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TRACHEAL ADENOID CYSTIC CARCINOMA A case report

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Abstract : Primary tracheal tumors are relatively rare and adenoid cystic carcinoma is one of the rare malignant tumors of the trachea. Here we report a case of tracheal adenoid cystic carcinoma who presented with uncontrolled asthma. On initial evaluation, the flattening of flow -volume loop in spirometry gave the clue for further evaluation and diagnosis. He underwent therapeutic bronchoscopy and removal of the tumor, followed by tracheal stenting and radiotherapy.

Keyword : Tracheal tumors, Adenoid cystic carcinoma, Bronchoscopy.

Introduction:

Primary tracheal tumors are rare and comprises of benign and malignant lesions. Tracheal malignancies occur 0.2 per 100000 population and accounts for 0.1% of the cancer deaths per year (1).

History and examination:

A 23 year old gentleman from North India presented with complaints of cough with productive sputum for the past 1 year , hoarseness of voice for 1 year, progressively worsening, breathing difficulty for 1 year, insidious onset, progressively worsening with effort tolerance of 200 meters, no seasonal, diurnal or postural variation associated with wheeze. He had no past history of tuberculosis or anti tuberculous drug intake. There is history of taking medications for asthma but no improvement. He had no addictions or co-morbid conditions. His vitals were stable. The upper airway examination revealed a shiny polypoidal mass in the right nostril. The chest examination revealed bilateral expiratory monophonic wheeze. The differentials of laryngeal tuberculosis, nasal polyp and bronchial asthma, necrotizing granulomatous vasculitis (formerly known as Wegener's granulomatosis) and tracheal malignancy was considered.

Investigations:

His sputum for acid fast bacilli & TB-PCR was negative. Blood investigations were non contributory. The chest x ray showed normal lung parenchyma with a localized area of tracheal narrowing at the level of sternal angle.

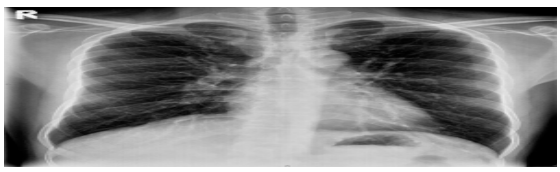


Fig.1 Chest x-ray-PA view- Tracheal narrowing at the level of sternal angle He underwent spirometry which showed severe obstructive ventilatory defect with no reversibility. The flow volume loop revealed flattening of both the inspiratory and expiratory loop suggestive of a fixed intra-thoracic obstruction.

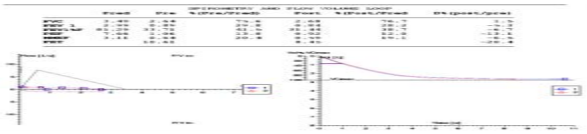


Fig.2 Spirometry & flow volume loop showing flattening of both inspiratory & expiratory loops An Oto-rhino-laryngologist evaluation revealed a granular polypoidal mass seen arising from the right inferior meatus going to the floor of the right nose on rigid nasal endoscopy. The naso-pharyngolaryngoscopy revealed left vocal cord paralysis. He underwent a computed tomography of the neck and thorax which revealed an irregular circumferential wall thickening involving the trachea with the left lateral and anterior wall being more severely involved. The mass is causing up to 75% reduction in tracheal lumen.

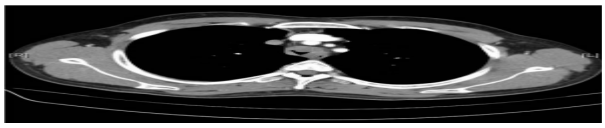


Fig. 3 CT Thorax at the level of thoracic inlet showing luminal narrowing of the trachea

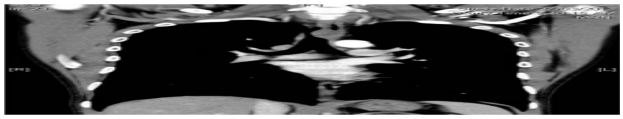


Fig.4 CT Thorax-Mediastinal window sagittal view showing mass in the distal trachea with luminal narrowing.

Bronchoscopy:

In view of significant airway compromise, he was planned for bronchoscopy in operating room under anaesthesia followed by biopsy. The bronchoscopy showed a pedunculated mass in the right nostril and left vocal cord palsy. There was a tumor in the lower trachea, circumferental, 2.5cm from the carina, 3cm long

and 5cm from the vocal cords. Biopsies were taken from the nasal and tracheal mass followed by electrosurgical debulking and balloon dilatation.

FIG 5-

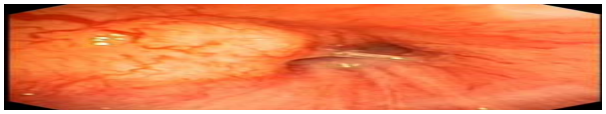
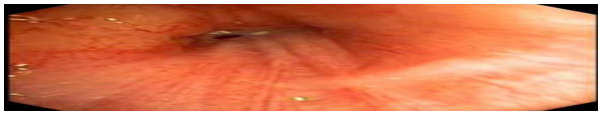


FIG 5- A. Showing tumor in lower trachea



B. Mass with luminal narrowing

FIG 6- Lower trachea after electrosurgical De-bulking and balloon dilatation.

The patient was comfortable and the breathlessness improved. He underwent spirometry which showed improvement in the flow volume loop.

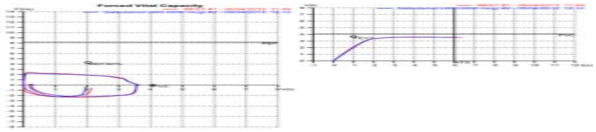


FIG 7: Spirometry & flow volume loop showing improvement of flow in both inspiratory & expiratory
loop after therapeutic bronchoscopy.

The mass from the right nostril was reported as Rhinosporodiosis and the tracheal mass was
reported as adenoid cystic carcinoma.

Discussion:

Primary tracheal tumors are rare and are slow growing which manifest late with cough, breathlessness and haemoptysis (2). Adenoid cystic carcinoma is the second common tracheal malignancy after squamous cell carcinoma. They are also called as adenocystic carcinoma, cribriform carcinoma or cylindroma(3). It is a slow growing low grade tumor

1). When associated with distant metastasis, the survival rate is less than 2 years. These tumors are more common between the ages 40 and 60 years and with no gender predilection. These tumors usually involve the upper one third of trachea with 15% intramural and 85% extramural component. About 20% of these tumors locally invade or metastasize to lymph nodes, nasal cavity, liver and lungs(4). The better survival rate of about 75% is seen with complete resection. The median overall survival rate is 18 years and median disease free survival rate is 10 years. The predictors of survival are margin status, extramural disease, perineural invasion and lymphnode metastasis(4). (The histological patterns are cribiform, tubular and solid. The solid form has a poor outcome compared to cribriform and tubular forms. The primary management of tracheal ACC is surgical resection followed by post operative radiotherapy. In unresectable ACC, a combination of carboplatin/paclitaxel and radiotherapy had shown good results(5). Bronchoscopic techniques have been used for both palliation and temporization until definitive therapy(3). In our patient, who had a locally advanced disease with evidence of perineural invasion, underwent bronchoscopic debulking of the tumor followed by tracheal stenting and radiotherapy. radiotherapy. In unresectable ACC, a combination of carboplatin/paclitaxel and radiotherapy had shown good results(5). Bronchoscopic techniques have been used for both palliation and temporization until definitive therapy(3). In our patient, who had a locally advanced disease with evidence of perineural invasion, underwent bronchoscopic debulking of the tumor followed by tracheal stenting and radiotherapy.

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