Case Report

Role of fluorine-18-fluorodeoxyglucose positron emission tomography-computed tomography in management of pulmonary mucoepidermoid carcinomas and review of literature

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ABSTRACT

Pulmonary mucoepidermoid carcinoma (PMEC) is a rare tumor of bronchial gland origin with a striking resemblance to MEC of the salivary glands. The World Health Organization classifies PMECs as "salivary gland type" tumors along with pulmonary adenoid cystic carcinomas and epimyoepithelial lung carcinomas. Their description in literature is largely limited to a few case series/case reports. Further, the experience of imaging in these tumors with fluorine-18 fluorodeoxyglucose positron emission tomography-computed tomography (^{18}F-FDG PET-CT) is also limited and evolving largely due to rarity of PMEC. We recently managed an interesting case of a PMEC and reviewed the literature surrounding this rare tumor with an emphasis on the role of ^{18}F-FDG PET-CT in its management. An ^{18}F-FDG PET-CT appears to be a useful imaging modality for predicting the tumor grade of patients with PMECs; further, there is emerging data to suggest the role of ^{18}F-FDG PET-CT for predicting the long-term prognosis of patients with PMEC.

Keywords: Fluorine-18 fluorodeoxyglucose positron emission tomography-computed tomography, grade, lung tumors, pulmonary mucoepidermoid carcinomas, prognosis

INTRODUCTION

Mucoepidermoid carcinomas (MEC) are one of the most common salivary gland malignancies. As a malignant tumor of bronchial gland origin, it was first described by Smetana in 1952. Pulmonary MECs (PMECs) have a presumed incidence of about 0.1–0.2% of all pulmonary tumors.[1] Its description in literature is largely limited to a few case series/case reports; mostly occurring in younger age groups as compared to the other nonsmall cell lung cancers. We recently managed an interesting case of a PMEC in a 46-year-old lady and reviewed the literature surrounding this rare tumor.

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CASE REPORT

A 46-year-old lady with no comorbid conditions presented to our center with a 10 months history of cough and occasional hemoptysis. Clinical examination revealed decreased air entry on the left lower chest. General medical examination and examination of the other systems were unremarkable. A contrast enhanced computed tomography (CT) scan of the chest revealed an ill-defined mass measuring 76 mm × 62 mm × 17 mm in the left lower lobe, which was extending onto the left lower lobe bronchus without any significant mediastinal adenopathy. The rest of the lung parenchyma was normal. An fluorine-18 fluorodeoxyglucose positron emission tomography-computed tomography (^{18}F-FDG PET-CT) scan was done subsequently.

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Figure 1 shows an isolated uptake in the left lower lobe lung (maximum standardized uptake value [SUVmax] 3.3). A bronchoscopic biopsy of the soft polypoidal mass seen extending onto the left lower lobe bronchus was suggestive of a low-grade mucoepidermoid carcinoma [Figure 2a-d]. A formal cardiopulmonary evaluation was subsequently done, and the patient was taken up for surgery. The patient underwent a left lower lobectomy with mediastinal lymph node sampling [Figure 3a and b] and had an uneventful recovery. The final histopathological examination showed an 8 cm × 6 cm × 2 cm polypoidal tumor, with features suggestive of a low-grade MEC without any mediastinal adenopathy and resected with clear margins. The patient was not considered for any further adjuvant therapy and is disease free and on regular follow-up for the past 6 months.

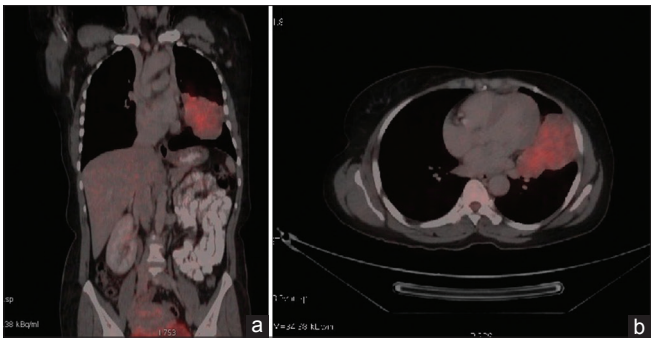

Figure 1: (a and b) Fluorine-18 fluorodeoxyglucose positron emission tomography-computed tomography shows an ill-defined mass measuring 76 mm × 62 mm × 17 mm in the left lower lobe (maximum standardized uptake value [SUVmax] 3.3)

DISCUSSION

PMEC originates from the glandular tissue identical with salivary glands located in the submucosa of the trachea and bronchus. The World Health Organization classifies PMECs as "salivary gland type" tumors along with pulmonary adenoid cystic carcinomas and epimyoepithelial lung carcinomas.[1]

The age group involved ranges from 3 to 78 years of age, with more than half of the cases occurring in patients younger than 30 years of age without any definite sex predilection. There appears to be no association with cigarette smoking or other risk factors implicated in the etiopathogenesis of nonsmall cell lung carcinomas. Patients with PMEC typically present with symptoms related to bronchial obstruction and atelectasis, such as cough, hemoptysis, wheezing, and postobstructive pneumonia; however, about 9–28% of patients have no definitive symptoms.[2] The clinical picture is strikingly similar to that of other inflammatory conditions such as asthma, chronic obstructive pulmonary disease, or pneumonia.

PMECs tend to have a central/endo or peribronchial location. Hence, bronchoscopic examination is critical in establishing the diagnosis. The CT image findings of PMECs are similar to other types of benign and malignant pulmonary nodules. Because the clinical and radiographic manifestations of PMEC are nonspecific, the diagnosis mainly relies on the histopathology.

Histologically, PMEC is comprised a mixture of various cell types including mucin-secreting glandular cells, squamous cells, and intermediate cells. Low-grade MEC is distinguished from high-grade MEC based on the lack of cytological atypia including nuclear pleomorphism and the absence of mitotic activity and necrosis.[3] The histological features helpful in differentiating PMECs from the other nonsmall cell lung cancers include the presence of mucous cells, rearrangement of MAML2, p63 expression, and lack of keratinization. TTF-1 and Napsin are typically not expressed in PMECs.[3]

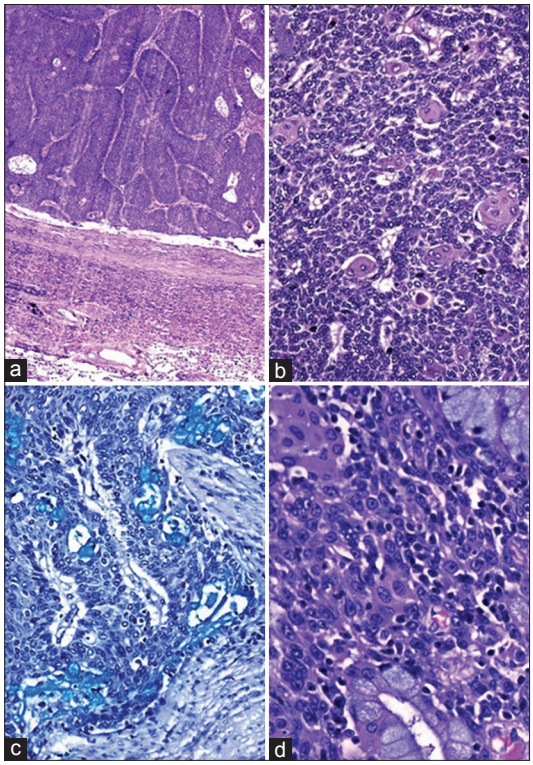

Figure 2: Histology suggestive of a low-grade pulmonary mucoepidermoid carcinoma (a) compressed lung tissue, (b) a foci of squamous differentiation and keratin pearls, and (c and d) cystic spaces filled with eosinophilic material lined by malignant columnar epithelium (H and E, ×20)

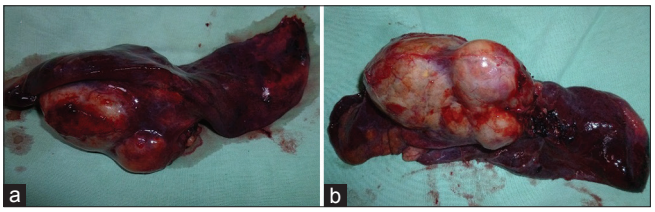

Figure 3: (a and b) Specimen photograph of the left lower lobectomy shows the 8 cm × 6 cm × 2 cm polypoidal tumor

The use of ^{18}F-FDG PET-CT in the management of PMEC was first reported by Kinoshita *et al.*[4] in 2005, many studies have tried to address this subject ever since. Studies have reported

^{18}F-FDG PET-CT to be a useful preoperative imaging modality for predicting tumor grade, nodal stage, and postsurgical prognosis.[2,5-8] Patients with SUVmax >6.5 were more likely to have a high-grade tumor, lymph node metastasis, and cancer recurrence.[8] A recent study reported the prognostic value of ^{18}F-FDG PET-CT in the long-term outcomes of patients with PMEC.[8]

Surgical resection with negative surgical margins remains the standard therapy for patients with PMEC.[5,9] Many different operative approaches have been described, including thoracotomy with conventional lobectomy, video-assisted thoracoscopic surgery lobectomy, sleeve lobectomy, and lobectomy with bronchoplastic closure with similar outcomes.

Histologic grade, tumor stage, and lymph node metastasis have been reported to be independent prognostic factors in patients with PMEC.[8,9] These prognostic factors, especially tumor grade have been verified in many studies. It is now accepted that low-grade PMEC behaves like a benign or low-grade malignant tumor, whereas high-grade PMEC shows aggressive characteristics such as any other nonsmall cell lung cancer.[8,9] The use of adjuvant therapy in the form of chemotherapy, targeted therapy, or radiation therapy is hence not recommended for low-grade PMECs given the excellent survival rates achieved with surgery alone (5 years survivals >95%).

Locally advanced high-grade PMECs have a guarded prognosis with the majority of patients succumbing to their disease. Effective treatment measures for high-grade PMECs have not been clearly established, although aggressive mediastinal lymph node dissections and adjuvant therapies have been attempted with limited success. Combination chemotherapy protocols are usually reserved for metastatic or inoperable tumors.

Molecular profiling of PMECs has identified a few "druggable" targets; however, the role of targeted therapies directed against EGFR or a novel CRTC1-MAML2 fusion protein expressed in some high-grade tumors is yet to be determined.[3,10]

CONCLUSION

PMEC is a rare malignant neoplasm with a striking resemblance to mucoepidermoid carcinoma of the salivary glands. The ^{18}F-FDG PET-CT appears to be a useful imaging modality for predicting the tumor grade of patients with PMECs; further there is emerging data to suggest the role of ^{18}F-FDG PET-CT for predicting the long-term prognosis of patients with PMEC.

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Conflicts of interest
There are no conflicts of interest.

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