# Case Report

## Primary Tracheal Papilloma: A Case Report

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## Abstract

Solitary papilloma in the respiratory tract is a rare benign epithelial tumor which is complete surgical excision of the current standard treatment for this type of tumor. Here a case of solitary tracheal papilloma treated by surgical resection is reported. Due to rarity and non-specific symptoms, tracheal papilloma always subjected to misdiagnosed and suffer from delayed treatment. In this case, a forty two years male has been presented with a recurrent non-productive irritative cough, a progressive shortness of breath, expiratory stridor and occasional hemoptysis. The patient was previously diagnosed as a case of bronchial asthma by a Pulmonologist and wrongly treated as well. CT scan revealed an intraluminal tracheal mass arises from the right side of the tracheal wall opposite c6-c7 vertebrae. The tumour was removed by endoscopic excision. The histopathological result confirms the diagnosis of squamous cell papilloma. No complications occur during surgery and no recurrence was observed in six months after surgery on followup.

Keywords: Trachea, papilloma, excision.

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# INTRODUCTION

Tracheal papilloma is a rare neoplasm in the respiratory system approximately 0.38 % of all lung tumors.<sup>1</sup> Most of the cases 90% of all cases of primary tracheal tumors are malignant. Most of these tumors are squamous cell carcinoma and adenoid cystic carcinoma.<sup>2</sup> A tracheal papilloma is characterized by the papillomatous growth from the bronchial epithelium which in response to HPV infection, most commonly HPV-6 & 11.<sup>3</sup> Tracheal papilloma can be classified into three categories according to histological type: squamous cell papilloma, glandular papilloma and mixed.<sup>4</sup> Here a case of primary tracheal papilloma in distal trachea treated by surgical resection is reported.

#### CASE REPORT

Mr. Masud, 40 years old male presented with cough for two years which was initially non- productive and intermittent but gradually became irritative and occasionally associated with hemoptysis. The cough was aggravated by exertion and relieved by taking rest. The patient also gave the history of dyspnoea and expiratory stridor during cough or exertion. He was a smoker and took 10-15 cigarettes per day for the last 15 years. He did not give any history of fever, common cold, chest pain, diurnal variation of cough, weight loss, any change of voice or difficulty in swallowing. He was treated for those complaints as bronchial asthma though the symptoms did not relive.

On fibre optic laryngotracheal examination, found there is pale irregularly surfaced mass in the tracheal lumen about 1.5 cm distal to the vocal folds almost occluding the tracheal lumen and moves with respiration. Other parts of larynx appear normal with normal vocal cord mobility. On examination, he did not have any hoarseness of voice. Other ENT and systemic examination reveals no abnormality.



Figure-1: Photograph of the patient with kind permission

X-ray Soft tissue neck lateral view reveled irregular soft tissue shadow occupying tracheal lumen opposite c6-c7 vertebra. (Figure-2).



Figure-2: X-ray Soft tissue neck lateral view

CT scan revealed heterogeneously enhancing irregular lesion measuring about  $2.1 \times 2 \times 1.4$  cm is seen at the right side of the tracheal lumen opposite the level of c6-c7

vertebrae, occupying most of the tracheal lumen (figure 3, 4). Decision was made for surgical intervention. Tracheostomy was done for ventilation, followed by endoscope assisted excision of the mass.



Figure-3: CT Axial View

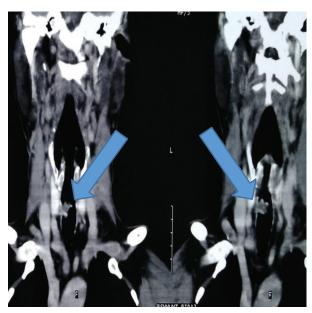


Figure-4: CT Coronal view

Histopathological examination of the specimen was reported as Squamous cell Papilloma

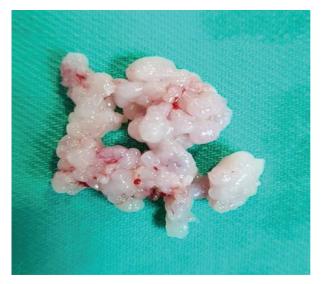


Figure-5: Specimen after excision

## DISCUSSION

A tracheal papilloma is an uncommon tumor of trachea consisting of 0.38 % of all lung tumours.<sup>1</sup> It may affect any part of the trachea but a proximal and distal third are most frequently affected.<sup>5</sup> It can be solitary, multiple or as few of recurrent respiratory papillomatosis. Respiratory papillomatosis is common in the larynx, although it can affect any part of the tracheobronchial tree. It is uncommon in the trachea. Distal trachea involvement was reported only in the 5% of recurrent respiratory papillomatosis cases.<sup>6</sup> In a study tracheal papilloma was reported in only 5 cases of 15,000 bronchoscopies.<sup>7,8</sup> Tracheal papilloma present in two forms - adult-onset or juvenile-onset. Adult-onset types are mostly to be malignant whether juvenile-onset are benign. Adult-onset types affect male and female at a ratio of 4:1.6 Most of the tracheal papilloma are due to HPV-6 and HPV-11. It is thought that HPV manifested by a vertical transmission during vaginal delivery of an infected mother.<sup>9</sup>

In this case, 42 years old male had no previous history of recurrent respiratory papillomatosis in childhood or maternal HPV. The clinical presentation of tracheal papilloma is non-specific. Symptoms may vary from cough and dyspnea to stridor and upper airway obstruction.<sup>10</sup> In this case the presenting complaints was a non-productive cough, shortness of breath and occasional hemoptysis. He was initially treated as bronchial asthma by a chest specialist. On chest X-ray, they could not identify the tracheal mass. When the patient comes to Bangabandhu Sheikh Mujib Medical University ENT OPD, initially, he

was advised X-ray soft tissue neck lateral view. Then found irregular shaped soft tissue lesion on tracheao-oesophagal region opposite c6 & c7 vertebrae. Then the patient was advised for CT scan and found heterogeneously enhancing irregular mass lesion measuring about  $2.1 \times 2 \times 1.4$  cm seen at the right side of the tracheal lumen opposite the level of c6-c7 vertebrae, occupying most of the tracheal lumen. The case diagnosed as a primary tracheal mass and decided to excise the mass surgically.

The histopathological report revealed squamous cell papilloma.

Squamous cell papilloma may transform squamous cell carcinoma and it is reported in 3 to 5% of all cases of tracheal papilloma.<sup>11</sup> Risk factors for malignant transformation is smoking, HPV infection (types 16 & 18), radiotherapy and chemotherapy.<sup>12</sup>

Treatment of tracheal papilloma is challenging and often required for recurrent manipulation. Treatment modalities dependon the type, severity, number and location of papilloma.<sup>13</sup> There are a variety of treatment modalities like endoscopic surgical method, excision by  $\rm CO_2$  laser, cryotherapy, photodynamic therapy , laser vaporization, electrocautery, argon laser coagulation, open approach etc.<sup>14</sup>

In this case, endotracheal intubation was not possible as the mass was almost occluding more than 70% of the lumen and there was anticipation for bleeding as was not confirmed about the mass. So tracheostomy was performed for ventilation and by properly securing the lower respiratory tract excision of the mass was done by endoscopically. Low lying tracheostomy was performed as the mass was opposite cervical 6 & 7 vertebrae.

Local recurrence is very common in papilloma after treatment. This patient is followed every 3 months interval and still, there is no recurrence.

# CONCLUSIONS

As tracheal papilloma is a rare disease. A Surgeon should have a high index in suspicion in patients presenting with obstructive symptoms, hemoptysis, persistent non-productive cough and history of exposure to HPV. Early diagnosis, proper surgical excision and close follow-up is mandatory for this type of patient.

**Conflict of interest**: The authors declare that they have no conflict of interest.

## **REFERENCES:**

- Popper HH, Wirnsberger G, JüttnerSmolle FM, Pongratz MG, Sommersgutter M. The predictive value of human papilloma virus (HPV) typing in the prognosis of bronchial squamous cell papillomas. Histopathology. 1992 Oct;21(4):323-30
- Kitada M, Yasuda S, Ishibashi K, Hayashi S, Matuda Y, Ohsaki Y, Miyokawa N. Leiomyoma of the Trachea: a case report. Journal of cardiothoracic surgery. 2015 Dec 1;10(1):78
- Valentino J, Brame CB, Studtmann KE, Manaligod JM. Primary tracheal papillomatosis presenting as reactive airway disease. Otolaryngology—Head and Neck Surgery. 2002 Jan;126(1):79-80.
- Li JW, Yan JX, Cao Y, Feng GZ, Gao W. Case Report Solitary mixed type papilloma in trachea: a case report and literature review. Int J Clin Exp Med. 2018;11(5):5286-9.
- Bhate JJ, Deepthi NV, Menon UK, Madhumita K. Rare Benign Tracheal Lesions. Int J Phonosurg Laryngol. 2012;2(1):37-40
- Anar C, Erer OF, Yavuz MY, Yücel N. An Isolated Tracheal Papilloma: A Case Report. Respiratory Case Reports. 2017 Jan 1;6(1).
- Miura H, Tsuchida T, Kawate N, Konaka C, Kato H, Ebihara Y. Asymptomatic solitary papilloma of the bronchus: review of occurrence in Japan. European Respiratory Journal. 1993 Jul 1;6(7):1070-3.

- Naka Y, Nakao K, Hamaji Y, Nakahara M, Tsujimoto M, Nakahara K. Solitary squamous cell papilloma of the trachea. The Annals of thoracic surgery. 1993 Jan 1;55(1):189-93.
- Kosko JR, Derkay CS. Role of cesarean section in prevention of recurrent respiratory papillomatosis—is there one? International journal of pediatric otorhinolaryngology. 1996 Mar 1;35(1):31-8.
- Breen DP, Lyons O, Barrett H, Burke C. Isolated tracheal papillomatosis—an infrequent cause of chronic cough. Respiratory Medicine Extra. 2007 Jan 1;3(1):21-2.
- Ogata-Suetsugu S, Izumi M, Takayama K, Nakashima T, Inoue H, Nakanishi Y. A case of multiple squamous cell papillomas of the trachea. Annals of thoracic and cardiovascular surgery. 2011 Apr 25;17(2):212-4.
- 12. Guillou L, Sahli R, Chaubert P, Monnier P, Cuttat JF, Costa J. Squamous cell carcinoma of the lung in a nonsmoking, nonirradiated patient with juvenile laryngotracheal papillomatosis. Evidence of human papillomavirus-11 DNA in both carcinoma and papillomas. The American journal of surgical pathology. 1991 Sep;15(9):891-8.
- Harris K, Chalhoub M. Tracheal papillomatosis: what do we know so far?. Chronic Respiratory Disease. 2011 Nov;8(4):233-5.
- Cömert SŞ, Parmaksız ET, Çağlayan B, Gülseven HT, Salepci B, Fidan A. Typical carcinoid and benign endobronchial tumour cases treated with interventional bronchoscopic techniques. Eurasian Journal of Pulmonology. 2013 Apr 1;15(1):39-44.