Transtracheal endoluminal resection of a pleomorphic adenoma occluding subglottis

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Word count of the abstract: 99

Word count of the text: 698

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Abstract

A 71-year-old male was treated for suspected bronchial asthma because of dyspnea and stridor for 3 months before presenting at our hospital. Chest computed tomogram and a laryngotracheoscopy revealed a mass occupying the subglottic cavity. Instead of a laryngotracheal resection, the tumor was extirpated from the posterior wall of the subglottis and the first 2 tracheal rings successfully through a vertical tracheotomy just above the life-saving trachestomy tube, and was diagnosed as pleomorphic adenoma. The patient is alive and well with no recurrent tumor 12 years after surgery, without any effect on the function of the voice or swallowing.

Keywords: Tracheal surgery, Tracheal tumor, Tracheostomy, Pleomorphic adenoma

1. Introduction

Primary tumor of the trachea is rare, and diagnosis is delayed in many cases because a large part of the trachea lumen has to be compromised before any localizing signs or symptoms appear. Also, the symptoms can mimic those of more common conditions such as asthma or bronchitis. A wide variety of tumors with histology other than squamous or adenoid cystic carcinoma, most often benign or low-grade malignancies, comprise one-quarter of the cases of primary tracheal tumor [1]. Due to their rare occurrence, management has not been standardized, and is performed on a case-by-case basis. This varies considerably and includes interventional endoscopy and extensive resection.

This case report presents a less invasive surgical procedure that can be used for benign tracheal tumors obstructing airway.

2. Case report

A 71-year-old male was referred to our department because of dyspnea and stridor over the previous 3 months. He had previously been treated for suspected bronchial asthma. A chest computed tomogram (CT) just before referral revealed a mass occupying subglottic cavity. At presentation he had biphasic stridor with suprasternal retraction. Emergent tracheostomy was performed on admission day 2 because of life-threatening asphyxia. Chest CT revealed a sharply marginated soft-tissue mass 2 cm in diameter occluding the lumen of the cervical trachea, which ruled out transmural invasion into the surrounding tissue such as the esophagus (Figure 1A). A laryngotracheoscopy revealed a rounded subglottic lesion that occupied 90% of the air space, originating from the posterior wall, and was covered with inflamed mucosa

(Figure 1B). Endoscopic biopsy was repeated from the submucosa after inflammation withdrawal, but did not yield a final histologic diagnosis including the grade of malignancy.

A cervical collar incision was made in the neck extension position around the tracheostomy. The mucosa of the posterior wall of the subglottis and the first 2 tracheal rings were incised and the well-demarcated tumor was extirpated from the tracheal wall through the vertical tracheostomy (Figure 2A). Incised mucosa and vertical trachestomy were closed using 4-0 absorbable monofilament sutures in an interrupted fashion. The resected spcimen was diagnosed as a benign pleomorphic adenoma (Figure 2B).

The tracheostomy tube was removed 20 days after surgery, and the patient recovered uneventfully without hoarseness or dysphagia. Bronchoscopy performed 6 and 18 months after surgery, chest CT taken every 6 months up to 3 years after surgery, and plain radiographs of frontal and lateral views of the neck every 6 months thereafter revealed no recurrent tumor, and the patient is alive and well 12 years after surgery.

3. Discussion

Tracheobronchial pleomorphic adenoma is a benign tumor arising from the mucous glands of the tracheobronchial tree [2]. It is rare in any one institution, although there have been multiple case reports [3-5]. In the middle or lower third of the trachea, segmental tracheal resection including the tumor and cuffs of normal tissue is considered to be the treatment of choice because of pseudopod extension of the tumor [6] or potential malignancy [7]. However, the anatomic and functional characteristics of that structure offer special problems when tracheal lesions affect the subglottic larynx.

In this case, a sleeve resection or a Grillo cricotracheal resection [8, 9] was not

adopted because of the well-circumscribed nature of the tumor, although patients and surgeons should be advised of a potential need for later laryngectomy. Larynx-sparing resection aims at prolonged palliation of vocal cord function, reserving complete laryngectomy for a distant future. In the present case, we were not sure of the negative surgical margin of the tumor cell on an intraoperative frozen section. Whether laryngeal function should be sacrificed is a matter of judgment and tumor type in each individual patient. In our case, however, the tumor did not recur though the patient has been followed up for more than ten years.

The endoluminal approach in this case was not technically complicated despite limited accessibility due to complex ventilation management. Despite the low incidence of subglottic benign tumors, this approach is preferable to laryngotracheal resection or laryngectomy in selected patients.

In conclusion, a transcervical endoluminal approach through a vertical tracheostomy is another good method for resection of benign tumors of the cervical trachea with good preservation of voice and relief of airway obstruction.

Figure legends

Figure 1. A chest CT scan (A) and a laryngotracheoscopic view (B) showing a mass (arrow) occluding the subglottic cavity.

Figure 2. (A) Intraoperative schema showing endoluminal resection through a vertical trachestomy. 1; Thyroid cartilage, 2; Cricoid cartilage, 3; Thyroid gland divided at the isthmus, T; trachestomy tube, the dashed line indicating mucosal incision of the membranous portion of the cervical trachea. (B) A microphotograph of the resected specimen showing small nests of tumor cells (arrow) with glandular components of various sizes and myxoid or hyalinous stroma (arrow head) (Hematoxylin-eosin, x100).

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