 2: (a) Flexible bronchoscopic image showing a smooth, polypoidal, well-circumscribed endobronchial tumor (arrow) at the orifice of bronchus intermedius. (b) Microphotograph of endobronchial biopsy showing squamoid cells, mucin-secreting cells, some lining glandular structures, and foci of extracellular mucin (H and E, ×100)

Low-grade EMEC is rare and a slow growing tumor. It is usually confined to the bronchus, does not involve adjacent lung parenchyma and presents with nonspecific signs and symptoms. Therefore, the diagnosis is generally delayed as in the present case. [6] It is treated by complete surgical resection of a lobe or a segment, which is associated with excellent prognosis with 5-year survival of 97.6%. [2] Adjuvant radiotherapy is generally not required. In the present case, surgery was performed, and the patient was relieved of his symptoms.

CONCLUSION

This report highlights the importance of keeping a high index of suspicion of an endobrochial growth in patients who present with endobronchial symptoms such as wheezing and hemoptysis. [10] Even in high TB prevalence countries, a thorough clinical and radiological assessment should be performed before treating a patient as sputum smear negative pulmonary TB. Any patient having radiological features suggestive of possible post-obstructive pneumonia should undergo prompt, flexible bronchoscopy to rule out endobronchial growth.

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Conflicts of interest

There are no conflicts of interest.

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