

# Trakeanın Epiteliyal Miyoepteliyal Karsinomu

## EPITHELIAL MYOEPITHELIAL CARCINOMA OF THE TRACHEA

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### Özet

Trakeanın primer tümörleri çok nadirdir. Sunumuz trakeal segmenter rezeksiyon ve uç uca anastomoz uyguladığımız trakeanın epiteliyal miyoepteliyal karsinomu olgusudur. Tümörün histolojik incelemesi WHO'nun tükürük bezleri ile ilgili kriterlerine uymaktadır. Hasta operasyondan bir yıl sonra asemptomatik olarak takip edilmektedir.

**Anahtar kelimeler:** Trakea, epiteliyal, miyoepteliyal karsinoma

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### Summary

Primary tracheal tumors are very rare. We presented here a case of epithelial-myoepithelial carcinoma of the trachea who was treated with segmental tracheal resection and end-to-end anastomosis. Histologically, the tumor had the criteria of WHO international classification of salivary glands. The patient did well one year after the operation.

**Keywords:** Trachea, epithelial, myoepithelial carcinoma

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### Introduction

Epithelial-myoepithelial carcinoma is a rare low-grade malignant salivary gland neoplasm that most commonly occurs in the parotid gland but can also arise in minor salivary glands. About 120 cases have been reported in the world literature, most of which were located in salivary glands, except for a few cases occurring in unusual locations such as breast, lachrymal gland, nose, paranasal sinus, trachea, bronchus, and lung [1-6]. We present here a patient with primary epithelial-myoepithelial carcinoma of the trachea, presenting itself as an asthma-like symptoms.

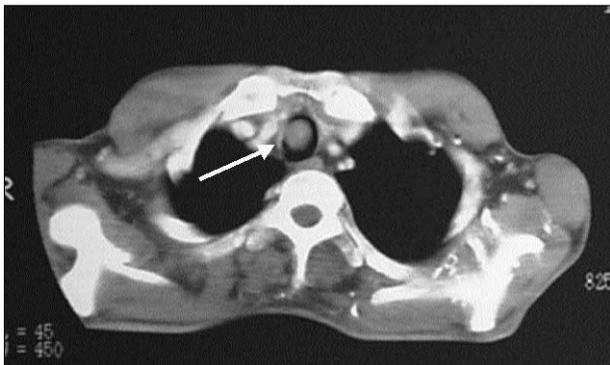
### Case Report

A 32-year-old male was admitted to our hospital with progressive exertional dyspnea and wheezing unrelieved by medical management for presumed asthma in January 2000. The patient did not have a history of asthma in childhood but he had been treated for asthma-like symptoms during the last three years. Physical examination was remarkable for severe stridor, wheezing and tachypnea. There were no palpable cervical lymph nodes or mass. A computed tomographic scan of the neck and chest showed an endotracheal mass of 20 ± 2 mm in diameter originating from right lateral wall, occluding 70–80% of the lumen (Figure 1). Examination with the rigid bronchoscope revealed a polypoid mass 3-4 cm after the cord vocales in the trachea, and it occluded the three quarters of the tracheal lumen. After the lumen was widened by bronchoscopic resection, the bronchoscope could be advanced beyond the

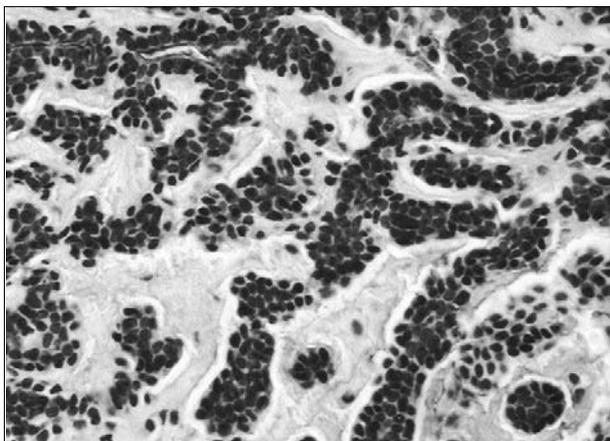
tumor and an adequate airway was maintained. Then an endotracheal tube was inserted into the trachea and ventilation was provided. A 3 cm length of tracheal segment including the tumor was resected through cervical incision. Primary tracheal anastomosis was performed by 3/0 vicryl using interrupted sutures. The patient was discharged uneventfully on the 9th postoperative day. He was symptom free one year after the operation and control bronchoscopy showed no residual stenosis or tumor recurrence.

Upon pathologic examination, macroscopically the specimen was 3 cm long in diameter and consisted of six tracheal rings. The cut surface of the tumor was mucoid and smooth. There was an irregular surface, polypoid tumor projecting into the lumen measured as 2.5x2.1x1.5 cm. Microscopically the tumor was composed of uniform epithelial cells, which were consisted of cuboidal cells with eosinophilic cytoplasm on the luminal surface and myoepithelial cells with clear cytoplasm on the outer layer. The tumor cells were uniform and mitosis was rare. Numerous tubules, whose secretion was positive for PAS, formed by an inner epithelial and outer myoepithelial layer were found. Immunohistochemical staining was performed using an avidin-biotin-peroxidase complex method. Immunohistochemical analysis showed that epithelial cells, which enclose the lumen, were positive for epithelial membrane antigen (EMA) and myoepithelial cells with the clear cytoplasm were positive for both S-100 protein and smooth muscle actin (SMA) (Figure 2-3). Local invasiveness or regional lymph node metastasis was not determined. Tumor wasn't seen on the resection line.

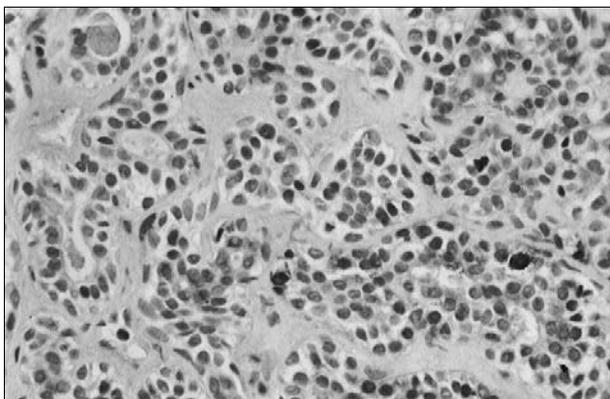
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**Figure 1.** A computed tomographic scan of the neck and chest demonstrating the endotracheal mass.



**Figure 2.** Immunohistochemical positive for EME (EMAx20).



**Figure 3.** Immunohistochemical positive for S-100 protein in the outer layer (S-100x40).

## Discussion

Primary tracheal tumors are rare neoplasms, most of them being epithelial in origin. Among these primary epithelial-myoeplithelial carcinomas of trachea are extremely rare. To date, only one patient with primary epithelial-myoeplithelial carcinoma of the trachea has been reported in the English literature [4]. The patient was producing bloody sputum.

Whereas our patient presented with long term and progressive dyspnea without hemoptysis. Besides causing delay in diagnosis, this clinical picture supports that epithelial-myoeplithelial carcinoma of the trachea is a tumor showing slow progression. Therefore tracheal tumors may be considered in the differential diagnosis of patients with intractable asthma-like symptoms. Epithelial-myoeplithelial carcinoma was described in the 1991 WHO international classification of salivary tumors [2]. According to this, epithelial-myoeplithelial carcinoma is composed of epithelial cells, which are positive for EMA by immunohistochemical analysis, are composed of cuboidal cells on the inner surface, and myoeplithelial cells with clear cytoplasm on the outer layer, are positive for both S-100 protein and SMA by immunohistochemical analysis [2,3]. This carcinoma occurs more frequently in salivary glands of head and neck. When it was taken in to consideration submucosal tracheo-bronchial glands are part of the minor salivary glands, epithelial-myoeplithelial carcinoma may originate from tracheo-bronchial glands [3]. Because there was no other tumor present in the salivary glands or other organs, and it was the single mass in the trachea, the tumor was accepted as primary. From pathological perspective, differential diagnosis of epithelial-myoeplithelial carcinoma includes a pleomorphic adenoma, adenoid cystic carcinoma, myoeplithelioma and the other primary salivary gland-type tumors. Furthermore, primary and metastases of clear cell carcinoma must be distinguished from epithelial-myoeplithelial carcinoma. In our case, the tumor has neither recurred nor metastasized, which is a evidence supporting the current opinion that epithelial-myoeplithelial carcinoma is a tumor of low-grade malignancy [3]. Although epithelial-myoeplithelial carcinoma is a low grade malignity , complete resection is necessary for a successful treatment in the trachea. Complete removal depends on the size of the tumor. Small size tumors like in our case may allow sleeve resection and primary anastomosis. When it was performed as an appropriate manner, short and long term result of surgery have been satisfactory. Finally clinical picture of our patient suggests that epithelial-myoeplithelial carcinoma of the trachea should be kept in mind as a rare cause of progressive and intractable asthma-like symptoms to prevent delay in diagnosis and treatment.

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