Case Report

Bronchial mucous gland adenoma presenting as massive hemoptysis: A diagnostic dilemma

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ABSTRACT

Mucous gland adenoma of the lung is an uncommon benign tumor that histologically resembles the mucus-secreting component of tracheobronchial gland. The majority arises within the main, lobar, or segmental bronchi but parenchymal involvement had also been reported. We herein present a case of mucous gland adenoma arising from the left lower lobe bronchus. The 32-year-old female presented with massive hemoptysis, productive cough, and dyspnoea and was clinically misdiagnosed as tuberculosis. Radiology proved to be inconclusive. This case highlights the importance of a complete lung work up in patients presenting with signs of respiratory tract infections.

KEY WORDS: Bronchus, hemoptysis, mucous gland adenoma

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INTRODUCTION

The bronchial mucous gland adenoma is an extremely rare and benign lung tumor. It is composed of mucus-containing acini-lined by cuboidal cells without pleomorphism. The majority arises within the main, lobar, or segmental bronchi.[1] MGA presenting as a peripheral lung mass is extremely rare. The lesion is classified as a benign epithelial tumor of the lung, among the salivary gland-type adenomas.[2] Total surgical resection is usually required for complete diagnosis and treatment. The main differential diagnoses are a low-grade mucoepidermoid carcinoma, primary adenocarcinoma, and benign adenomatous lesions of lung. We herein report a case of this rare tumor presenting as massive hemoptysis.

CASE REPORT

A 32-year-old Kashmiri woman was referred to our hospital with chief complaints of hemoptysis, cough, and fever of 2-year duration. She was diagnosed as pulmonary tuberculosis and took antitubercular treatment for 6 months. But gradually she started developing chest pain and dyspnoea. There was a past history of fibroadenoma of right breast. There was no history of nausea, vomiting, diarrhea, or weight loss. She was a nonsmoker. Laboratory investigations revealed hemoglobin of 9.0 g/dl with normal total leucocyte and platelet counts. Erythrocyte sedimentation rate was 53 mm at the end of first hour. Liver and kidney function tests were within normal range. HIV, hepatitis B virus, and hepatitis C viral serologies were negative. Chest X-ray showed consolidation and collapse of left upper lobe. Contrast-enhanced computed tomography (CECT) chest showed a well-circumscribed homogeneously enhancing mass lesion in the apical segment of left lower lobe occluding the left lower main bronchus. There was no evidence of any calcification or necrosis on CT scan [Figure 1a]. Radiological diagnosis of pulmonary hamartoma or intrapulmonary hydatid cyst was made. Fiberoptic bronchoscopy done revealed a polypoidal growth arising from the lower lobe-left causing occlusion of left main bronchus and suggested the possibility of a carcinoid tumor. Finally preoperative diagnosis of carcinoid tumor arising from the left lower lobe bronchus involving the apical segment of the lower lobe was made. A left lower lobectomy was performed via left posterolateral thoracotomy and specimen was sent for histopathological examination. Macroscopic examination of the resected specimen revealed a whitish solid mass,

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5.5 cm in maximum diameter, arising close to the bronchus and compressing the adjacent lung parenchyma. The cut surface showed mucoid material without hemorrhage or necrosis. On close examination, several small cystic spaces were noted [Figure 1b]. Microscopically, the tumor was well circumscribed, lined by respiratory epithelium, and consisted of numerous irregularly arranged cysts, tubules, and glands lined by bland columnar, cuboidal, or flattened, mucus secreting cells. Tumor was found projecting into the bronchial lumen, however, not invading the underlying cartilage [Figure 2a and b]. There was no cytological atypia or mitoses. No intermediate cells or squamous component of mucoepidermoid carcinoma was found. The intervening stroma consisted of delicate connective tissue and lymphoplasmacytic infiltrate [Figure 2c]. The lumen contained mucus that was alcian blue positive [Figure 2d]. The Ki-67 proliferation index was low (less than 1%). Immunohistochemistry for thyroid transcription factor-1 was negative [Figure 2e]. Taken together, these observations led us in making a diagnosis of a bronchial mucous gland adenoma. The postoperative course was uneventful and the patient made a complete recovery.

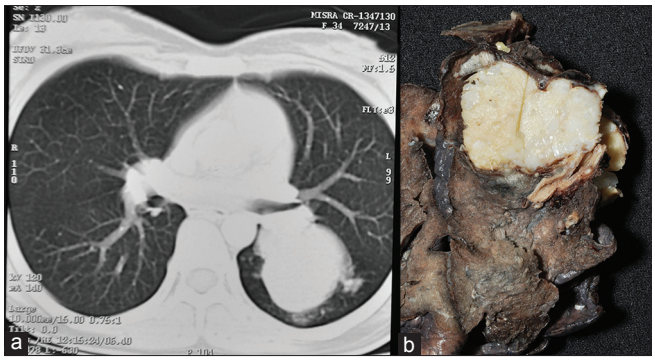

Figure 1: (a) CECT Chest showing a well-circumscribed homogenously enhancing mass lesion in the apical segment of left lower lobe, (b) Cut surface of mucous gland adenoma showing mucoid gelatinous surface with numerous small cysts of variable sizes. Tumor is arising from the bronchus and compressing the adjacent lung parencyma

DISCUSSION

Mucous gland adenoma is a rare benign tumor originating from the mucous secreting glands of the larger airway mucosa. This tumor was first reported in 1882 by Muller as a pathologic entity separate from carcinoma of the lung and was first named bronchial adenoma arising from mucous gland.[3] The majority of the cases are seen in the bronchus, but it has also been described in the trachea or peripheral airways.[4-7] The tumor occurs with equal frequency in men and women and at any age (mean 52), including children.[1] Most commonly reported symptoms are cough, fever, and recurrent pneumonia and these are the result of bronchial obstruction by the tumor. The clinical presentation may be mistaken for asthmatic disorders or chronic obstructive pulmonary disease for long periods of time.[6,8] Other nonspecific observations such as a chronic cough, unilateral wheezing, hemoptysis, or pulmonary infections can also be present for a long time before a correct diagnosis is made.[9]

In this case, the patient had symptoms since 2 years and was wrongly diagnosed as tuberculosis and treated with ATT for 6 months. Even radiology examination proved to be inconclusive. There is nothing on bronchoscopic findings that would distinguish mucous gland adenoma from other neoplastic and non-neoplastic bronchogenic lesions, although the presence of surface microcysts and a gelatinous coating over the surface of tumor is more in keeping with mucous gland adenoma. Thus, removal of the tumor and pathological examination are key to the diagnosis.

Pulmonary adenomas are uncommon and can be classified as alveolar adenoma, papillary adenoma, mucinous cystadenoma or adenomas of the salivary-gland type, which

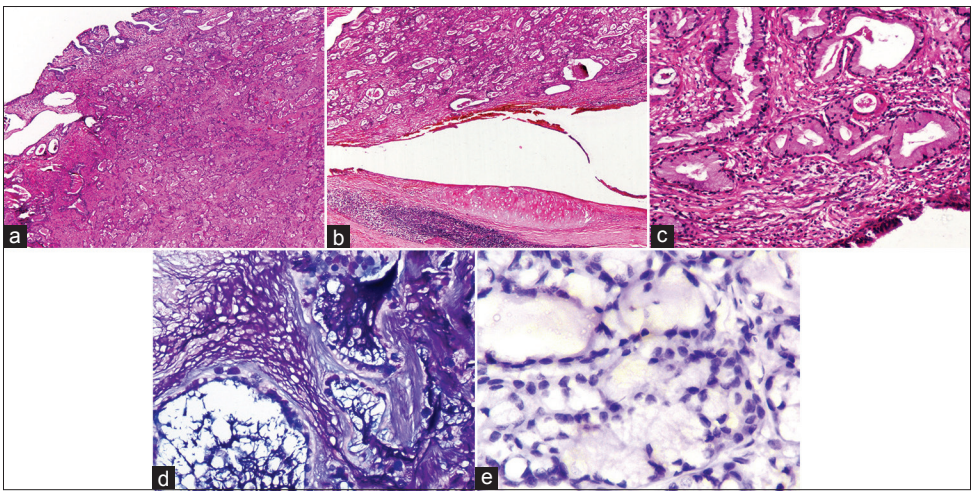

Figure 2: (a) Tumor is lined by respiratory epithelium and consists of numerous irregularly arranged tubular glands (H and E, ×20), (b) Photomicrograph showing tumor projecting into the bronchial lumen without invading the underlying cartilage (H and E, ×20), (c) The glands are lined by single layer of mucous secreting tall columnar cells and the interveining stroma shows lymphoplasmacytic infiltrate (H and E, ×20), (d) Glands showing alcian blue positivity (Periodic acid-Schiff and alcian blue stain, ×40), (e) Epithelial cells showing TTF-1 negativity (IHC, ×40)

include pleomorphic adenoma, monomorphic adenoma, myoepithelioma, and mucous gland adenoma.[10] Histological differential diagnoses include low-grade mucoepidermoid carcinoma, primary adenocarcinoma as well as the benign adenomatous lesions, glandular papilloma, papillary adenoma, alveolar cell adenoma, and mucinous cystadenoma. The mucoepidermoid carcinoma is a rare malignant tracheobronchial tumor but encountered more frequently than the bronchial mucous gland adenoma. The careful assessment of squamous and intermediate cells will confirm this diagnosis.[1,6] An adenocarcinoma shows the typical features of malignancy, such as cytological atypia, mitoses, and an infiltrative growth pattern along with strong TTF-1 immunopositivity in the neoplastic glands.[11] Immunohistochemically, a bronchial mucus gland adenoma usually expresses high molecular weight keratins, but it is negative for TTF-1.[11] The glandular papilloma has an endobronchial growth pattern and typical fibrovascular cores, lined by ciliated or nonciliated columnar cells and a varying proportion of cuboidal and goblet cells.[12] Mucinous cystadenomas, papillary adenomas, and alveolar cell adenomas are parenchymal lesions. Mucinous cystadenoma of the lung is a benign lesion histologically similar to the bronchial mucous gland adenoma. However, it occurs in the peripheral pulmonary parenchyma and is a true mucin-filled cyst lined by mucous epithelium, with variable expression of TTF-1 antigen.[13] A papillary adenoma, however, consists of fibrovascular cores lined by cuboidal or columnar epithelium and it is positive for TTF-1, whereas alveolar adenomas are well-circumscribed unencapsulated multicystic masses with ectatic spaces lined by cytologically bland flattened, cuboidal, and hobnail cells that show positivity with broad spectrum keratins, TTF-1, and CEA.[14] Mucous gland adenoma is a benign tumor and does not recur or metastatise. Nevertheless, some authors have pointed out the malignant proliferative potential of the tumor.[7] There is no tumor recurrence in our patient 6 months after the surgery. The treatment of choice is the complete excision of the mucous gland adenoma involving the bronchus or pulmonary segment or lobe.[15]

CONCLUSION

Benign glandular tumors of the tracheobronchial tree remain out of the limelight of pulmonary medicine. They are capable of provoking mechanical and symptomatic bronchial obstruction. Nevertheless, many patients with mucous gland adenoma go on for years with repeated hemoptysis and recurrent pneumonias prior to the diagnosis. Recognition of this unusual neoplasm by bronchoscopist and pathologist is vital. This observation gives credence to the necessity of an in-depth lung workup in patients presenting with signs of respiratory tract infections and such unusual differential diagnoses should be considered.

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