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Case Report

Masquerading Adenoid cystic Carcinoma of Trachea. A case report

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ABSTRACT
Primary malignant tracheal tumors are not common and adenoid cystic carcinoma (ACC) of trachea is very rare. Hereby we discuss an extremely rare case of ACC of proximal trachea, which was diagnosed in a 60-year-old male who presents with recently diagnosed stridor and cough since 2 months which was ever misdiagnosed as chronic obstructive pulmonary disease (COPD). Fibreoptic bronchoscopy and computerised tomographic (CT) scan of chest revealed an endotracheal mass that was on histopathological examination showed adenoid cystic carcinoma of trachea. As per search in medline very few such cases are reported in india itself. The patient was treated by surgical sleeve resection followed by radiotherapy and is successfully on regular follow-up.

Key words: Pulmonary disease, Adenocystic carcinoma, Trachea.

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INTRODUCTION

Tumors of tracheal origin are exceedingly rare; the incidence of primary tracheal tumors is less than 0, 2 per 100,000 persons per year.[1] Malignant tumors are more common than benign tumors and account for 60 to 83% in adults.[2] Adenoid cystic carcinoma (ACC) is a rare primary tracheal tumor that is second most common tracheal malignancy at histology after squamous cell carcinoma.[3] The clinical and pathologic features of ACC of the trachea; which was formerly named "cylindroma" and "adenoicystic carcinoma", were initially reported in 1859 by Billroth.[4] It is usually found in younger patients and it appears to be unrelated to smoking, with an equal distribution in males and females.

CASE REPORT

60-year-old male who presented with severe strider to our department. He had history of wheeze and cough since 2 months. He had similar complaints four months ago and was treated with antibiotics and bronchodilators by a general practitioner. Since 10 year was taking treatment for COPD.

General examination was unremarkable. On auscultation he had bilateral wheezing. The hematological and biochemical profile was essentially normal. The chest radiograph was normal. (Figure 1) As biased towards COPD patient was treated with bronchodilator and inhaled steroids but symptoms were progressively increased without any improvement.

Further evaluation was done via endoscopy and imaging. Endoscopic evaluation revealed larynx was normal (Figure 2), But there was a polypoidal pinkish growth in upper part of trachea that appear arises from anterior wall and occluding >30% of the lumen (Figure 3).

Computed tomography imaging revealed endotracheal enhancing soft tissue density lobulated lesion arising from anterior wall of the proximal trachea occluding >30% of the lumen (Figure 4) additional patchy consolidation and mild bronchiectatic changes in b/l lung fields Bronchoscopic biopsies of the lesions showed adenoid cystic carcinoma.

Emergency tracheostomy was done and planned for elective tracheal tumour resection. Tracheal sleeve resection was performed, intraoperative tumor was found arises from anterior wall of third tracheal ring excised along with 1 cm margins.

Histopathology report revealed tissue lined by respiratory epithelium with a polypoidal neoplasm seen in sub epithelial region neoplasm is composted of basoloid cells and myoepithelial cells forming cribriform and tubular pattern microscopic pattern in these patterns are filled with hyalinized material tumour invade in between the hyaline cartilage in to the pericartilegenous adipose tissues. Maximum diameter of the tumour is 2 cm. circumferential resected margin is free of tumour with 1 mm clearance. Superior and inferior mucosal resected margins are free from tumour with 1 mm clearance.

Patient underwent radiotherapy and is successfully on regular follow-up. Patient was re-evaluated after 6 month by fiberoptic bronchoscopy which shows patent tracheal lumen with mild tracheal stenosis and no residual lesion with healed mucosa.(Figure 5)

Follow-up at 6 months post-treatment showed no local recurrence or distant metastases.

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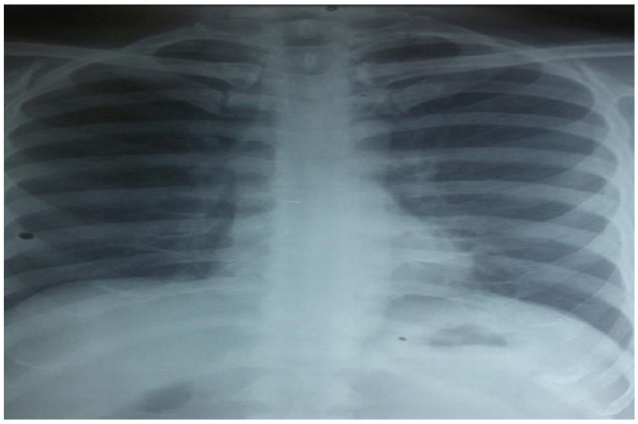

Figure 1: shows chest x-ray with no obvious finding.

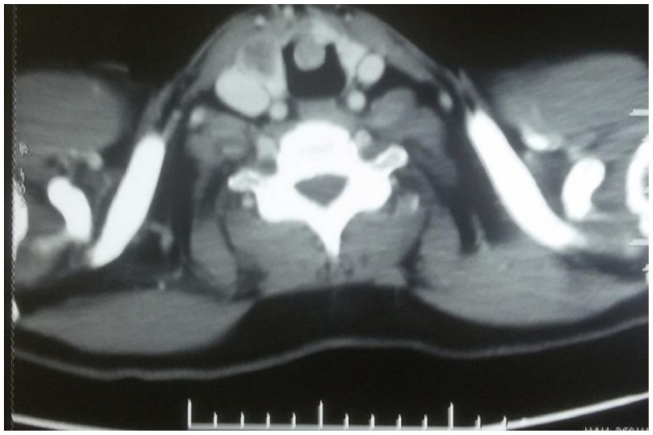

Figure 4: Computed tomography imaging revealed endotracheal enhancing soft tissue density lobulated lesion arising from anterior wall of the proximal trachea.

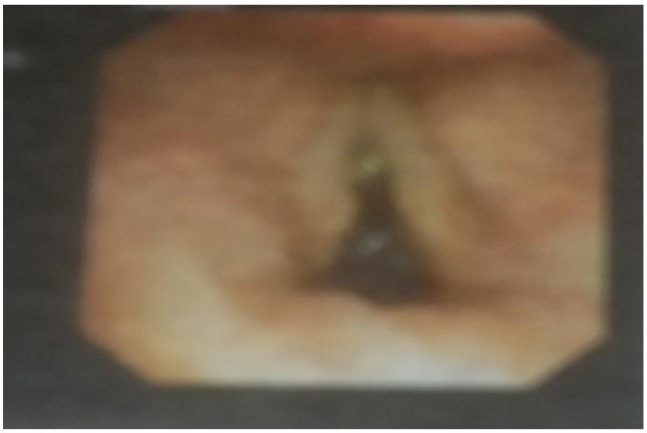

Figure 2: Endoscopic picture showing normal larynx.

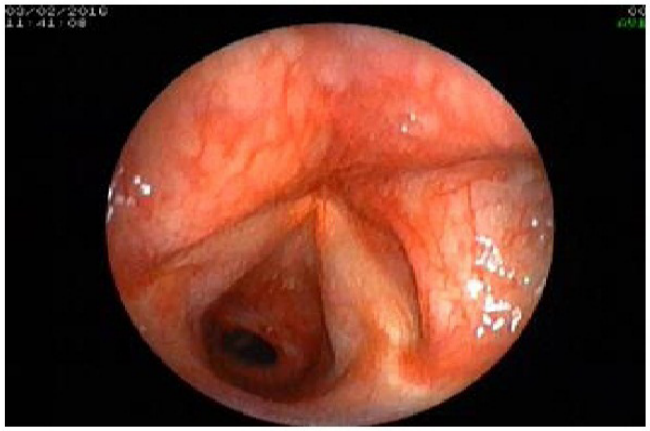

Figure 5: shows patent tracheal lumen with mild tracheal stenosis and no residual lesion with healed mucosa.

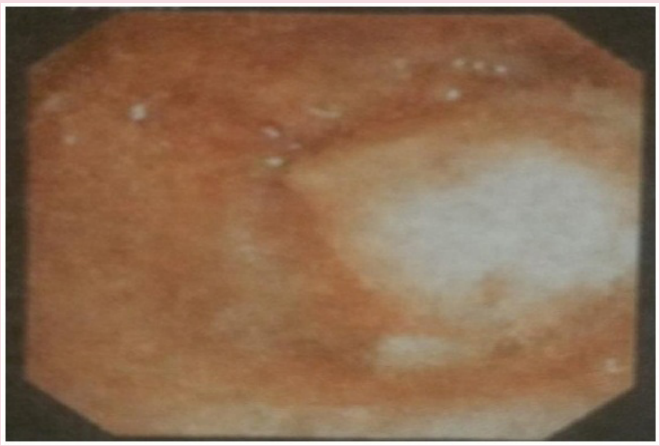

Figure 3: Showing mass occluded the tracheal lumen.

DISCUSSION

Tracheal tumors mostly arise in the lower or upper third, with a tendency to originate at the lateral and posterolateral wall near the junction of the cartilaginous and membranous portions. Adenoid cystic carcinoma (ACC) of the trachea is rare, it represents 1% of all respiratory tract cancers. It's generally considered as a slow-growing, with prolonged clinical course. Most ACCs are discovered in middle age with no gender predilection.[5]

ACC arises from glands of the upper respiratory more commonly in the minor salivary glands and in the seromucinous tract. ACC is a non encapsulated tumor, it spreads most commonly by direct extension, submucosal or perineural invasion, in transverse and longitudinal planes.[6] Lymphatic spread is uncommon. More than 50% of patients with tracheal ACC have hematogenous metastases. Pulmonary metastases are the most common and can remain asymptomatic for many years.[7] Metastases to the brain, bone, liver, kidney, skin, abdomen; and heart have also been reported. The usual clinical presentation is directly related to the size and location of the tumor within trachea. Most patients with ACC trachea are symptomatic at the time of presentation and symptoms

usually relate to airway obstruction i.e. cough, hoarseness, hemoptysis, shortness of breath and wheezing respiration. Patients may be incorrectly diagnosed and treated for asthma or chronic bronchitis for months or years before the lesion is recognized

Here we report one case of primary adenoid cystic carcinoma of the trachea which was ever biased and treated for COPD. The CT scan is a useful imaging procedure for ACC. It is highly accurate in the assessment of the tumor location, extra luminal extensions, cranial involvement and distant metastasis.[7]

Most of the adult primary tracheal tumors arise in the distal third region of the trachea. In our patient, there was involvement of proximal trachea and mainly from anterior wall. ACC of proximal trachea is rare. Treatment of ACC includes surgery, radiotherapy, and combination of both. However, recommended treatment consists of surgical resection with postoperative radiotherapy.[8] Early diagnosis and treatment by surgery with radiation therapy provide significantly prolonged survival or even the possibility of complete remission. Maziak showed a five-year cumulative survival rates for patients with complete and incomplete resection were 82% versus 77%.[9]

Gaissert *et al* has published the largest series of tracheal ACCs, with 135 occurrences in patients during 40 years; 71% of these patients were treated with resection and showed with a 52% 5-year overall survival. Radiotherapy was administered in 70% of the patients postoperatively. In the unresectable ACC patients, radiotherapy alone was recommended, with a overall survival of 30%.[10]

Our patient is doing very well 12 months after surgery and postoperative radiotherapy without any evidence of local recurrence or distant metastases. Long-term survival of patients of ACC depends on the presence of distant metastases. Therefore, regular follow-up at 6-month interval is very important.

CONCLUSION

In conclusion, while evaluating and treating COPD with aggrevating symptoms and not relived by medial management, one must be vigilant for tracheal tumors from extra-thyroidal sites or primary tracheal tumor. ACC should be included in the differential diagnosis of the possibility should be considered that might arise in trachea, as illustrated by our case. Unfortunately, many patients with ACC are mistakenly diagnosed and treated as asthma patients for months or years. Early diagnosis of ACC is important as it may improve surgical resistibility and thereby improve prognosis.

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