CASE REPORT

Pleomorphic Adenoma of the Trachea

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SUMMARY

Primary tracheal tumors are very rare. Pleomorphic adenoma is rarely found in the trachea, despite being the most common histological form of salivary gland neoplasm. We present a case of pleomorphic adenoma of the trachea. Bronchoscopic excision using cold instruments resulted in apparently normal tracheal mucosa.

KEY WORDS:
Pleomorphic adenoma, Tracheal neoplasms

INTRODUCTION

Tracheal neoplasms occur infrequently, accounting for less than 1% of all malignancies. The majority of tracheal tumors in adults are malignant but in children, 90% of the tumors are benign[1]. The most common benign tumor is squamous papilloma, and other types include pleomorphic adenoma, granular cell tumor, and benign cartilaginous tumors.

Pleomorphic adenoma of the trachea is extremely rare. There are only 22 cases reported in World literature[2]. The behavior of this tumor in the trachea appears to be similar to it counter part in other sites and distinctly different from the more frequently encountered epithelial tumors.

CASE REPORT

A 37-year old gentleman presented with a spontaneous hemoptysis of eight months duration. He had two episodes, where he coughed out about 30ml of fresh blood. He did not complaint of dyspnea, chest pain, chronic cough, dysphagia, fever, loss of appetite, loss of weight, epistaxis, vomiting, melena or aspiration symptoms. He was previously a smoker and had discontinued cigarettes three years previously. Clinically, he was well and not in any respiratory distress. He was not cyanosed, nor had he clubbing. There was no bruising, petechiae, or bleeding from any other orifice nor lymphadenopathy. His head and neck examination were unremarkable. His chest examination revealed good air entry bilaterally with no adventitious sounds. Results of cardiovascular examination were unremarkable.

Computed tomography (CT) scan of thorax revealed a small polypoid intraluminal mass with hemogenous density in the posterior wall of mid trachea (Figure. 1). Bronchoscopic examination showed well-defined, glistening mass, arising from the posterior wall of the trachea and 5cm from the vocal cords (Figure 2). Transbronchoscopic biopsy was performed and reported as adenoma.

Intraoperatively, he was placed in supine position. After induction of anaesthesia using muscle relaxant and analgesia, suspension laryngoscopy was placed. Jet ventilation device was positioned proximally using a slim metal cannula in the lumen of the laryngoscope to maintain oxygenation. A well-demarcated, 2x2 cm in cross diameter, peduncluted and flushy tumor was found and excised with cold instruments. Postoperative, he was well and nursed in normal ward. He was treated with corticosteroid for two days. The final

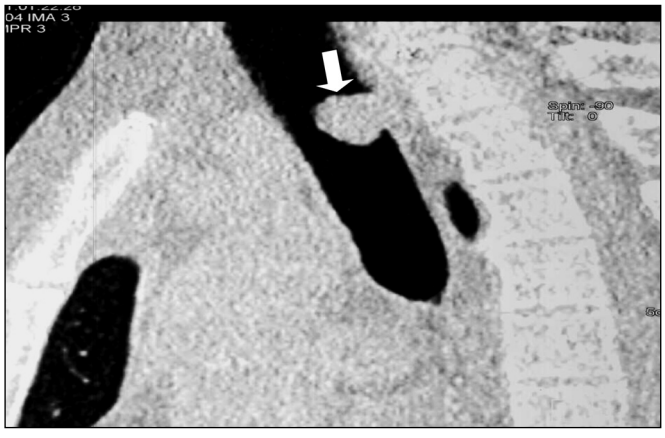

Fig. 1: Pleomorphic adenoma of trachea. Sagittal CT scan shows smooth intratracheal soft tissue mass based on the posterior wall of the trachea.

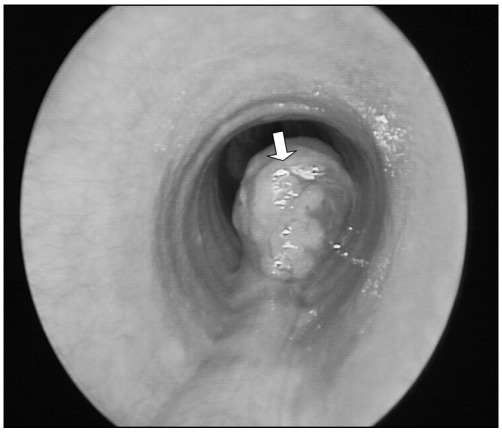

Fig. 2: Bronchoscopy shows an intratracheal polypoid mass with smooth and glistening surface.

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pathological diagnosis was pleomorphic adenoma. He was seen at three months post operation and presently he remains asymptomatic.

DISCUSSION

Primary tracheal tumors represent only 2% of upper airway tumor[3]. Its incidence is 0.1 per 100,000[1]. There is 1:100 ratio of tracheal to bronchial tumors in autopsy material. Their rarity often contributes to significant delays in diagnosis. Benign epithelial neoplasm of the trachea and main bronchi are rare. They consist of surface epithelial neoplasms and tracheobronchial gland neoplasms. Surface epithelial neoplasms consist of squamous cell papillomas, transitional cell papillomas, and papillary adenomas. Benign tracheobronchial gland neoplasms usually form intralumenal polypoid lesions. The varieties that have been described include mucous gland (cell) adenoma, cystadenoma, pleomorphic adenoma, and oxyphil adenoma[3].

Pleomorphic adenoma is the most common form of major salivary gland tumors, but it is exceedingly rare in the trachea. The behavior of these tumors in the trachea appears to parallel those of their counterparts in salivary glands with high propensity for recurrence and on rare occasions metastasize. The most frequent presenting symptom for pleomorphic adenoma is dyspnea, hemoptysis, cough, wheezing and stridor. In many cases the diagnosis is delayed because fifty percent of the tracheal lumen has to be compromised before any localizing signs and symptoms appear. Heifetz *et al*[2] stated that some of the patients with pleomorphic adenoma of the trachea are mistaken for having asthma on the initial evaluations. However, in our patient as the only presenting symptom was hemoptysis, tuberculosis was the initial diagnosis.

Tracheal tumors are notoriously difficult to diagnose on plain chest X-ray, however. CT scanning gives the best estimation of the tracheal involvement. CT characteristics of pleomorphic adenoma include polypoidal intraluminal soft tissue mass with homogeneous density. Mixed tumour of salivary gland type in the trachea can be diagnosed by transbronchoscopic biopsy or aspiration cytology[1]. Microscopically, tumors arise in the sub epithelial tissues and characterized by biphasic composition showing admixture in varying proportions of epithelial and stromal elements. A mixture of respiratory and squamous epithelium covers the tumors. The gland has a tubular pattern, with a varying amount of eosinophilic material. Foci of squamous metaplasia have been identified in the tumor. The stroma usually infiltrated with connective tissue myocin (myxoid), some shows metaplasia to cartilage.

For most primary tracheal tumors, surgery is the modality of choice. The treatment of choice for pleomorphic adenoma is complete excision with cuffs of normal tissue to avoid recurrence, which is attributed to pseudopod extension of the tumor. Various therapeutic modalities have been used such as segmental resection of the trachea and end-to-end anastomosis, bronchoscopic removal with cold instrument, laser excision, cryotherapy, and radiation. Our center does not have any lasers that would be suitable for removal of this tracheal tumor. Instead, the cold instrument via suspension laryngoscopy was used to excise the tracheal tumor with adequate exposure and access.

Conventional anesthetic techniques for tracheal tumors are either rigid bronchoscopy, to bypass the lesion with a small-sized endotracheal tube or to ventilate through a standard tube placed just above the lesion. In our case we used jet ventilation via a laryngoscope with ability to maintain oxygenation and good assess to surgical field.

CONCLUSION

Pleomorphic adenoma of the trachea is an extraordinarily rare tumor. The behavior of these tumors in the trachea appears to be similar to it is counterpart in salivary glands with a tendency for recurrence. These tumors are easily mistaken for common diseases such as bronchial asthma on initial evaluation.

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