

CASE REPORT

Laryngotracheal Pleomorphic Adenoma: A Rare Entity

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ABSTRACT

Laryngotracheal pleomorphic adenoma is a rare tumor with a lot of dilemma in establishing histological diagnosis and with no definitive treatment guidelines. Very few cases have been reported in the literature with good outcomes in open approach procedures. We, in our report, will discuss the detailed surgical procedure and the outcome with open approach.

Keywords: Cricotracheal resection, Larynx, Pleomorphic adenoma, Salivary gland tumor, Trachea.

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INTRODUCTION

Pleomorphic adenoma is the most common benign tumor of major salivary glands. Laryngotracheal pleomorphic adenoma is very rarely described. There is a lot of challenge involved in establishing diagnosis because of the location and airway-related issues. Due to their rare occurrence, management has not been standardized. Definitive procedures are carried out on a case by case basis. This varies considerably and includes interventional endoscopy and extensive resection.¹ Although it is difficult to determine the best treatment approach in individual cases, segmental resection of the trachea, including the tumor and cuff of normal tissue, is thought to be the treatment of choice.² However, the anatomic and functional characteristics of laryngotracheal complex offers special problems during resection and reconstruction when tracheal lesions affect the subglottic larynx or *vice versa*.¹ Stringent respect for the native tissues during resection and reconstruction makes it possible to reduce postoperative complications.³ There are about 20 cases reported in the literature.⁴ Our case is different in the way we have managed by open approach excision and obtained good results postoperatively.

Henceforth, meticulous dissection and good knowledge of the anatomy as well as the disease process is very important for good postoperative results. We have taken consent from the patient to publish this as a case report for academic purpose.

CASE REPORT

A 50-year-old homemaker presented to us with history of hoarseness and occasional dyspnea on exertion since 1 year. She was evaluated for similar complaints in outside hospital 2 years ago. She had undergone endoscopic laryngeal procedure for the same. The histopathology was suggestive of low-grade mucoepidermoid carcinoma. On fiberoptic examination, there was a submucosal, smooth surfaced lesion in the right posterolateral aspect of the subglottis (Fig. 1). The undersurface of the true vocal fold was normal. Bilateral vocal folds were mobile. Rest of the laryngeal examination was unremarkable. The computed tomography (CT) scan revealed that the mass was nonenhancing. It was abutting posterolateral aspect of the cricoid cartilage (Fig. 2). There was minimal erosion of the underlying cartilage. Inferiorly, the mass was extending up to the upper border of 2nd tracheal ring. There was partial airway compromise (Fig. 3). The histopathology of the mass was low-grade mucoepidermoid carcinoma. We planned for partial cricotracheal resection and thyrocricotracheal anastomosis. Patient was induced by orotracheal intubation with a no. 6 size flexometallic endotracheal tube. A horizontal neck crease incision was taken at the level of thyroid cartilage and the subplatysmal flaps elevated. Cricoid cartilage, thyroid



Fig. 1: Laryngoscopic examination showing submucosal mass (arrow) in subglottis

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Fig. 2: Axial view showing mass abutting the right cricoid cartilage compromising the airway



Fig. 3: Coronal view showing mass abutting the right cricoid cartilage compromising the airway

cartilage and the upper six rings of trachea were exposed. The recurrent laryngeal nerve was identified early in the course of dissection and preserved bilaterally. The cervical trachea was separated from the esophagus. After reflection of the thyroid gland, horizontal tracheal incision was made at the level of upper border of third tracheal ring. A fresh endotracheal tube was inserted through the inferior tracheostomy. The tumor was visualized in the subglottis with extension up to level of the 1st tracheal ring (Fig. 4). The proximal cut was taken at the level of the lower border of the thyroid cartilage through the cricothyroid membrane and muscle. Retaining the posterior lamina of the cricoid cartilage along with the muscular attachments and the bilateral cricoarytenoid joint, the cricoid cartilage was cut using heavy scissors. The remnant mucosa was normal and grossly free of tumor tissue. The tumor was excised into along with overlying cartilage (Fig. 5). The trachea was pulled upward to be anastomosed with the remnant cricoid cartilage and lower border of the thyroid cartilage. The airtight closure was confirmed using bubbling test.

Suction drains were placed in the neck and the wound was closed in layers. She had uneventful recovery. The histopathology was pleomorphic adenoma. Postoperative flexible laryngoscopy showed normal bilateral mobile vocal folds with adequate airway. She was followed up for 1 year and there was no evidence of recurrence. She has no airway-related issues and swallowing issues. She is happily back into her profession.

DISCUSSION

The term pleomorphic adenoma implies derivation from a multipotential stem cell, whereas the more commonly used term, benign mixed tumor implies the simultaneous transformation of both epithelial and myoepithelial precursors.⁴ Tracheobronchial pleomorphic adenoma is a benign tumor arising from the mucous glands of the tracheobronchial tree.⁵

Males predominate slightly over females. Age ranges from as young as 15 years to as old as 82 years. There was no specific patient age associated with appearance

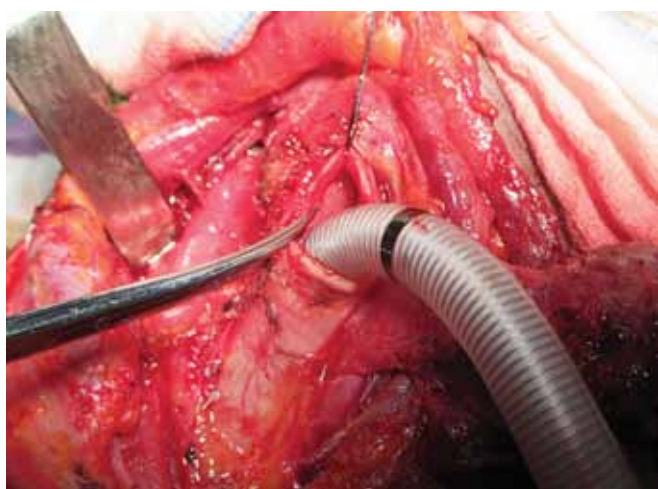


Fig. 4: Submucosal subglottic mass seen after tracheotomy being pointed using instrument

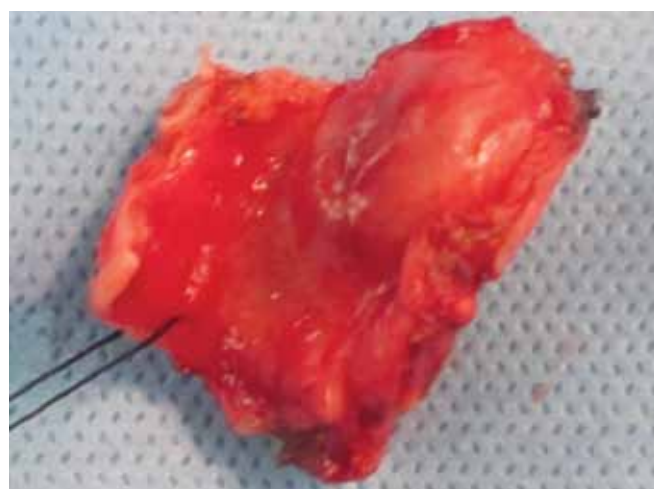


Fig. 5: Specimen containing part of cricotrachea and the submucosal mass

of these tumors, though most of the cases were noted in the 7th decade of life.⁶

The more common symptoms related to phonatory and respiratory complaints *viz.* hoarseness and dyspnea, followed by dysphagia. The type and severity of symptoms were dependent upon the size and site of the tumor mass *viz.* subglottic tumors were usually associated with dyspnea.⁶

Histologically, there is morphologic diversity including mucoid, chondroid, osseous and myxoid elements.⁷

Immunohistochemical stains prove to be positive for various cytokeratins, S100 protein, vimentin and smooth muscle actin. This describes the 'mixed' nature of the tumor. Pleomorphic adenoma may resemble aggressive epithelial tumors because of the high cellularity and lack of a stromal component which can lead to misdiagnosis as a carcinoma.⁸

Radiologic imaging plays an important role in the diagnosis of tracheal tumors and assists in treatment planning.⁹

Endoscopic treatment of tracheal pleomorphic adenoma has been described and may include tumor debulking, enucleation, laser ablation or cryotherapy. The risk of local recurrence is substantial with endoscopic techniques and multiple treatments are usually required.⁹

The treatment for pleomorphic adenoma of the trachea consists of complete surgical resection with end-to-end tracheal anastomosis.¹⁰ But, when the lesion in the subglottis or extending into the upper trachea, it is a surgical challenge. Cricoid cartilage is a part of the basic functional unit of larynx; hence, any alteration in it alters the functional outcome. Larynx-sparing resection aims at prolonged preservation of vocal cord function, reserving complete laryngectomy for a distant future. Whether laryngeal function should be sacrificed is a matter of judgment and tumor type in each individual patient.¹

The recurrence rate of an ordinary pleomorphic adenoma depends almost entirely on the adequacy of the primary excision and most cases of recurrence take place within 18 months after surgery.¹¹ In rare cases, pleomorphic adenoma has been reported to have metastasized to the lymph nodes or other organs.¹¹

In our patient, we planned for an open approach in view of lack of complete visualization of the tumor by endoscopic approach and also because it was referred to us with a histopathology of low-grade mucoepidermoid

carcinoma. Surgery is the treatment of choice for such lesions. Our case shows that even with open approach with conservative resection, the morbidity is minimal and recovery is equivalent to endoscopic resection. Even in this era, good quality open approach resection yields outcomes similar to endoscopic procedures.

CONCLUSION

The goal of the debriefing is to learn lessons and make improvements that can help us to avoid the misdiagnosis and unnecessary treatment in future. Laryngotracheal resection and immediate reconstruction for subglottic tumors is achieved with good preservation of voice, low morbidity and no compromise of long-term survival.

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