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Case Report

Mucoepidermoid Carcinoma Lung- Both Primary and Metastasis: A Case Report

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Abstract: Mucoepidermoid carcinoma (MEC) is a rare malignant tumour of lung, accounts for <1% of all lung carcinomas. MEC is a salivary gland-type tumor that consists of mucin-secreting cells,squamoid cells and intermediate-type cells. Lung is the most common site of distant metastasis from MEC salivary gland and lung is a rare site of Primary MEC. We report two cases of Mucoepidermoid carcinoma lung. First case is a 59 year old lady who presented with fever, cough and breathlessness.Imaging studies revealed a mass involving left lower lobe bronchus. Lobectomy was done and the diagnosis was MEC.Second case case is a 50 year old lady who had a previous history of total conservative parotidectomy MEC right parotid gland at the age of 35 years.After 15 years, she presented with cough and breathlessness. Imaging studies showed multiple lesions in bilateral lung fields, liver, mediastinal and supraclavicular nodes, with possibility of metastasis. Pleural fluid and biopsy revealed MEC metastasis.We also emphasize here the utility of fluid cell block study and special stains like mucicarmine to reach the diagnosis of MEC.
Keywords: Mucoepidermoid carcinoma (MEC), lung, primary and metastasis.

INTRODUCTION

Primary MEC lung is a rare salivary gland type tumor of lung which accounts for less than 1% of all pulmonary tumors (Adel, K. *et al.*, 2017; William, D. *et al.*, 2015). Histologically it is similar to those arising from major salivary gland which is an admixture of squamous (epidermoid) cells, mucus secreting cells and intermediate cells. Primary MEC is believed to be arising from minor salivary glands lining the trachea-bronchial tree (Anirban, H. *et al.*, 2017; Ahmed, Q. *et al.*, 2016). Mucoepidermoid carcinoma(MEC) is the most common malignant salivary gland neoplasm, mostly in parotid.Incidence of distant metastasis from MEC of salivary glands is about 13%, most common site being lung, followed by liver, bone, skin, intestine and so on (Adel, K. *et al.*, 2017; Anirban, H. *et al.*, 2017). Here we report two cases of Mucoepidermoid carcinoma lung; one is a case of Primary MEC lung and the other is a case of pulmonary metastasis from MEC parotid gland.

CASE PRESENTATION

Case1

A 59 year old lady presented with fever and cough for 1 week duration. She had acute onset of breathlessness. On examination ,there was reduction in breath sounds on left side with bilateral crepitations and rhonchi.Chest X ray and CECT thorax revealed a mass lesion involving the left lower lobe towards the hilum(figure a).Fibreoptic bronchoscopy showed a polypoidal endobronchial mass projecting through the left main bronchus(figure b). We received Left lower lobectomy specimen .Gross examination showed a whitish polypoidal firm mass seen protruding near the bronchus.Cut section showed an irregular whitish growth measuring 5x3x2 cm.Growth is seen involving the resected end of bronchus(figure c). Microscopy showed a neoplasm composed of squamous cells, mucus secreting cells and intermediate cells arranged in nests and sheets with focal lymphocytic infiltration of stroma (figure d). Mucicarmine stain was positive (figure e).

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Diagnosis: Mucoepidermoid carcinoma,low grade lung-Primary.

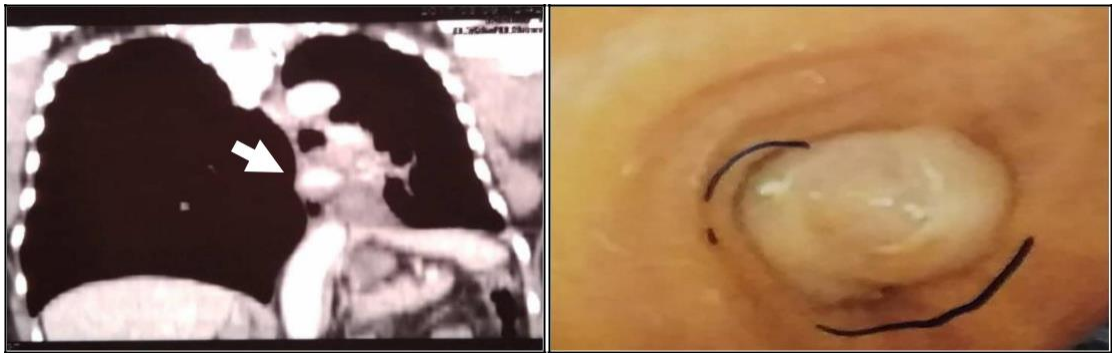

Figure (a) show CECT thorax showing a mass lesion involving left hilar region. Figure (b) show bronchoscopic view of endobronchial polypoidal mass protruding through the left bronchus.

Figure (c) Macroscopy shows an irregular whitish growth involving the adjacent bronchus obliterating the lumen

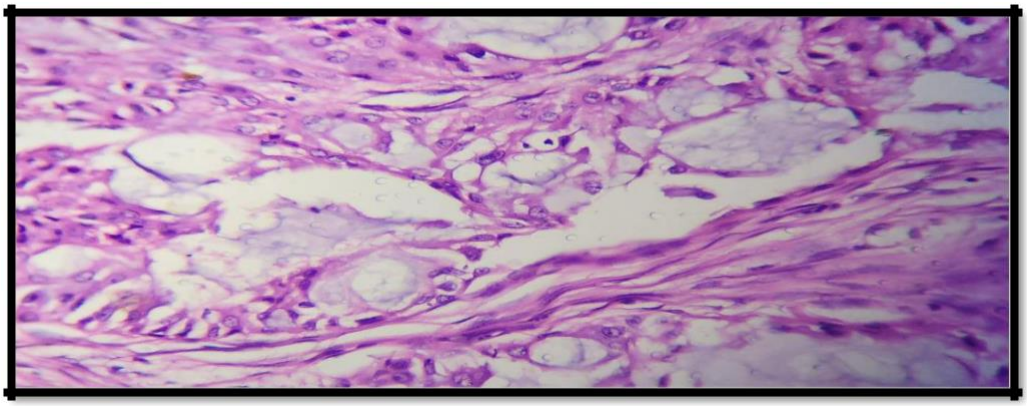

Figure (d) H & E Stain 400X showing mucus secreting cells,squamoid cells and intermediate cells in glandular pattern and nests.

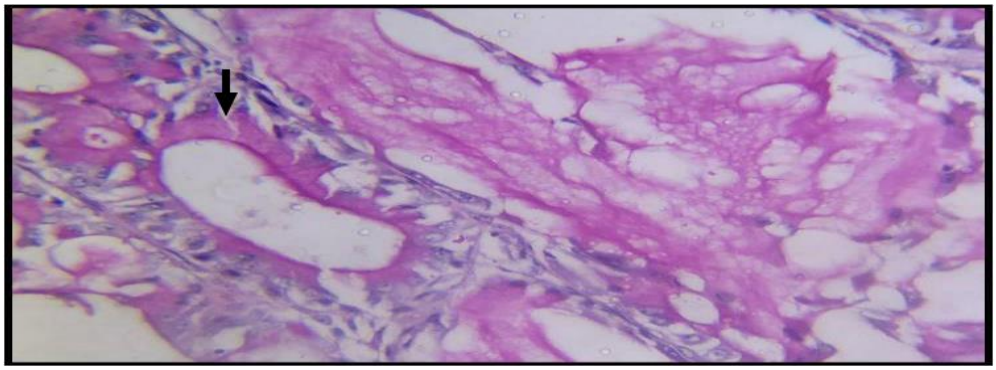

Figure (e) Mucicarmine stain 400X showing mucicarmine positivity.

Case 2

A 50 years old lady had a previous history of total conservative parotidectomy for MEC right parotid gland at the age of 35 years. Histopathological report was Mucoepidermoid carcinoma –low grade with excision margins involved; thereafter she received chemoradiation and was on regular follow up. Follow up for the past 3 years was lost. After 15 years, she presented difficulty in breathing and non-productive cough for duration of 2 months.On examination, she was pale with right supraclavicular lymphadenopathy. Examination of chest showed decreased movement and breath sounds on right side with dullnote on percussion.Chest X-ray revealed moderate right sided pleural effusion obscuring the lung fields. Fluid was tapped by chest tube insertion and her symptoms almost relieved (figure f). For further evaluation, CECT thorax was done, it showed multiple nodular lesions in bilateral lung fields with a right hilar mass lesion and similar lesions in right lobe of liver, mediastinal and right supraclavicular lymphnodes, likely metastasis (figure g).Thoracoscopy revealed multiple discrete nodules involving parietal pleura and diaphragm. We received 300 ml yellow coloured pleural fluid for cytological study, which was positive for malignant cells. Cell block study of the same showed three populations of cells; squamous, mucus secreting and intermediate cells. Mucicarmine staining was positive (figure h and i). We received thoracoscopic pleural nodule biopsy, which showed a neoplasm composed of squamous cells, mucus secreting cells and intermediate cells arranged in nests, sheet and diffusely with focal lymphocytic infiltration of stroma. Mucicarmine stain was positive (figure j).

Diagnosis: Mucoepidermoid carcinoma,high grade lung- Metastasis from salivary gland

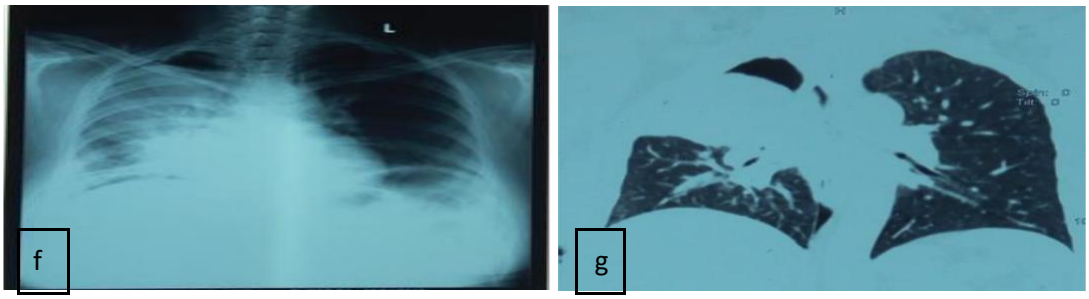

Figure (f) shows chest X ray showing right sided opacity in the lung. Figure (g) shows right sided upper lobe mass lesion and multiple nodules in bilateral lung fields.

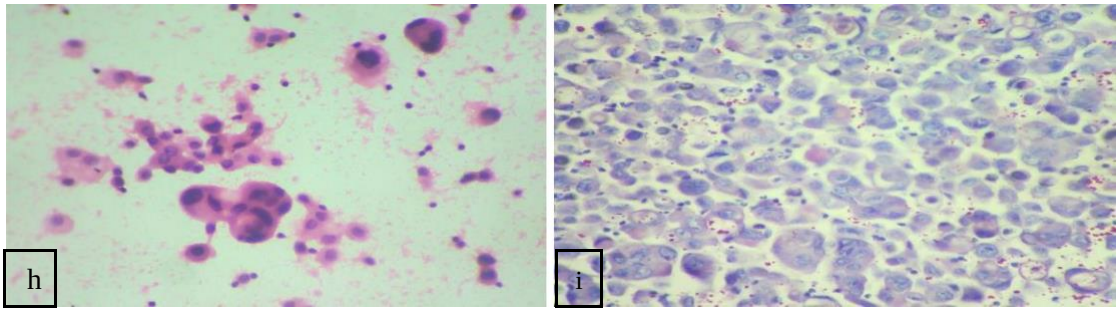

Figure (h) Pleural fluid cytology shows malignant cells. Figure (i) cell block showing mucicarmine positivity.

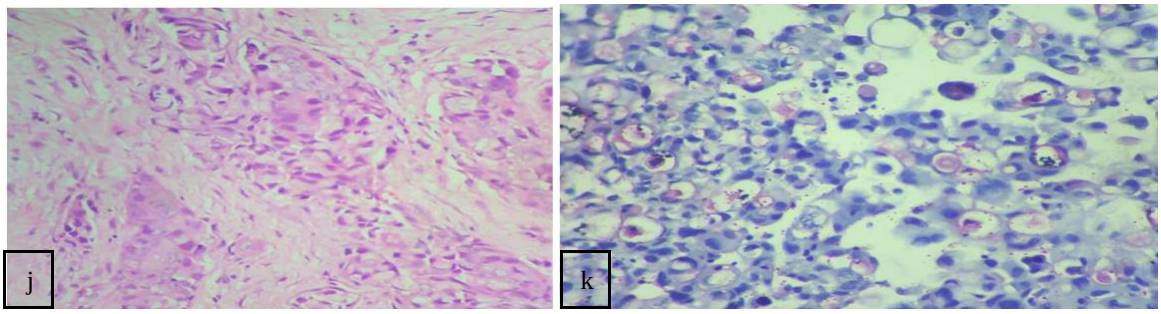

Figure (j) Pleural biopsy: H& E stain 400X showing mucus secreting cells, squamoid cells and intermediate cells.Mucicarmine stain show positivity.

Discussion

Mucopidermoid carcinoma (MEC) is the most common malignant tumor of salivary gland and a rare primary tumor of lung which constitutes about <1% of all lung tumors (Adel, K. *et al.,* 2017; William, D. *et al.,* 2015). Diagnosis of Mucoepidermoid carcinoma

can be made based on presence of squamous (epidermoid) cells, mucus secreting cells and intermediate cells in varying proportions in both cytology and histology (Adel, K. *et al.*, 2017; Anirban, H. *et al.*, 2017). Now a days, MEC grading is done using Three tier systems (example:Brandwein system and AFIP system) and can be classified as low,intermediate and high grade MEC (Derek, K. O. *et al.*, 2004; Ahmed, Q. *et al.*, 2016). High grade MEC is more aggressive and most likely to recur and metastasize to distant sites, most commonly in lungs, followed by liver,bone,skin,intestine and so on (Adel, K. *et al.*, 2017; Ahmed, Q. *et al.*, 2016). Primary MEC occurring in lung is believed to be originating from submucosal minor salivary glands in large airways, usually present as exophytic endobronchial mass. First case is a 59 year lady presented as left endobronchial mass (Anirban, H. *et al.*, 2017). Second case is a 50 year old lady with bilateral pulmonary lesions and similar lesions in liver and lymphnodes with previous history of Mucoepidermoid carcinoma 15 years back which showed the possibility of pleural metastasis in this case. As initial diagnosis of MEC parotid was made at the age of 35 years and she was managed with Total conservative parotidectomy to avoid facial nerve palsy. Incomplete removal of tumor and loss of follow up may be the reason for late distant metastasis in this patient. Available studies showed that genetic aberrations leading to MEC in lung and salivary gland is the same (William, D. *et al.*, 2015).A fusion gene CRTC1-MAML2 occurs in both low grade and high grade Mucoepidermoid carcinoma also seen in the salivary gland counterpart. There is no separate immunohistochemical marker available to prove the bronchial origin.Primary MEC lung lacks TTF-1 and Napsin A expression which makes difficulty in proving bronchial origin John, R. *et al.*, Cytological evaluation including cellblock study with supportive special stains like Mucicarmine can predict the possibility of Mucoepidermoid carcinoma which can be confirmed by histopathology.Strict follow up with adequate imaging studies and pathological confirmation by cytology and histology of specimens are important to pick up the cases of late metastasis.

CONCLUSION

Mucoepidermoid carcinoma is a malignant neoplasm of salivary gland, mostly involving parotid and rarelyoccur as primary in lung (Adel, K. *et al.*, 2017; William, D. *et al.*, 2015). Lung is the most common site of metastasis from MEC salivary glands (William, D. *et al.*, 2015). Histological grade, Tumor staging and tumor resection margins are the important prognostic factors contributing to late recurrence and distant metastasis (Anirban, H. *et al.*, 2017; Ahmed, Q. *et al.*, 2016). Possibility of metastatic pleural effusion from Mucoepidermoid carcinoma can be picked up using cell block study supported by special stains like Mucicarmine , with histopathology being the gold standard for final diagnosis (Jennifer, J. *et al.*, 2016).

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