

## Seven-Year Survival of a Patient with Primary Tracheal Squamous Cell Carcinoma after Surgery: A Case Report

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### ABSTRACT

The primary tracheal cancers are very rare, and squamous cell carcinoma is its most common form, especially in smokers. Due to the late presentation of symptoms, these tumors are usually diagnosed too late, and thus, they have poor survival. The gold standard protocol for these cancers is surgical excision and adjuvant radiotherapy. Primary radiotherapy is applied for the advanced and inoperable patients.

In this study, we present the case of a 78-year-old woman with a history of heavy smoking, coughing, and dyspnea for a long time. During the diagnostic evaluations, bronchoscopy was carried out and a vegetated tumor was observed about 5 cm below the vocal cords. The patient was referred to the Department of Cardiothoracic Surgery, Ghaem Hospital of Mashhad University of Medical Sciences, Iran, for surgical management. The tumor was removed with rigid bronchoscopy, and her dyspnea temporarily improved. The pathology report indicated that the patient was suffering from squamous carcinoma. Therefore, she required reoperation to excise the invaded trachea with a tumor-free margin. Ultimately, considering no marginal involvement, lymphatic metastasis, or distant metastasis, the patient was discharged and referred to receive additional oncological treatments with the recommendation of annual surveillance bronchoscopy. Seven years after the operation, the patient is still alive and healthy without any local recurrence or metastasis at the age of 82.

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### Introduction

Given that the primary tracheal cancers are very rare, studying the natural history of the tumor and its treatment policy is too challenging and the knowledge about them is limited. The primary tracheal tumors present with different clinical and histological manifestations (1).

Most of the primary tracheal tumors are malignant and include adenoid cystic carcinoma (ACC), squamous cell carcinoma (SCC), adenocarcinoma, mucoepidermoid carcinoma, carcinoid tumor, and small cell carcinoma (oat

cell carcinoma) (2, 3). In some reports, SCC was more prevalent in smokers and ACC in nonsmokers (2).

Jimmie Honings and colleagues in Massachusetts General Hospital (MGH) reported 59 cases of tracheal SCC and stated that 50% of the primary tracheal tumors were of SCC type (4). The thyroidal and esophageal tumors may invade the trachea; at the same time, it is a site of metastasis of recurrent carcinoid of the left main bronchus, larynx, and lungs (1).

The radiological appearance of these tumors

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**Figure 1.** The anterior-posterior chest X-ray in which the deviation of the narrow column of air inside the trachea and its constriction is marked with arrows

falls into three categories of exophytic, wall-thickening, and intraluminal. In addition, the endoscopic appearance of these tumors is usually an obstructive prominent lesion in the trachea, just like in our patient (5).

Generally, due to nonspecific symptoms like hemoptysis, cough, dyspnea, hoarseness, and stridor, these tumors are diagnosed too late and have a poor survival rate. If there are no serious symptoms such as hemoptysis, patients are not usually referred to specialists and are treated as asthma and airway hypersensitivity patients for years. High-resolution computed tomography (CT), a very useful imaging technique for diagnosing tumors, coupled with endoscopy, a complementary method in tumor diagnosis, can be useful in the reconstruction of the trachea (5). Thus, the patient was admitted to the hospital and X-ray (Figure 1) and cervicothoracic CT scan (Figure 2) were performed.

### Case Presentation

A 78-year old woman with a long history of smoking (up to 15 years), cough, and dyspnea (for months), which aggravated recently, was referred to the Department of Cardiothoracic Surgery, Ghaem Hospital of Mashhad University of Medical Sciences, Iran. After admission, and obtaining X-ray (Figure 1) and neck and chest CT scan images (Figure 2), she went under diagnostic fiber optic bronchoscopy. Despite normal vocal cords, there was a 2-cm tumoral protrusion from the posterior and lateral tracheal walls at approximately 5 cm below the vocal cords. The tracheal tissue below the tumor was



**Figure 2.** The coronal section of the chest and neck computed tomography that shows the narrowing of the tracheal space

uninvolved. Biopsies were taken and the pathology report indicated papilloma with focal high-grade dysplasia.

The patient was admitted to the Department of Surgery in December 2009. After a rigid bronchoscopy and removal of the tumor, the lumen became patent. The pathologists reported that the tumor was of the squamous cell carcinoma type, and accordingly, the patient was re-admitted 10 days later and prepared for surgical excision. Three centimeters of the trachea was resected through a cervical incision, and then an end-to-end anastomosis was performed. The final histological diagnosis was squamous cell carcinoma without any marginal involvement. The patient was discharged without any dyspnea and in a good general condition. She was also referred to the Department of Oncology for adjuvant radiotherapy. Annual follow up was performed using neck and chest radiography and bronchoscopy. Four years after the surgery at the age of 82, the patient is still living healthy without any local recurrence or distant metastasis.

### Discussion

The knowledge regarding primary tracheal cancer is limited due to its rarity. The primary tracheal tumors are usually malignant and mostly of the squamous cell carcinoma type. They are more prevalent in smokers like in our case, a 78-year-old heavy smoker, while primary tracheal ACC is more prevalent in non-smokers. Some other rare tumors such as carcinoid tumors, small cell carcinoma (oatcell carcinoma), benign

tumors, and tracheal involvement from thyroidal, esophageal, or laryngeal cancers have been reported (1). Webb and colleagues from the University of Texas studied 74 patients with tracheal tumor during 60 years (1945–2005). Thirty-four cases were detected with SCC (45.9%). The most prevalent symptoms were dyspnea (55.4%), hemoptysis (48.6%), cough (41.9%), and hoarseness (35.1%). Most of the patients were farmers and had a history of smoking. Among these cases, those who received primary surgical treatment and adjuvant radiotherapy, had better survival rate than those who underwent primary radiotherapy and/or chemotherapy (2).

Thotathil and colleagues studied 15 patients with primary tracheal malignancy from 1983 to 2000. Most of their cases were aged over 40 years and the majority of them were male (87%). Most of the tumors were of the SCC (40%) and ACC (27%) types. One of the patients was treated using primary surgical resection, anastomosis, and adjuvant radiotherapy. Others underwent different protocols of chemotherapy or radiotherapy. Moreover, two of the patients received intraluminal brachytherapy. The researchers concluded that radiotherapy is significantly effective for unresectable tumors (6).

In another study, Gaissert and colleagues examined 270 cases of tracheal tumor from 1962 to 2002. In general, 135 patients had SCC, and the remaining had ACC. Furthermore, 191 (71%) patients underwent surgical resection and 79 (67%) did not receive the operation on account of tumor length (67%), local invasion (12.4%), distant metastasis (7%), and other reasons (2%). Five- and ten-year survival rates in patients who had tracheal SCC and were treated through tumor resection were 39% and 18%, respectively (7).

Grillo and colleagues investigated 135 cases of tracheal SCC in a period of 26 years. They reported that the most prevalent site of metastasis was the paratracheal lymph nodes. The hematogenous metastasis to the lungs, liver, bones, and sometimes adrenals may also occur. Gender, age, and prevalence of tracheal SCC are similar to those of lung SCC that mostly present in the sixth and seventh decades of life and more frequently in men (8).

Besides the plain radiographs that may show the tumor in the trachea (Figure 1), CT scan and spiral CT reconstruction of the trachea clearly exhibit the location and spread of the tumor in the trachea (Figure 2). Ultimately, fiberoptic bronchoscopy and biopsy (if possible) would confirm the diagnosis. If the biopsy was not possible (because of probable bleeding), rigid bronchoscopy and sampling under general anesthesia would show tumor location and length

and confirm the diagnosis (similar to our case).

Surgical decision-making is performed after endoscopic evaluations. The proper surgery is tumor resection followed by an end-to-end anastomosis, while adjuvant radiotherapy is effective in cases with regional lymph nodes or microscopic marginal involvement, which provides them with a better survival rate (8).

Currently, the tracheal resection and reconstruction method is the procedure of choice in more than 50% of tracheal SCC tumor patients. The need for complementary radiotherapy after the surgery is based on the invasion to the regional lymph nodes or microscopic marginal involvement of the anastomosis (9).

Ahn and colleagues studied 37 patients with tracheal tumor, of whom 14 and 23 cases had benign and malignant tumors, respectively. Among the malignant tumor patients, 11 cases had tracheal SCC. In that study, six patients were treated with surgery, three patients with surgery and radiotherapy, and two with radiotherapy alone. The researchers demonstrated the outcomes of the different treatment methods and ascribed that surgical resection was the best treatment for tracheal SCC (10).

The most commonly accepted staging classification of the primary tracheal tumors that is based on Bhattacharyya staging system is presented below:

T1: tumors less than 2 cm and confined to the trachea

T2: tumors more than 2 cm and confined to the trachea

T3: tumors spread outside the trachea but not to the adjacent structures

T4: spread to the adjacent structures

It should be noted that TNM staging is related to the presence of lymphatic or distant metastasis (2).

Based on the staging, our patient was staged as T2N0M0 and underwent surgery. Three centimeters of the trachea was resected followed by an end-to-end anastomosis, but due to the negative surgical margins, radiotherapy was not performed. Until now, annual follow-ups by using radiography and bronchoscopy have been conducted for seven years, and the patient is living healthy without any local recurrence or distant metastasis.

## Conclusion

Primary tracheal tumor is very rare and primary squamous cell carcinoma is one of its histological forms occurring mostly in smokers. Generally, because of the common and nonspecific symptoms, these tumors are diagnosed too late and have a poor survival rate. Presenting this patient in our study, the researchers aim to draw attention to this

uncommon malignancy and recommend considering it as a probable diagnosis when evaluating a patient with treatment-resistant respiratory symptoms. This case has reached a good state five years after the surgery and is living healthy without any local recurrences.

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None.

### Conflict of Interest

The authors declare no conflict of interest.

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