

Case Report

Solitary mixed type papilloma in trachea: a case report and literature review

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Abstract: Solitary papilloma in the respiratory system is thought to be a rare benign epithelial tumor, and complete surgical resection is currently the standard treatment for this type of tumor. We report a case of solitary mixed-type tracheal papilloma treated with a snare under a tracheoscope. Tracheal papilloma is rare, and mixed-type papilloma is rarer. The patient's clinical manifestations were recurrent cough. After removal of the tumor by using a snare under bronchoscopy, the patient's cough resolved. No recurrence was observed in the following 3 months after the treatment. No complications occurred during the treatment. Mixed-type tracheal papilloma in adults can be clearly diagnosed using bronchoscopy. Local excision is thus recommended for solitary mixed-type tracheal papilloma.

Keywords: Solitary papilloma, mixed type of papilloma, bronchoscopy

Introduction

Solitary bronchial papilloma is a rare neoplasm in the respiratory system, accounting for approximately 0.38% of all lung tumors [1]. Solitary bronchial papillomas are subclassified into three categories according to histological type: squamous cell papilloma, glandular papilloma, and mixed squamous cell and glandular papilloma (mixed papilloma). Solitary mixed-type tracheal papilloma is even rarer. The present case report features a rare case of solitary tracheal papilloma and describes the clinicopathological characteristics, diagnostic challenges, and selection of appropriate and adequate treatment.

Case presentation

The patient, a 52-year-old man, was hospitalized owing to the complaint of interrupted cough for more than 1 year. The patient experienced recurrent cough with no obvious cause since June 2016, which appeared to be a mild,

mainly dry cough, occasionally with little white phlegm and no blood in the sputum; further, there was no history of mild fever or sweating, chest tightness, shortness of breath, regurgitation belching, or congestion. Early diagnosis and treatment failed to attach importance. However, the cough recurred with no significant improvement. The patient visited our hospital for an outpatient examination on June 20, 2016. Chest computed tomography revealed a trachea-occupying (**Figure 1A**). Subsequently, fiberoptic bronchoscopy (performed in June 22) indicated a cauliflower-like neoplasm approximately 4 cm away from the subglottis of the trachea on the left side (**Figure 1B**). The patient was admitted to the hospital for further treatment. During the course of the disease, the patient had no fever and no significant weight loss. He had a smoking history of more than 30 years (20/day), with a history of hypertension for 5 years. He was given oral Adalat 30 mg once daily for antihypertensive treatment. Physical examination revealed a temperature of

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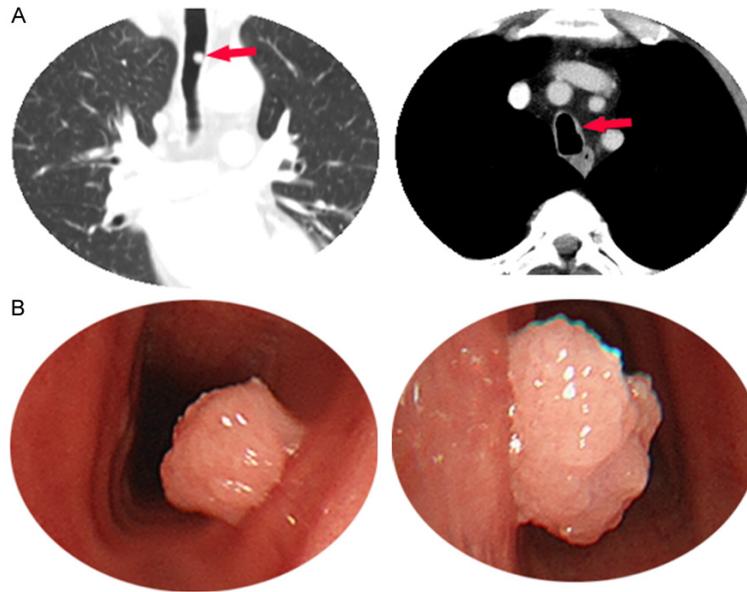


Figure 1. Imaging and flexible bronchofiberscope findings in the case. A. Chest computed tomography (CT) shows a 2 × 2-cm nodule in the middle of the trachea (arrow). B. A cauliflower like neoplasm is located at about 4cm away from subglottis of the trachea.

36.7°C, a pulse rate of 72 beats per minute, respiratory rate of 18 times per minute, blood pressure of 132/78 mm Hg, lucid spirit, and lips without cyanosis. The thorax was symmetrical, without tenderness of the sternum. Respiratory motion on both sides was symmetrical. Tactile fremitus was normal and bilaterally symmetrical. Double lung percussion was voiceless. Auscultation of the lungs was rough and dry, with wet rales. No abnormality was observed in the heart and abdomen, and no edema was found in the lower limbs. The admission examination results were as follows: white blood cell count, $4.88 \times 10^9/L$; neutrophil count, 61.5%; red blood cell count, $4.78 \times 10^{12}/L$; hemoglobin level, 135 g/L; platelet count, $206 \times 10^9/L$; and tumor markers: CA50 (-), CA125 (-), CA19-9 (-), CA15-3 (-), CEA (-), and NSE (-). A pulmonary function test revealed normal ventilation function (FEV1 accounted for 95%, FEV1/FVC = 90.97%). His coagulation function was normal. In the preliminary diagnosis, the nature of the airway tumor remained unclear. With the signed consent of the patient on June 24, 2016, interventional bronchoscopy of the laryngeal mask airway was performed under general anesthesia. By using a disposable laryngoscope in fiberoptic bronchoscopy (Olympus 1T-260) under general anesthesia, a

papillary neoplasm with 1-cm diameter was detected in the middle and upper parts of the trachea under the fiberscope. The color was consistent with the tracheal mucosa. The neoplasm was connected with a wide base to the tracheal wall with an uneven surface, and the other part of the lumen was patent. In the operation, a snare to trap the base of the papilloma was used, removing it by coagulation under 30-W power. Most of the prominent luminal part of the tumor was resected without transoperative bleeding, removed using biopsy forceps, and then sent to the pathology department. Postoperative pathological examination revealed mixed-type, phosphorus-like cells and glandular papilloma (**Figure 2**).

After removal of the tumor by a snare under bronchoscopy, the patient's cough resolved. No recurrence was observed in the following 3 months after the treatment. No complications occurred during the treatment.

Discussion

Papilloma is common in the larynx but rare in the trachea and bronchus. Tracheal papillomas originate from the trachea, bronchial epithelium, and mucous glands. With regard to the number of papillomatous lesions in the trachea and bronchus, pulmonary papillomas are divided into two types, namely multiple and solitary. Multiple-type papilloma is the most prevalent, mainly found in adolescents, which is related to human papilloma virus (HPV) infection, usually occurring in the upper respiratory tract and partly spreading to the lower respiratory tract [2]. The three pathological types of papilloma are squamous cell type, adenoid cell type, and mixed squamous epithelium and glandular epithelium (mixed type), among which the squamous cell type is the most common. None of the patients in the cases reported in the English literature had a solitary mixed-type papilloma at the main trachea. Similar cases published in the literature are summarized in **Table 1**. In

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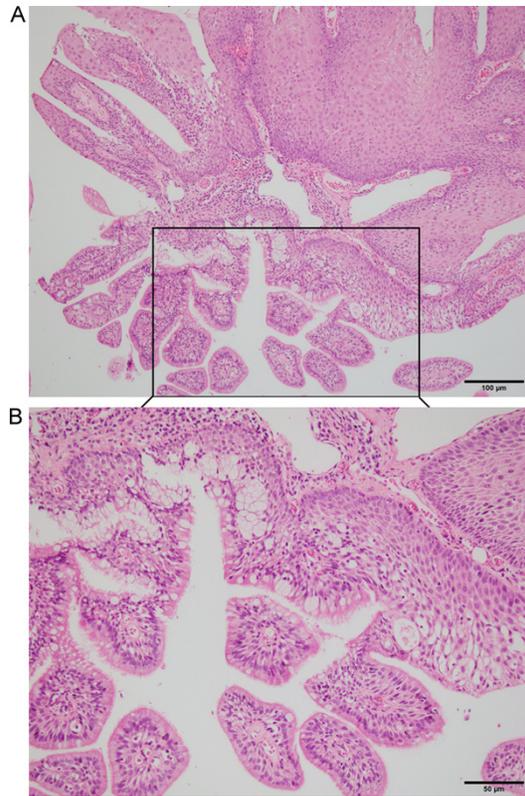


Figure 2. Pathological findings in the case. A. Low-power histologic view of the resected tumor HE (hematoxylin-eosin) staining. The tumor is composed of a fibrovascular core and papillomatous fronds lined by pseudostratified columnar epithelium. B. High-power view of a HE staining. The pseudostratified columnar epithelium consists of ciliated columnar cells and numerous mucous cells, with no cytologic or architectural atypia.

adults, mixed-type papillomas mainly occur in elderly individuals aged 50-60 years. Further, they occur more commonly in male patients (more than half of whom have a history of smoking). The male-to-female ratio is approximately 4:1 [3]. The cause of solitary mixed-type papilloma remains unclear. Studies indicate that this disease is associated with smoking and HPV infection [4]. Trvfon et al. [5] reported that the proportion of patients with the disease who had a smoking history was 78%. However, Inamura et al. [6] reported that 8 patients underwent an HPV test and tested negative. The results of the HPV tests of 7 domestic cases of solitary mixed-type tracheal papilloma, 4 of which were reported by Lu and 3 by Dong, were all negative. The present case was also HPV negative. The clinical manifestation

of solitary papilloma is complex. It can be manifested as cough, wheezing, hemoptysis or bloody sputum, chest tightness, and shortness of breath, and no clinical symptoms are observed; thus, it is detected only in a physical examination. Our patient had a dry cough, which he ignored and was detected in the routine examination. The biopsies of most mixed-type papillomas by using bronchoscopy indicate the central-type lesion, which is often misdiagnosed as lung cancer. According to the literature report, the bronchoscopy results of mixed-type papillomas vary; most papillomas show a new, cauliflower-like morphology, part of which shows occult growth. Some bronchoscopic examinations have revealed that part of the bronchial wall shows a slight uplift, but other bronchoscopic examinations revealed bronchial stenosis.

In this case, the patient was successfully treated with local excision. To the best of our knowledge, all reported cases revealed that complete resection of tumor lesions, including surgical resections or endoscopic interventional therapies, provides cure [7]. Surgical treatment confers great trauma and high risk, requiring sophisticated technology, with slow recovery, high costs, potential complications, and high mortality. By contrast, endoscopic interventional therapy causes lesser trauma, has lower risk, takes shorter time, and is associated with fast recovery and less complications. If the lesion is small enough to be removed under endoscopy, the snare-and-burn technique is effective for the treatment of such lesions. Snare burn can obtain more complete specimens for pathological examination because the cancer often develops in the basal part of the tumor, and diagnosis of potential cancer may be missed when the specimen is inadequate. In addition, laser ablation should be avoided as much as possible because it can cause damage to the entire lesion tissue and make it difficult to obtain tissue specimens for pathological examinations, resulting in possible missed diagnosis. In some cases, endoscopic electrocautery with a snare or argon plasma coagulation is used. However, because of the possibility of carcinomatous change, endoscopic resection may lead to missed diagnosis of potential cancer. To maximize the retention of lung function, according to the location of the papilloma, the size of the tumor,

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Table 1. Summarize similar cases published in literature

1st author (year)	Location	Histologic features	Case number
Basheda S et al. (1991)	Bronchus	Squamous type	1
Popper HH et al. (1992)	Bronchus	Squamous type	5
Flieder DB et al. (1998)	Lung	Glandular type	7
Flieder DB et al. (1998)	Lung	Squamous type	27
Flieder DB et al. (1998)	Lung	Mixed type	7
Nakagawa M et al. (2008)	Lung	Glandular type	1
Tryfon S et al. (2012)	Bronchus	Glandular type	6
Tryfon S et al. (2012)	Bronchus	Squamous type	21
Tryfon S et al. (2012)	Bronchus	Mixed type	5
Kaseda K et al. (2014)	Lung	Glandular type	2

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and so on, tumor resection should be followed by selection of local bronchus, lung resection, and lobotomy [8]. We report a case of a tumor located in the main trachea, with solitary and small lesions, that was removed using a high-frequency snare, thereby achieving a good therapeutic efficacy.

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Disclosure of conflict of interest

None.

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