Journal of
Cardio - Thoracic Medicine

http://jctm.mums.ac.ir

The Five Years Survival after Surgery of Primary Tracheal Squamous Cell Carcinoma: A Case Report and Review of the Literature

Seyed Hossein Fattahi Masoom[1], Seyed Hossein Ahmadi Hoseini[2], Amir Mohammad Hashem Asnaashari[2]*, Yavar Shams Hojjati[3], Aida Shams Hojjati[4]

[1] *Thoracic Surgeon, Endoscopy Minimally Invasive Surgery Research Center, Mashhad University of Medical Sciences, Mashhad, Iran*
[2] *Pulmonologist, Lung Diseases Research Center, Ghaem Hospital, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran*
[3] *Fellowship Plastic Surgeon, Lung Diseases Research Center, Ghaem Hospital, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran*
[4] *Emergency Medicine, Mashhad University of Medical Sciences, Mashhad, Iran*

ARTICLEINFO

Article type:
Case report

Article history:
Received: 04 Nov 2015
Revised: 24 Dec 2016
Accepted: 24 Apr 2017

Keywords:
Bronchoscopy
Squamous Cell Carcinoma
Trachea

ABSTRACT

Unlike the primary tracheal tumors, squamous cell carcinoma of trachea is common, especially in smokers. This type of tumor has a low rate of survival and it is diagnosed too late on account of late presentation of its signs. The treatment of choice is surgical removal followed by adjuvant radiotherapy; Primary radiotherapy is the appropriate treatment in inoperable cases. In this study, we present the case with a long history of smoking, who was suffering from cough and dyspnea for a long time. During diagnostic evaluations a vegetated tumor was observed about 5 centimeters below the vocal cords. Pathologists reported the tumor as a squamous cell carcinoma, and the patient underwent an operation for resection of involved trachea.

Through presenting this patient, we aimed to draw attention to this uncommon malignancy and recommend considering it as a probable diagnosis when evaluating a patient with treatment-resistant respiratory symptoms.

►*Please cite this paper as:*
Fattahi Masuom SH, Ahmadi Hoseini SH, Hashem Asnaashari AM, Shams Hojjati Y, Shams Hojjati. The Five Years Survival after Surgery of Primary Tracheal Squamous Cell Carcinoma: A Case Report and Review of the Literature. J Cardiothorac Med. 2017; 5(2): 587-590.

Introduction

Primary tracheal tumor has a rare incidence and studying the natural history of the tumor and its treatment policy is too difficult given the scarcity of data on this issue. Primary tracheal tumors have different clinical and histological manifestations (1).

Most of the primary tracheal tumors are malignant and include adenoid cystic carcinoma (ACC), squamous cell carcinoma (SCC), adeno-carcinoma, mucoepidermoid carcinoma, carcinoid tumor and small cell carcinoma (oat cell carcinoma) (2, 3). SCC has been reported to be more prevalent in smokers, while ACC is more common in nonsmokers (2).

Honings and colleagues in Massachusetts General Hospital reported 59 cases of tracheal SCC and stated that about 50% of primary tracheal tumors were SCC (4). Thyroidal and esophageal tumors may invade trachea (trachea is a site of metastasis from recurrent carcinoid of left main bronchus, larynx and lung) (1).

Radiological appearance of this tumor is divided into three categories: exophytic, wall thickening and intraluminal. Endoscopic appearance of this tumor is usually as a bulky and obstructive lesion in the trachea, similar to our

**Corresponding author:* Amir Mohammad Hashem Asnaashari, Lung Diseases Research Center, Ghaem Hospital, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran. Tel: +9805138436199; Email: emis@mums.ac.ir

case (5). Due to common and nonspecific symptoms like hemoptysis, coughing, dyspnea, hoarseness and stridor, these tumors are diagnosed too late and have a low rate of survival. Moreover, in case no blood in the sputum is observed, the patient is usually diagnosed as asthmatic. Computerized tomography (CT) reconstruction of trachea is a very useful imaging technique for diagnosing these tumors and endoscopy is a complementary method (5).

Case Presentation

In this study, we presented the case of a 78-year-old female with a long history of smoking (up to 15 years ago), suffering from cough and dyspnea for months which aggravated recently. The patient was referred to the thoracic surgery ward of Mashhad University of Medical Sciences, Iran, 2009. After admission and taking X-ray (Figure 1), as well as neck and chest CT scan, (Figures 2, 3) the patient underwent diagnostic fibrotic bronchoscopy due to tracheal stenosis caused by a tumoral lesion. Vocal cords were normal. From approximately 5 centimeters below the vocal cords a 2-centimeter tumoral protrusion from posterior and lateral tracheal walls was observed. The trachea below the tumor was tumor-free and had no other lesions. Biopsies were taken from the tumor, showing papilloma with focal high grade dysplasia.

After rigid bronchoscopy and removal of the tumor the lumen became patent. Pathologists reported the tumor as squamous cell carcinoma, therefore, the patient was re-admitted 10 days later for surgery.

By a cervical incision, about 3 centimeters of trachea was resected followed by an end-to-end anastomosis. The final histological diagnosis was squamous cell carcinoma without any marginal involvements. The patient was discharged without any dyspnea and in a good general condition. Annual follow-up by neck and chest radiography and bronchoscopy was performed.

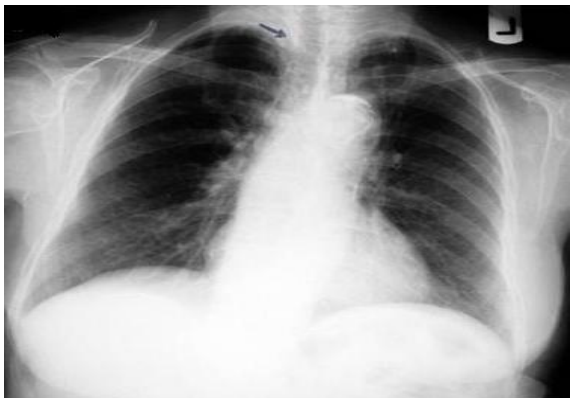


Figure 1. Chest X-ray of our patients Arrow: tracheal stenosis due to the tumor

Five years after the surgery, the patient was healthy without any local recurrence or distant metastasis at the age of 82.

Discussion

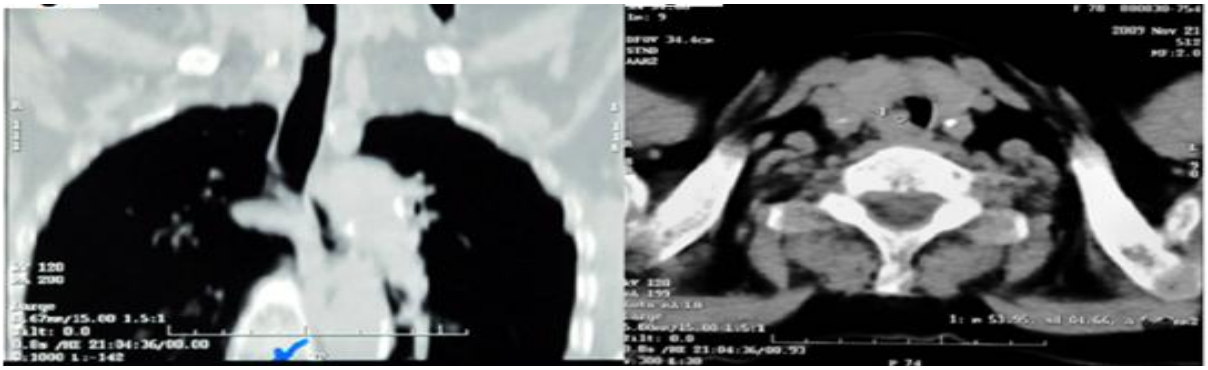
Primary tracheal cancer has a low rate of incidence; therefore limited data on this issue is at hand. Primary tracheal tumors are mostly malignant. Primary tracheal ACC is mostly prevalent in nonsmokers and primary tracheal SCC, on other hand, is more common in smokers, similar to our case, a 78-year-old woman who was a smoker with primary tracheal SCC. Some other rare tumors such as carcinoid tumor, small cell carcinoma and benign tumors have been reported (1).

Webb and colleagues studied 74 patients with tracheal tumor within 60 years (1945–2005). It was found that 34 cases had SCC (45.9%), among whom, the most prevalent symptoms were dyspnea (55.4%), hemoptysis (48.6%), coughing (41.9%) and hoarseness (35.1%). Most of the patients were farmers and had a history of smoking. Among them, those who had primary surgical treatment and adjuvant radiotherapy had a higher rate of survival, as compared to those who had primary radiotherapy and/or chemotherapy (2).

Thotathil and colleagues studied 15 patients with primary tracheal malignancy between. Most of the cases were aged more than 40 years, and the majority of them were male (87%). Among them, most of the tumors were SCC (40%) and ACC (27%), respectively. One patient was treated through primary surgical resection, anastomosis and adjuvant radiotherapy, and the others underwent different protocols of chemotherapy or radiotherapy. Two patients received intraluminal brachytherapy. They concluded that radiotherapy was significantly effective for treating unre-sectable tumors (6).

Gaissert et al. reported 270 cases of tracheal tumor during 1962-2002. A total of 135 patients had SCC and 135 had ACC. Moreover, 191 patients (71%) received surgical resection and a number of cases did not undergo operation because of tumor length (67%), local invasion (12.4%), distant metastasis (7%) and other reasons (2%). In addition, 5 and 10-year survival rate in patients who had tracheal SCC and were treated by tumor resection were 39% and 18 %, respectively (7).

Grillo et al. reported 135 cases of tracheal SCC in a period of 26 years and suggested that the most prevalent site for metastasis was the paratracheal lymph nodes. Hematogenous metastasis to lung, liver, bone and sometimes to adrenals may also occur.



Figures 2 and 3. CT Scan of neck and chest from our patients showing tracheal stenosis due to tracheal tumor

Gender, age and prevalence of tracheal in SCC patients were similar to SCC of lung, mostly occurring in sixth and seventh decades of life frequently in males (8).

Unfortunately, malignant tracheal tumors are diagnosed too late and initially experience treatment for asthma, and at the time of diagnosis mostly are unresectable and suffocation may occur before surgery (9). Plain radiographs may show the tumor in the trachea (Figure 1), CT scan and also spiral CT reconstruction of trachea clearly demonstrate the location and spread of the tumor in the trachea (Figures 2, 3), and fibrotic bronchoscopy and biopsy (if possible) would confirm the diagnosis. If taking biopsy was not possible (because of possible bleeding), rigid bronchoscopy and sampling under general anesthesia would present tumor location and length, and confirm the diagnosis (similar to our case).

After endoscopic evaluation, decision to perform surgical procedure is made. The proper surgery is tumor resection followed by end-to-end anastomosis. Adjuvant radiotherapy is effective in cases with regional lymph nodes or microscopic marginal involvement, which can offer a better survival (9).

Nowadays, this tracheal resection and reconstruction method is the procedure of choice in more than 50% of tracheal SCC tumors, and complementary radiotherapy after the surgery is required in case of invasion to the regional lymph nodes or microscopic marginal involvement of the anastomosis (10).

Ahn and colleagues studied 37 cases of tracheal tumor, of whom 14 cases had benign and 23 cases had malignant tumors. Among the malignant tumor cases, 11 had tracheal SCC. They compared different treatment methods and surgical resection was recommended as the best treatment for tracheal SCC (11).

The staging classification for primary tracheal tumors which is most commonly accepted in the literature and is based on Bhattacharyya's staging system is outlined below:

T1: tumor < 2 centimeters and confined to trachea

T2: tumor > 2 centimeters and confined to trachea

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

T4: spread to adjacent structures

N and M staging are related to presence of lymphatic or distant metastasis (2, 12).

Based on this staging system, our patient was staged as a T2N0M0, in whom 3 centimeters of trachea was resected followed by end-to-end anastomosis, and due to the negative surgical margins, radiotherapy was not used. Annual follow-up by radiography and bronchoscopy was performed for five years (up to now). She is living healthy without any local recurrence or distant metastasis.

Conclusion

Primary tracheal tumor has a low rate of incidence and primary squamous cell carcinoma is one of its histological forms which occurs mostly in smokers. Given the common and nonspecific symptoms of these tumors, they are diagnosed too late and have a low rate of survival. Through presenting this patient, we aimed to draw attention to this uncommon malignancy and recommend considering it as a probable diagnosis when evaluating a patient with treatment-resistant respiratory symptoms. Our case had a good outcome after the surgery, and five years following the operation, she is currently healthy without recurrence.

Acknowledgments

This study was financially supported by Mashhad University of Medical Sciences, Mashhad, Iran.

Conflict of Interest

The authors declare no conflict of interest.

References

1. Abbate G, Lancella A, Contini R, Scotti A. A primary squamous cell carcinoma of the trachea: case report and review of the literature. Acta Otorhinolaryngol Ital. 2010; 30:209-12.
2. Webb BD, Walsh GL, Roberts DB, Sturgis EM. Primary tracheal malignant neoplasms: the University of Texas MD Anderson Cancer Center experience. J Am Coll Surg. 2006; 202:237–46.
3. Schneider P, Schirren J, Muley T, Vogt-Moykopf I. Primary tracheal tumors: experience with 14 resected patients. Eur J Cardiothorac Surg. 2001; 20:12-8.
4. Honings J, Gaissert HA, Ruangchira-Urai R, Wain JC, Wright CD, Mathisen DJ, et al. Pathologic characteristics of resected squamous cell carcinoma of the trachea: prognostic factors based on an analysis of 59 cases. Virchows Arch. 2009; 455:423-9.
5. Li W, Ellerbroek NA, Libshitz HI. Primary malignant tumors of the trachea: a radiologic and clinical study. Cancer. 1990; 66:894–9.
6. Thotathil ZS, Agarwal JP, Shrivastava SK, Dinshaw KA. Primary malignant tumors of the trachea–the Tata Memorial Hospital experience. Med Princ Pract. 2004; 13:69–73.
7. Gaissert HA, Grillo HC, Shadmehr MB, Wright CD, Gokhale M, Wain JC, et al. Long-term survival after resection of primary adenoid cystic and squamous cell carcinoma of the trachea and carina. Ann Thorac Surg. 2004; 78:1889–96.
8. Grillo HC. Primary tracheal neoplasms: surgery of the trachea and bronchi. London: Decker Inc; 2004. P. 207-8.
9. Honings J, Gaissert HA, Weinberg AC, Mark EJ, Wright CD, Wain JC, et al. Prognostic value of pathologic characteristics and resection margins in tracheal adenoid cystic carcinoma. Eur J Cardiothorac Surg. 2010; 37(6):1438-44
10. Li ZJ, Tang PZ, Xu ZG. Experience of diagnosis and treatment for primary cervical tracheal tumours. Chinese J Otorhinolaryngol Head Neck Surg. 2006; 41:208-10.
11. Ahn Y, Chang H, Lim YS, Hah JH, Kwon TK, Sung MW, et al. Primary tracheal tumors: Review of 37 cases. J Thorac Oncol. 2009; 4:635-8.
12. Bhattacharyya N. Contemporary staging and prognosis for primary tracheal malignancies: a population-based analysis. Otolaryngol Head Neck Surg. 2004; 131:639-42.