



ORIGINAL RESEARCH PAPER

Otorhinolaryngology

CASES OF PRIMARY SQUAMOUS CELL CARCINOMA OF TRACHEA IN NON-SMOKERS: OUR EXPERIENCE

KEY WORDS: Trachea, malignant, squamous cell carcinoma.

Dr. Tanusree Choudhury

Postgraduate trainee, Department of Otorinolaryngology, Silchar Medical College,

Dr. Smrity Rupa Borah Dutta*

Associate Professor, Silchar Medical College*Corresponding Author

Dr. Abhinandan Bhattacharjee

Associate Professor, Silchar Medical College

ABSTRACT

Primary tracheal tumours are extremely rare and overwhelmingly malignant. Squamous cell carcinoma is most common in smokers, whereas adenocarcinoma is most commonly found in non-smokers. Patients most commonly presents with hemoptysis, respiratory distress, cough and stridor. The symptoms appear in late phase and often are mistaken for chronic obstructive pulmonary disease. This leads to delay in management. Surgery, followed by adjuvant radiotherapy and chemotherapy, is the treatment of choice.

INTRODUCTION

Tumours, arising primarily from trachea, are rare. Most primary tracheal tumours are malignant. Squamous cell carcinoma is most common in smokers, whereas, adenoid cystic carcinoma is most commonly found in non-smokers. The tumour can be identified radiologically as intra-luminal, wall-thickening, exophytic lesion. The symptoms of this tumour is hemoptysis, dyspnoea, hoarseness, stridor, cough, which occur in late phase and thus, there is delay in diagnosis. Surgery, followed by adjuvant radiotherapy, is the choice of treatment. In inoperable cases, radiotherapy and chemotherapy is the treatment of choice. A case of primary tracheal cancer is described and a review of literature is presented.

CASE REPORT - 1

A 60 years old, non-smoker, presented with shortness of breath and noisy breathing for 1.5 months, and 2 swelling over anterior aspect of neck. There is no history of hoarseness or dysphagia. There is no pain over the swelling. The patient was initially treated elsewhere as suspected COPD.



Fig. 1,2,3- Presentation and clinical examination of the patient.

On examination, we find two spherical swellings, of size 2x2 cm² and 2x1 cm², were seen over the anterior aspect of neck, which were firm, well-defined with regular surface, extended from anterior border of right sternocleidomastoid to sterna head of left sternocleidomastoid, and did not move on deglutition. The overlying skin was normal and the swellings were fixed to the underlying structures. The patient also complained of weight loss for 1 month.

On laryngoscopy, we find normal laryngeal study with tracheal lesion.

Fine needle aspiration cytology from right sided level III and IV neck nodes, shows moderately differentiated squamous

cell carcinoma. Biopsy from mass, taken by bronchoscopy, shows features of squamous cell carcinoma.

On Computed Tomography of neck, we find a mass lesion, arising from lateral wall of proximal trachea, causing severe narrowing of tracheal lumen, posteriorly abutting prevertebral soft tissue from D1 to D4 level, posteromedially invading trachea-esophageal groove and proximal thoracic esophagus, anteriorly encasing right common carotid artery, laterally abutting proximal part of right subclavian artery, antero-superiorly invading inferior aspect of lobe of thyroid and infero-laterally apical pleura of right lung. Heterogenously enhancing lymph nodal mass in right lower jugular location, compressing right internal jugular vein.

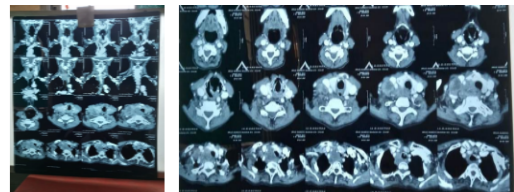
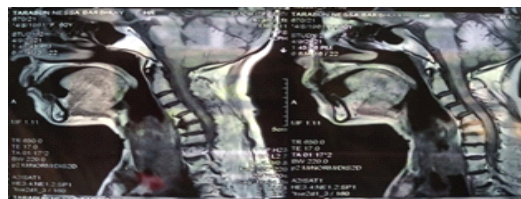


Fig. 4,5- CT scan shows tracheal infiltration.

The patient received six cycles of palliative chemotherapy with 28.2 mg cisplatin and 470 mg of 5-fluorouracil. The patient could not be treated surgically due to extent and general condition of the patient.

After 1 year, the patient presented in ENT OPD of Silchar Medical College with stridor, which was expiratory in nature. After that, MRI of neck and chest was done, which showed a well-defined intraluminal lobulated soft tissue density mass with internal septations, arising from the right posterolateral wall of trachea at the level of T2 and T3 vertebrae, causing expansion of trachea and luminal narrowing. The lesion appears isointense to the surrounding muscles on T1WI, and hyperintense on T2/STIR sequences. The lesion shows heterogenous enhancement on post contrast study.



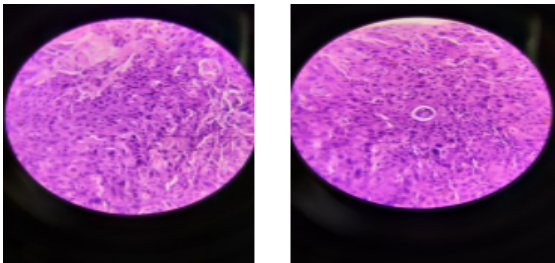
The patient was planned for radiotherapy, but the patient expired before that.

CASE REPORT- 2

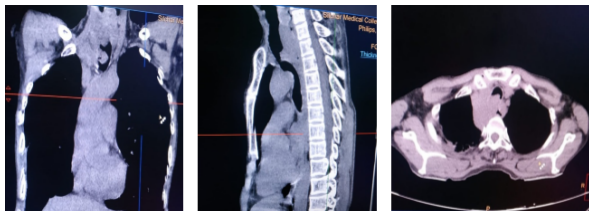
A 45 year old, non- smoker, presented with noisy breathing for 3 months, respiratory distress for 1 month and dysphagia for 15 days. The patient does not give any history of hoarseness of voice. This patient was also suspected to have COPD and was treated for the same.

On examination, the patient was found to be having expiratory stridor, which got slightly relieved if the patient sat. No lymph nodes were palpable in neck.

Bronchoscopy was done, which showed a mass at the level of carina. Biopsy was also taken from the mass, which showed squamous cell carcinoma.



Contrast enhanced CT of neck and thorax showed a circumferential asymmetrical growth, arising from posterior wall of trachea and protruding into tracheal lumen, at the level of C7 to T4 vertebral levels. The growth extends into cervical and upper thoracic esophagus.



The patient was planned for radiotherapy and chemotherapy, but the patient expired before that.

DISCUSSION

Primary carcinoma of the trachea is not common.¹⁻³ This rarity makes research into the natural history and treatment very difficult.⁴ Most primary cervical tracheal tumours are malignant: adenoid cystic carcinoma (ACC), squamous cell carcinoma (SCC), adenocarcinoma⁵⁻¹², mucoepidermoid carcinoma, carcinoid tumour³, oat cell carcinoma¹³.

SCC is the most common pathology in smokers, ACC is more prevalent in non-smokers.⁵ But SCC in non-smokers is extremely rare.

Benign tumours are xanthogranuloma and pleomorphic adenoma.³

A tumour, arising in the thyroid or oesophagus, can spread to trachea. Trachea can be site of metastasis from recurrent carcinoid tumour in the left main bronchus⁹, larynx, lung^{1,7}, colon².

The radiological appearance of the tumours can be classified as: intra-luminal, wall-thickening, exophytic form.⁷ Endoscopic evaluation reveals that the majority of the lesions are bulky and obstructive in nature.⁵

The tumours tend to be diagnosed late on account of delayed specific symptoms²: hemoptysis, dyspnoea, cough, hoarseness, stridor^{1,5-7}. The initial diagnosis is thought to be bronchial asthma.¹⁴

Computed tomography(CT) demonstrates tracheal wall thickening and extra-luminal changes.⁷

Progressive local disease can cause complications: fatal hemorrhage, oesophago-tracheal fistula, tracheal necrosis¹⁵, tracheal stenosis⁸.

Management of tracheal tumours includes interventional endoscopy, surgery, radiotherapy, endoluminal brachytherapy.³ Extensive segmental resection of the trachea is the treatment of choice.³ Upto 50% of the trachea can be resected with modern techniques.^{4,8} Surgery, followed by adjuvant radiotherapy and primary radiotherapy in inoperable cases represent potentially curative treatment options.^{5,9,16} The sleeve trachea resection is one of the optimal surgical modalities, the other options are- partial tracheal wall resection, immediate tracheal reconstruction, total laryngectomy + partial resection of trachea and thyroid lobectomy^{8,12}, resection and primary reconstruction, laryngotracheal resection, cervico-mediastinal exenteration, carinal resection and reconstruction¹⁶. Trachea anastomosis is suitable for small defects. The platysma myocutaneous flap combined with facial flap of the sternohyoid muscle, sternocleidomastoid myoperiosteal flap and the pectoralis major musculocutaneous flap are applied to reconstruct the defects of cervical trachea.¹¹ Endo-bronchial high dose - rate brachytherapy may be used for tracheal tumours, even as a boost for external beam irradiation or in recurrences. Long-term survival may also be expected, particularly for tumours with adenoid cystic histology.¹⁷ With tracheostomy, a curative excision or a palliative excision is possible.¹⁸

Palliation has improved with the introduction of laser resection, brachytherapy and stents. SCC may have better prognosis in the trachea than in lungs.⁴

After surgical management, the 3- year and 5-year survival rates are 79.80% and 48.36% for ACC, 80% and 20% for SCC.⁸ Carvalho Hde, et al. presented their experience with high dose-rate endobronchial brachytherapy; they reported a good local control, at the time of the first bronchoscopic control. They treated 4 patients with non-resectable tracheal tumour, out of which two patients with SCC died at 6th and 33rd month, after treatment, only second patient presented local recurrence. The other two patients were alive after 64 and 110 months of follow-up.¹⁷

The 1-, 3-, 5- year survival rates, following radiation therapy alone, are 64.7%, 64.7% and 26% for SCC, 85.7%, 85.7% and 85.7% for ACC. Patients with ACC and those with complete remission following treatment had a significantly better survival probability.¹⁵

The 5-year survival rate for low grade malignant tumours arising in trachea, like, carcinoid, muco-epidermoid carcinoma, adenoid cystic carcinoma, is 78.8%.³

CONCLUSION

Primary tumours of trachea are rare. Out of which, SCC is found in smokers, and ACC is found in non-smokers. SCC in non-smokers, are extremely rare. Average duration of delay of diagnosis is 12 months.¹⁵ In our case, it was 4 years. The annual incidence of primary tracheal malignancies is 0.142 per 100,000 people.²⁰ Male:female is 8:3 in SCC, 3:6 in ACC.²¹ Patients with airway tumours, typically have a delay in diagnosis as the symptoms are mistaken for bronchial asthma or chronic obstructive pulmonary disease. In our case, the patient was treated initially as a case of COPD. Treatment depends on general condition of the patient and extension of the lesion. Surgical treatment is possible only in small lesions, which are resectable.

In our case, non-surgical intervention was done due to extent of the disease, and the patient received palliative chemotherapy.

An elderly patient with long-standing respiratory distress, should be assessed for tracheal lesions to prevent delayed diagnosis and treatment.

REFERENCES

1. Goldstein J. Primary carcinoma of the trachea: report of two cases. *Southern Medical Journal*. 1977;70(4):434-436.
2. Morency G, Chalaoui J, Samson L, Silvestre J. Malignant neoplasms of the Trachea. *Canadian Association of Radiologists Journal*. 1989;40(4):198-200
3. Schneider P, Schirren J, Muley T, Vogt- Moylopf I. Primary tracheal tumors: experience with 14 resected patients. *European Journal of Cardio-Thoracic Surgery*. 2001;20(1):12-18.
4. Hetzel M. Tracheal tumours: Could treatment be better?. *Clinical Oncology*. 1993;5(5):272-276.
5. Webb B, Walsh G, Roberts D, Sturgis E. Primary tracheal malignant neoplasms: the University of Texas MD Anderson Cancer Centre experience. *Journal of the American College of Surgeons*. 2006;202(2):237-246.
6. Howard D, Haribhakti V. Primary tumours of the trachea: analysis of clinical features and treatment results. *Journal of Laryngology and Otolaryngology*. 1994;108:230-232.
7. Li W, Ellerbroek N, Libshitz H. Primary malignant tumours of trachea. A radiologic and clinical study. *Cancer*. 1990;66(5):894-899.
8. Li ZJ, Tang PZ, Xu ZG. Experience of diagnosis and treatment for primary cervical tracheal tumours. *Zhonghua Er Bi Yan Hou Ke Za Zhi* 2006;41:208-210.
9. Grillo H. Reconstruction of the trachea after resection for neoplasm. *Head and Neck Surgery*. 1981;4(1):2-8.
10. Mathisen D. Primary tracheal tumour management. *Surgical Oncology Clinics of North America*. 1999;8(2):307-326.
11. Pan X, Lei D, Xu F, et al. Surgical management of primary cervical tracheal cancer. *Zhonghua Er Bi Yan Hou Ke Za Zhi*. 2003;38:437-490.
12. Yamazaki K, Kubo Y, Hirasawa M, et al. A study on low grade malignant tumours arisen in the trachea and the bronchus. *Kyobu Geka* .1997;50:939-941.
13. Sweeney E, Hughes F. Primary carcinoma of the trachea. *Histopathology*. 1977;1(4):289-299.
14. Inoue H, Ishihara T. Tracheal tumour resection and reconstruction surgery. *Nihon Kyobu Shikkan Gakkai Zasshi*. 1990;28:272-277.
15. Harms W, Latz D, Becker H, et al. Treatment of primary tracheal carcinoma. The role of external and endoluminal radiotherapy. *Strahlenther Onkol*. 2000;176:22-7.
16. Grillo H, Mathisen D. Primary tracheal tumours: Treatment and results. *The Annals of Thoracic Surgery*. 1990;49(1):69-77.
17. Carvalho H, Figueiredo V, Pedreira W, Aisen S. High dose-rate brachytherapy as a treatment option in primary tracheal tumours. *Clinics* .2005;60(4):299-304.
18. Yuan X, Li H, Wang P. Primary tumour of the trachea with a report of 24 cases. *Zhonghua Zhong Liu Za Zhi*. 1995;17:311-313.
19. Gaissert H, Burns J. The Compromised Airway: Tumors, Strictures and Tracheomalacia. *Surgical Clinics of North America*. 2010;90(5):1065-1089.
20. Madariaga M, Gaissert H. Overview of malignant tracheal tumors. *Annals of Cardio-thoracic Surgery*. 2018;7(2):244-254.
21. Ahn Y, Chang H, Lim Y, Hah J, Kwon T, Sung M et al. Primary Tracheal Tumours: Review of 37 cases. *Journal of Thoracic Oncology*. 2009;4(5):635-638.