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Primary Adenoid Cystic Carcinoma of Bronchus- A Rare Case Report

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Abstract: **Adenoid cystic carcinoma (ACC) is a malignant epithelial neoplasm most commonly presents in the minor salivary glands and also present in the respiratory tract as counterpart of salivary gland tumor arising from a sub mucosal gland of the tracheobronchial tree in the respiratory tract. ACC of the lung and bronchus are rare locally aggressive malignant tumor often require extensive surgery or palliative therapy is given in cases of inoperable tumor. We report a case of ACC of bronchus in a 30 year old male presenting with the chief complaints of non-productive cough and haemoptysis.**

Keywords: **Adenoid cystic carcinoma, Bronchus, Salivary gland.**

I. INTRODUCTION

Primary adenoid cystic carcinoma of lung and bronchus comprises less than 1% of all lung malignancy.[1,6,9]

Within the tracheobronchial system, ACC are the most common salivary-type gland tumors of the trachea, accounting for 20% to 35% of tracheal tumors and 75% to 80% of all tracheobronchial gland tumors.[2,3,8] Within the lung parenchyma adenoid cystic carcinomas are much less common.[4]

It has an equal sex distribution and tends to occur in the fourth and fifth decades of life.[5,9] They may present as nodules within, or a generalized constriction of, the major airways. Symptoms, usually resulting from partial airway obstruction, include wheezing, progressive shortness of breath, and hemoptysis.[8,9]

II. CASE STUDY

30 year old male patient was present to the pulmonary department with the chief complaints of cough and haemoptysis since 2 months. He had proximal airway obstruction with shortness of breath, cough, wheeze, chest pain and haemoptysis. Cough was non-productive. On chest examination crepitation was present.

X ray chest shows soft tissue mass around the left side of bronchus.

CT scan of chest reveal a hypodanse soft tissue mass present around the left side of bronchus. Mass was hypodanse and causes narrowing of lumina and collapse of lung. Right side of lung parenchyma appears to be normal.

Bronchoscopy shows a growth present around the left side of bronchus measuring 3x2x1.5cm. The growth was well-defined circular and outer surface was smooth. Growth was irregular and causes narrowing of bronchus. Biopsy was taken from growth and sent to our department for histopathological examination.

Received several gray, white soft tissue pieces collectively measuring 0.2x0.2 cm.

Microscopy- Section shows an unencapsulated, infiltrating neoplasm consisting predominantly of classic cribriform patterns demonstrating arrangement of cells in a “Swiss cheese” configuration with many oval or circular spaces. These spaces contain hyalinized light eosinophilic material. At places tubular pattern also present. The tumor cells are composed of uniformly sized cells with small, hyperchromatic round to oval nuclei, with scanty eosinophilic to clear cytoplasm, and indistinct cell borders. The cystic spaces seen in the cribriform pattern are pseudocysts, which are extracellular and lined

by a basement membrane. Ductal cells are scattered among these abluminal cells, which surround small true lumens forming glands. The interstitial stroma was from myxoid to hyalinized. Cellular and nuclear pleomorphism, necrosis, and mitotic activity are absent.

On the basis histopathological finding a diagnosis of ACC was made. The patient was referred to higher centre for thoracic surgery due to lack of facility at our centre. Patient loss follows up after this.

III. FIGURES

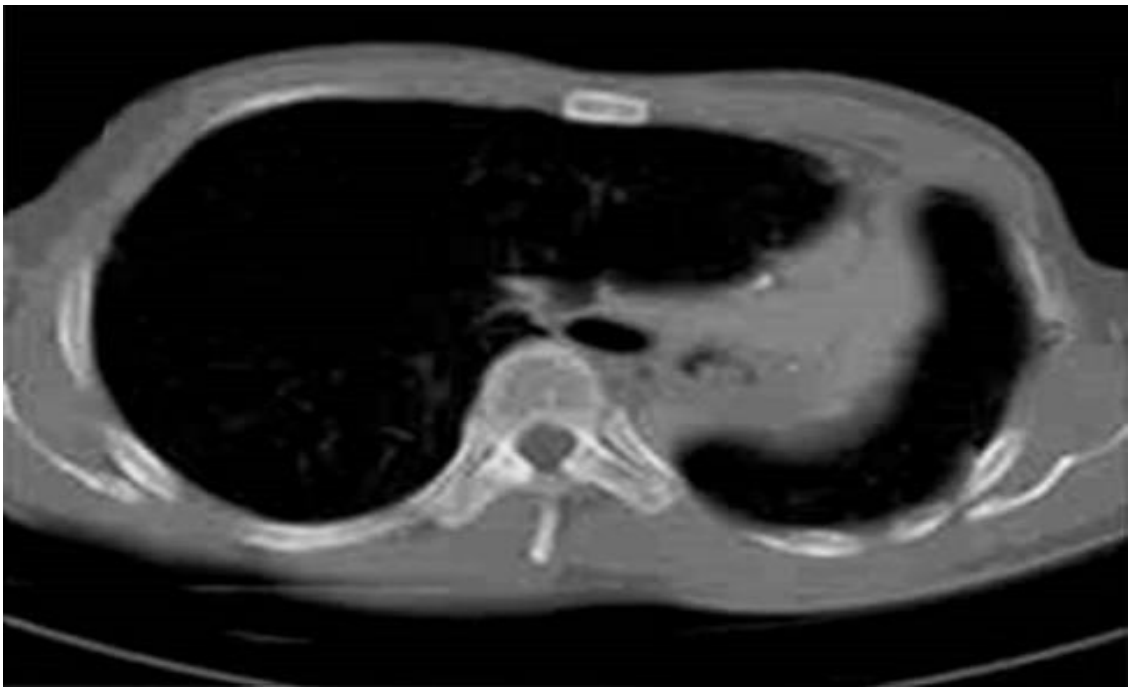

Figure 1-Chest computed tomography scan showing hypodanse lesion around the left bronchus.

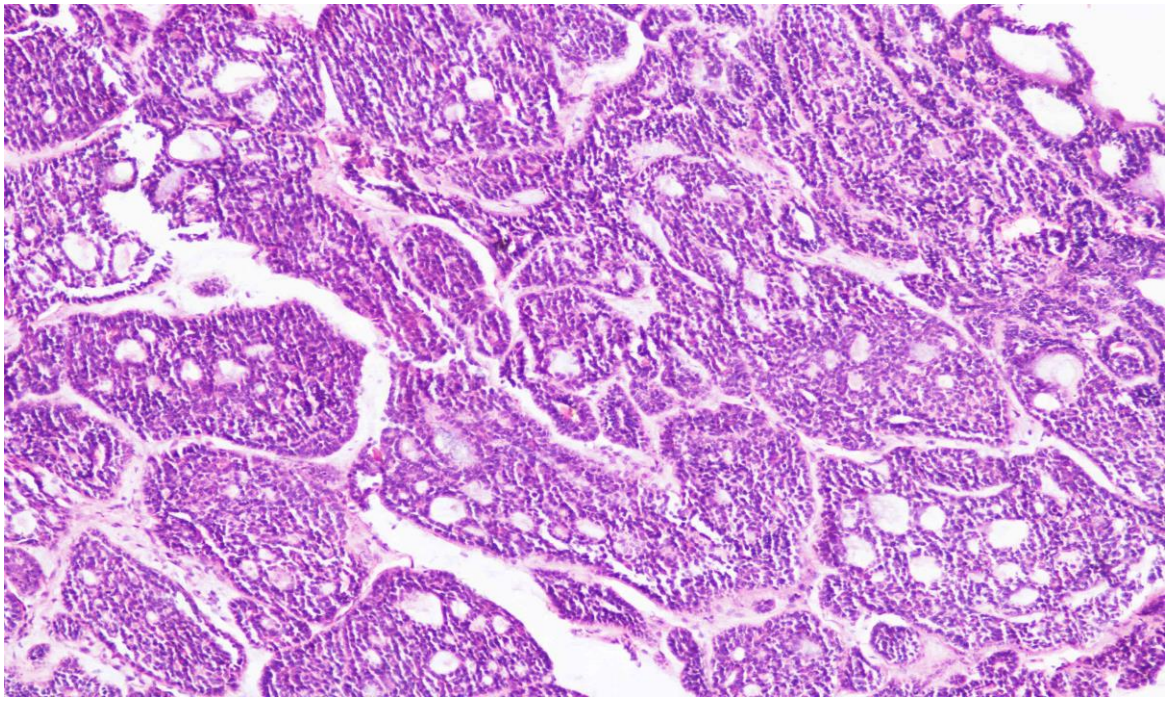

Figure 2- Section shows predominantly of cribriform patterns (H&E, Low Power)

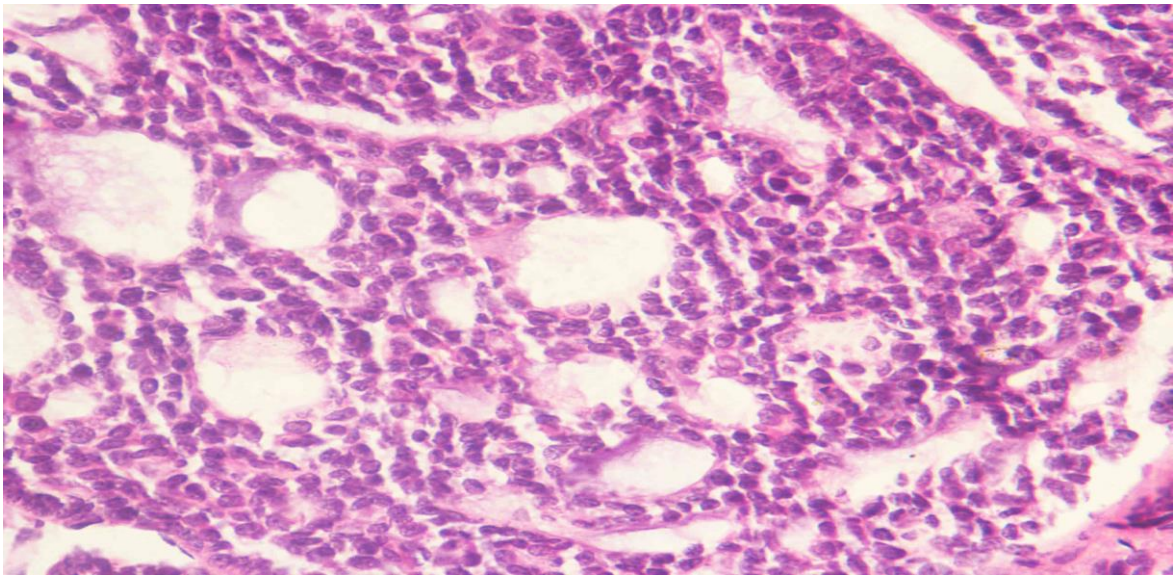

Figure 3- Section shows tumor cells arrange around oval or round cystic spaces forming cribriform pattern (H&E, High power)

IV. DISCUSSION

Adenoid cystic carcinoma, also called cylindroma, is a malignant tumor that can arise from a primitive cell of sub mucosal glands of the respiratory tract, or tracheobronchial gland, which shows differentiation characteristics of ductal and myoepithelial cells and most commonly involved site in the lung is the trachea, and rarely involve bronchial tree and comprises less than 1% of all lung tumours.[7,8 ,9]

ACC of respiratory tract presents most commonly in the fourth and fifth decades of life with an equal sex incidence behaves in an insidious and indolent fashion with multiple local recurrences preceding metastases with9. ACC not associated with cigarette smoking or other risk factor [8, 9].

Within the tracheobronchial system, adenoid cystic carcinomas are the most com-mon salivary-type gland tumors of the trachea, accounting for 20% to 35% of tracheal tumors and 75% to 80% of all tracheobronchial gland tumors.[5,8] Within the lung parenchyma adenoid cystic carcinomas are much less common[8]. In some instances, a peripheral (apical) pulmonary location may result in the development of a pancoast syndrome. [8]

Symptoms, usually resulting from partial airway obstruction, include wheezing, progressive shortness of breath, and hemoptysis. Hemoptysis is caused by surface ulceration, but may also be related to the effects of more distal obstruction. Other obstructive symptoms may include chronic cough, which becomes more productive, fever, and general cachexia in advanced cases.[7, 8, 9]

Adenoid cystic carcinomas are rarely diagnosed on FNAC. Cytodiagnosis of adenoid cystic carcinomas include features such as three-dimensional clusters of basaloid cells and hyaline basement membrane material forming cylinders or spheres that are sharply demarcated from the tumor cells. Monotonous tumor cells and mucinous spaces and separation by often hyalinized stroma are helpful indicators of adenoid cystic carcinoma. 8

The histochemical features of ACC include the presence of diastase-resistant, periodic acid–Schiff–positive, and mucicarmine-positive material within the pseudo-cysts. Alcian blue staining is also present within the pseudocysts. The immuno-histochemistry of ACC varies according to cell type. The myoepithelial cells show cytokeratin, S-100 protein, p63, calponin, vimentin, and actin positivity with variable glial fibrillary acidic protein reactivity. The ductal cells show cytokeratin, EMA, and carcinoembryonic antigen positivity.[8,9]

Differential diagnosis of ACC includes adenocarcinomas, small cell carcinoma and metastatic adenocarcinomas. In small cell carcinoma typical solid sheet of cell with euchromatin, nuclear crowding or moulding and without adenoid pattern. Rarely solid areas with neuroendocrine markers (such as CD56, chromogranin, and synaptophysin) will be demonstrate to exclude small cell carcinoma occurring in combination with glandular components.[8]

Metastatic adenoid cystic carcinomas of lung, which in contrast to primary lesions are usually peripheral, small, and multiple.

A correlation between the clinical behavior of ACC and their histological patterns has been suggested. The solid histological pattern has been associated with a more aggressive clinical course and early distant metastases, in contrast to the cribriform type which shows a more benign behavior. However in our case, the cribriform subtype-predominant pattern showed a very aggressive clinical course that is quite unusual for such tumors. Surgical resection appears to be the mainstay of treatment in these tumors. Adjuvant radiotherapy and chemotherapy has been used in a limited number of patients.

The behavior of ACC is one of multiple recurrences with late metastases and survival needs to be analyzed over a prolonged period (10-15 years).[9] Patients are prone to develop local recurrence because of difficulty obtaining clear margins and it is recommended that margins of resection be analyzed by frozen section at the time of primary surgery[1,9].

Primary treatment is surgery with supplemental radiation, especially by linear accelerator. Poor prognosis is related to stage of the tumour at the time of diagnosis, the presence of positive margins, and a solid cellular growth pattern[1,9].

V. CONCLUSION

Adenoid cystic carcinoma is a rare tumor of bronchus and it is a locally aggressive tumor requires extensive surgery often diagnosed by biopsy. Alternatively radio-therapy given for palliative therapy. So Adenoid cystic carcinoma always kept as a differential diagnosis in the tumor of respiratory tract.

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