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CASE REPORTS

Primary tracheal papilloma disguised as asthma: A case reportYan-bin Chen, MD, PhD¹, Jun-hong Jiang, MD, PhD¹, Ling-chuan Guo, MD, PhD², and Jian-an Huang, MD, PhD¹¹Department of Respiratory Medicine, the First Affiliated Hospital of Soochow University, Suzhou, China and ²Department of Pathology, the First Affiliated Hospital of Soochow University, Suzhou, China**Abstract**

Introduction: Tracheal papilloma presenting as asthma is a rare occurrence. **Case Study:** We report a case of a 32-year-old male patient who presented with features of asthma. Flexible bronchoscopy demonstrated a large growth arising from the lower end of the trachea. Successful treatment using snare loop and argon plasma coagulation (APC) of the polypoid growth was performed via flexible bronchoscope. **Results:** The patient had immediate relief of airway obstruction and histopathological examination of the neoplasm demonstrated features of papilloma. **Conclusion:** Primary tracheal papilloma is mimicker of asthma, CT scan should be considered in patients with persistent chronic cough, or stridor. Endoscopic papillectomy is a safe and effective treatment and should be considered as first-line therapy for tracheal papilloma.

Keywords

Tracheal neoplasm, papilloma, asthma, interventional pulmonology

History

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Introduction

Endotracheal benign tumors are rare. Papillomas are among the commonest endotracheal benign tumors, however, primary tracheal papillomas presenting as asthma are uncommon. We herein report a case of a young male patient who presented with asthma. His plain chest film was normal, yet chest computed tomography (CT) scan showed endotracheal mass. Bronchoscopic examination of the airways demonstrated the presence of a large tracheal tumor, which was successfully removed with diathermic snare loop and argon plasma coagulation (APC).

Case study

A 32-year-old man suffering from chronic cough for 1 year came to our outpatient department. The patient's health had been good until 1 year before, when he began to have episodes of cough. He was a hairdresser without history of allergic rhinitis. He had no history of unconsciousness or foreign body aspiration, no history of smoking. The patient was well developed and well nourished. General physical examination was unremarkable. Plain chest films did not find any abnormalities (Figures 1A and 1B).

Bronchial dilation test revealed the forced expiratory volume in 1 second (FEV₁) increasing from 1.86 L to 2.09 L after inhaled bronchodilator, and FEV₁/forced vital capacity (FVC) was 44.06%, which demonstrating severe obstruction. According to Global Initiative for Asthma (GINA) 2014, the patient

was diagnosed as asthma, and budesonide/formoterol (Symbicort) 160/4.5 was prescribed, two inhalations twice daily. His condition improved to some extent, but 2 months later, he came to our clinic again with chief complaint of coughing up pieces of papillomatous tissue, and several times of the episode occurred during the past 1 year, yet he did not share it with the clinician in his first visit.

Chest CT was arranged immediately and an endotracheal mass arising from the lateral wall of the trachea was found (Figures 1C, 1D). There was no mediastinal lymph node enlargement and both lungs appeared normal. Flexible fiberoptic bronchoscopy (FOB), under local anesthesia, was performed for further assessment, which revealed that the larynx and vocal cords appeared normal, but a fleshy polypoid lesion with a wide base and a cauliflower-like appearance was seen in the trachea about 2.5 cm above the carina from 12 to 6 o'clock position occluding approximately 60% of the lumen, its surface was lustrous with a little purulent secretion (Figure 2A).

The bronchoscopy can pass through the lumen to the carina and main bronchus. The major bronchi and the distal bronchial segments were normal. A biopsy specimen was taken and the tumor was removed using snare loop and APC (Figure 2B). The snare loop (Micro-Tech, Nanjing, Co., Ltd.) was used to encircle and firmly tighten around the basal part of the polypoid growth and the snare loop was pulled very slowly to avoid bleeding. Then APC was performed with an argon plasma coagulator unit (VIO 200S APC2, ERBE).

APC monopolar probe was inserted through the working channel of the bronchoscope. The target tissue was endoscopically visualized and then coagulated. The power was 40 W. No procedural complication was observed. The patient reported marked relief from cough after bronchoscopy.

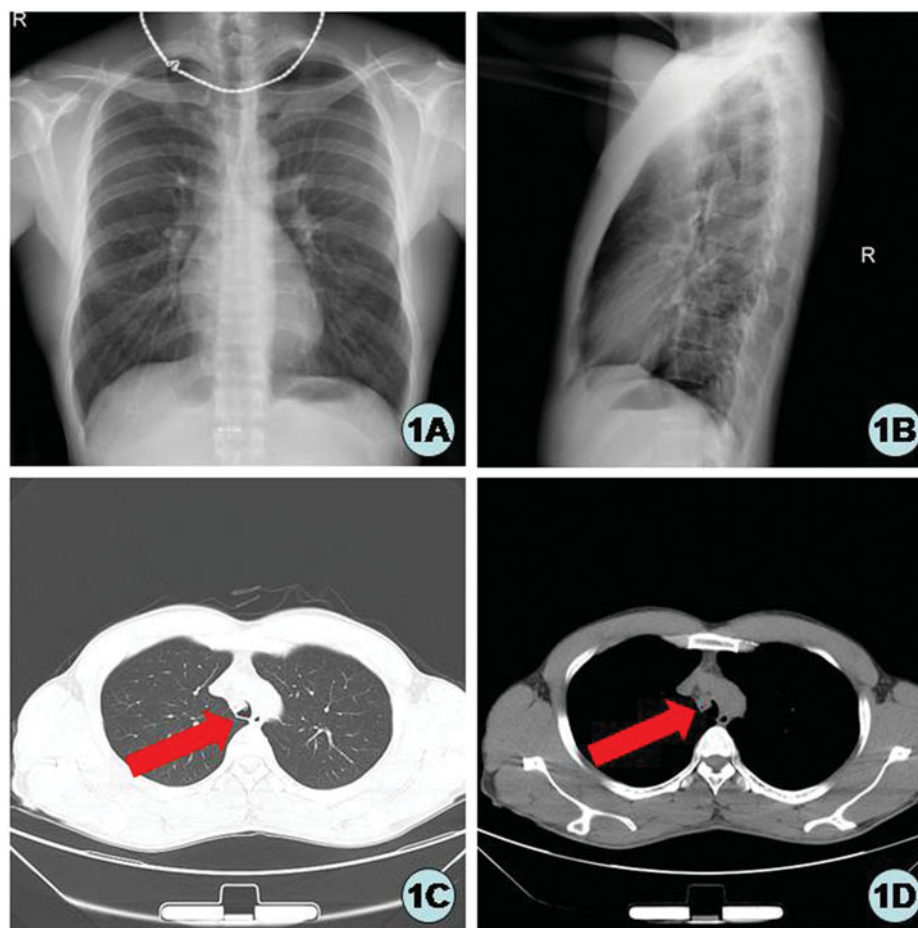


Figure 1. (A, B) Anteroposterior and lateral radiographs showing no tumor. (C, D) CT scan (lung window) showing an endotracheal mass protruded into the trachea, resulting in an approximately 60% reduction of the cross-sectional area. The tumor also can be seen in the mediastinal window.

And follow-up bronchoscopic examination 1 week later showed that the lumen of trachea was nearly normal (Figure 2C). All bronchodilators were discontinued and FEV₁ and FVC with bronchodilators were 3.49 L (93.3% of predicted) and 4.46 L (102.6% of predicted), respectively, FEV₁/FVC was 78.2%. The histological appearance was that of a tracheal papilloma with some focal mild atypical hyperplasia, no evidence of malignancy was found (Figure 2D). The patient is under regular follow-up to date.

Discussion

The patient, who was a hairdresser, manifested chronic cough. His bronchial dilation test was positive and also showed persistent chronic airflow limitation. At the same time, his plain chest film was normal. So we diagnosed him as asthma and prescribed budesonide/formoterol for him. His condition improved to some extent until two months later when he coughed up pieces of papillomatous tissue, and received chest CT and FOB examination. Primary tracheal papilloma was diagnosed at last.

The papilloma is one of the most common benign tumours of the trachea with a small possibility of malignant transformation, which occurs in 3% to 5% of patients [1]. According to previous reports, most papillomas are found in larynx, only 5% of patients had distal involvement of trachea [2]. Its incidence is reported to be only five cases in 15,000 bronchoscopies [3]. Or

1.8 cases per 100,000 people in adolescents older than 15 years [1]. Tracheal papilloma may be caused by human papillomaviruses (HPV), either HPV-6 or HPV-11 [4]. HPV-negative biopsies occurred more frequently in adult-onset patients [5]. These tumors are 4 times more likely to be found in males than in females and usually present in the fifth to seventh decade of life [6]. Yet, the patient in our report did not show any hints of HPV infection and was younger than the data in the literature.

Clinical symptoms of tracheal papilloma depend on the type of tumor, extent of endotracheal obstruction, and the degree of distal parenchymal damage. The major clinical features are chronic cough, dyspnea on exertion, wheezing, repeated respiratory infection, intermittent hemoptysis, and blood-stained sputum. The most common physical finding is airway obstruction manifested by stridor, wheeze or rhonchi, but usually the physical examination is negative [7]. As to the patient in our report, chronic cough was the main symptom, stridor or rhonchi was not to be heard in spite of more than half of the tracheal lumen obstructed.

The chest radiograph is usually not helpful, just as the one in our report. CT is a useful tool, which can document the size, location, and involvement of surrounding structures. When viewed endoscopically, papilloma appears as a warty or pedunculated mass, and is prone to hemorrhage after touching due to its fragile nature. Histology shows papillary lesions with keratinized epidermoid cells. The degrees of metaplasia and dysplasia are index of malignant transformation. The viral

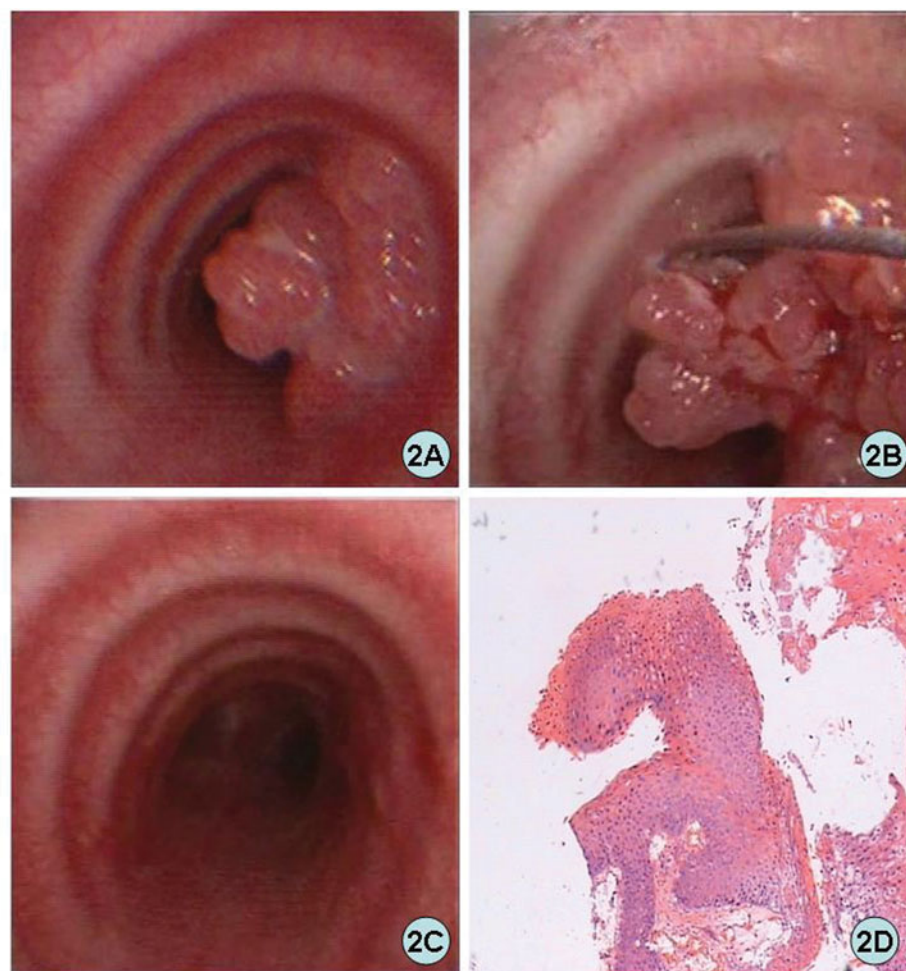


Figure 2. (A) Bronchoscopy showing a cauliflower-like lesion in the trachea about 2.5 cm above the carina from 12 to 6 o'clock position. (B) Bronchoscopic electrosurgical removal of tumor of lower third of the trachea with diathermic snare loop. (C) After snare and APC therapy, most parts of the tumor are removed. (D) HE stain showing histological appearance of papilloma with some focal mild atypical hyperplasia, no evidence of malignancy was found ($\times 20$).

infected pattern, koilocytes, is also found sometimes. We did not find any koilocytes from the histopathologic slide of our patient. The final diagnosis is usually made by thorax CT, bronchoscopy and histological examination. Pulmonary function studies can partially reflect its severity [8].

The patient in our report was diagnosed as asthma in his first visit, partially due to his ignoring history of coughing up papillomatous tissue before, partially due to the doctor's choice of plain chest film. If the patient could provide past history in detail, or if the doctor chose chest CT instead of plain chest film, tracheal papilloma should be diagnosed earlier.

Tracheal papillomas can be divided into three groups: multiple papillomatosis, inflammatory polyps and solitary papilloma [9]. Solitary papilloma, mostly found in elderly people, can be divided into two categories. One is defined as solitary papilloma covered with columnar or cuboidal epithelium, with or without squamous metaplasia. The other type consists of solitary squamous papilloma, which has well-formed connective tissue stroma, covered by stratified squamous epithelium [10].

The aim of treatment for tracheal papilloma is to accomplish a tumour-free state and maintenance of the appropriate airway for breathing. Currently, bronchoscopic ablation is a useful and efficacious modality for the management of tracheal papilloma as removal is both diagnostic and curative. Electrosurgery

with snare loop has been reported as a safe procedure that may be used with flexible bronchoscopy requiring only local anesthesia and is associated with short procedure time [11]. APC, which uses a high-frequency electrical current fed from a probe tip through ionized argon plasma, is useful in causing superficial thermal coagulation of tissue, and does not result in tissue carbonization or vaporization. All these advantages make it suitable for the treatment of papillomas, which is a superficial epithelial tumour [7].

The patient in our report obtained remarkable relief after bronchoscopic snare loop and APC therapy. The second check of FOB one week later showed lumen of trachea was nearly normal.

Tracheal papillomas may recur at any time and approximately 2–3% or 3–7% of the patients can undergo malignant conversion (squamous cell carcinoma) [12, 13]. And fewer than 1 in 5 adult patients will develop spread of disease to the lungs [4].

Conclusion

Primary tracheal papilloma is mimicker of asthma and chest radiographs can be normal; hence, CT scan should be considered in patients with persistent chronic cough, or stridor.

Endoscopy is the main method to make a definite diagnosis. Electrocautery with snare loop and APC under local anesthesia can lead to rapid palliation and immediate tumor debulking. Endoscopic papillectomy is a safe and effective treatment and should be considered as first-line therapy for tracheal papilloma. Although the patient has an excellent outcome, close follow-up is necessary for early detection and treatment of malignant transformation and recurrence [13, 15–17].

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Yan-bin Chen and Jun-hong Jiang contributed equally to this work as co-first authors.

Declaration of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the article.

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