

Case Report

Case report: combination of multiple intervention methods under bronchoscope for tracheal adenoid cystic carcinoma

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Abstract: A 56-year-old male diagnosed with tracheal adenoid cystic carcinoma in 2009 was admitted to our department in February 2014 with complaints of progressively worsening breathlessness and wheezing. Computed tomography (CT) revealed a large tumor with near-complete obstruction of the distal trachea, and flexible bronchoscopy revealed a smooth, well-vascularized mass that almost completely obstructed the distal trachea. The patient was treated with high-frequency electrical snare, and about 2/3 of the foreign body was resected. After treatment, the patient's dyspnea was clearly relieved. From 2014 to 2017, the patient underwent bronchoscopic high-frequency electrical snare, argon plasma coagulation (APC), and CO₂ cryotherapy on several occasions due to local recurrence, and achieved good results. Surgical resection is the preferred treatment of adenoid cystic carcinoma. For patients who can't undergo surgical resection at the time of consultation and/or have serious complications, bronchoscopic intervention is a new treatment method.

Keywords: Adenoid cystic carcinoma, fibrobronchoscopy, tracheal neoplasmst

Introduction

Upper airway tumors are relatively rare and usually malignant [1]. Primary adenoid cystic carcinoma (ACC) is a low-grade malignant tumor that originates in the mucous gland or submucosal gland, and has an incidence of about 0.1/100,000 to 0.2/100,000 [2]. Tracheal adenoid cystic carcinoma (TACC) is a low-grade malignant tumor that originates in the trachea, and because it's early clinical symptoms lack specificity, it is easily missed or misdiagnosed.

At present, surgical resection is considered as the first choice treatment of ACC. Endoscopic options for managing malignant airway obstruction, such as use of lasers and stents, are palliative and reserved for unresectable tumors or patients with contraindications to surgery [1].

ACC characteristically shows infiltrative growth and perineural invasion, and tends to metastasize

size to the lungs, bones, liver and brain [3]. Once distant metastasis occurs, most patients die within 2 years [4]. A case of TACC was treated with a variety of bronchoscopic interventions in our department, and achieved good results; which is reported as follows.

Case report

A 56-year-old man presented with complaints of progressively worsening breathlessness and wheezing in our department in February 2014. He had a history of TACC from about five years prior that was treated with chemoradiotherapy. Physical examination: the patient was conscious without cyanosis on his lips, nor jugular vein bulging. The three concave signs of inhalation were visible, the thorax was symmetric without deformity, the bilateral breathing momentum and tremor were of the same degree, the breathing sounds on both lungs were thick, and the neck and both lungs had wheezing sounds. No obvious abnormality was

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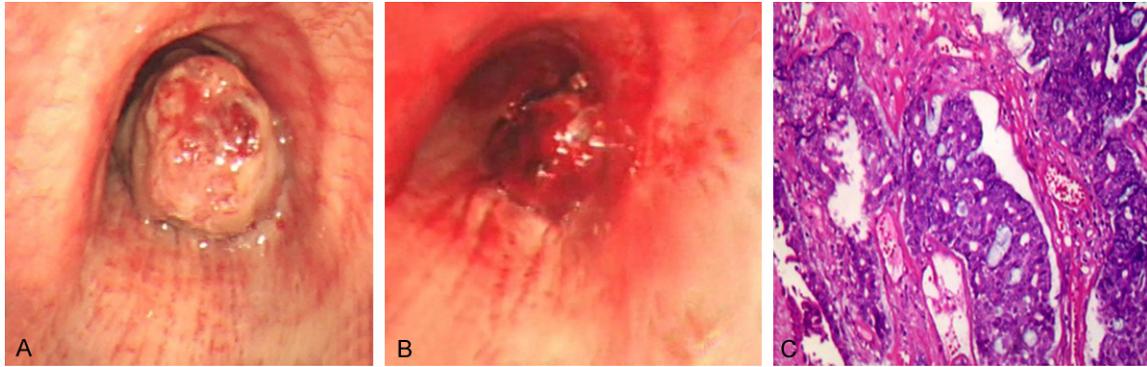


Figure 1. Bronchoscopic bronchopathy and the pathological examination of 2014. Before microscopic treatment (A): the endobronchial view seen on flexible bronchoscopy-vascularity of the new organism. The new organism is about 3 cm in length and blocks the trachea by about 90%. After microscopic treatment (B), the new organism is significantly smaller than before, and the bronchoscope can enter the left and right main bronchi and reveal unobstructed bronchial lumen of each section. (C) Photomicrographs of biopsy specimen (H&E, $\times 100$) adenoid cystic carcinoma with a cribriform and tubular growth pattern infiltrates beyond the level of the trachea. The tumor cells are composed of basal cells and myoepithelial cells.

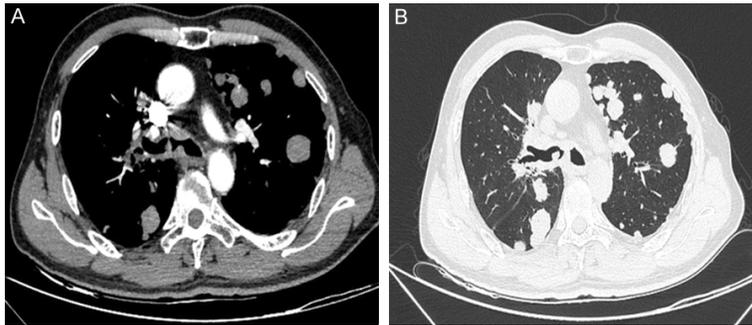


Figure 2. Chest CT scan: the walls of the lower part of general trachea, bulge area, and proximal part of the bilateral main bronchus are widely irregularly thickened and narrowed, which is obvious in the proximal section of the main bronchus with the lumen being significantly narrow with a minimum diameter of about 3 mm. There are multiple metastatic nodules and masses in both lungs and the pleural margins.

found in the examination of the heart and abdomen, and no edema was found in either lower limbs. CT scan showed that the lower part of the lumen of the trachea was occluded by more than 95%. Bronchoscopy showed a smooth, well-vascularized mass that almost completely obstructed the distal trachea. The length of the obstruction was about 3 cm and blocked the trachea by about 90% (**Figure 1A**). After received high-frequency electric snare treatment, about 2/3 of the growth was resected (**Figure 1B**), and the patient's dyspnea was significantly relieved. The histology of the biopsies was consistent with ACC (**Figure 1C**). From 2014 to 2017, the patient repeatedly underwent high-frequency electric snare, argon plas-

ma coagulation (APC), and CO₂ cryotherapy in our department due to local recurrence, and achieved good results. In June 2018, the patient was re-admitted to our unit due to shortness of breath and wheezing. Chest CT scan (**Figure 2**) revealed that the wall of the lower trachea, carina region and the proximal segment of bilateral main bronchus were extensively and irregularly thickened, and the lumen was narrowed. The lumen of the proximal segment of the right main bronchus was clearly narrowed,

with a minimum diameter of about 3 mm. There were multiple metastatic nodules and masses in both lungs and pleural margins. On June 6, rigid bronchoscopy was performed under general anesthesia. Under the microscope, most of the lumen was blocked by the new growth in the lower trachea, the left main bronchus opening showed changes of tumor infiltration, and the growth of new cauliflower-like organism in the right main bronchus caused stenosis of the lumen with the narrowest diameter as about 2-3 mm (**Figure 3**). Most of the new growth in the lower trachea was scooped out by rigid bronchoscope, and APC and CO₂ cryoablation was repeatedly performed for resecting the growth in the lower trachea and right main

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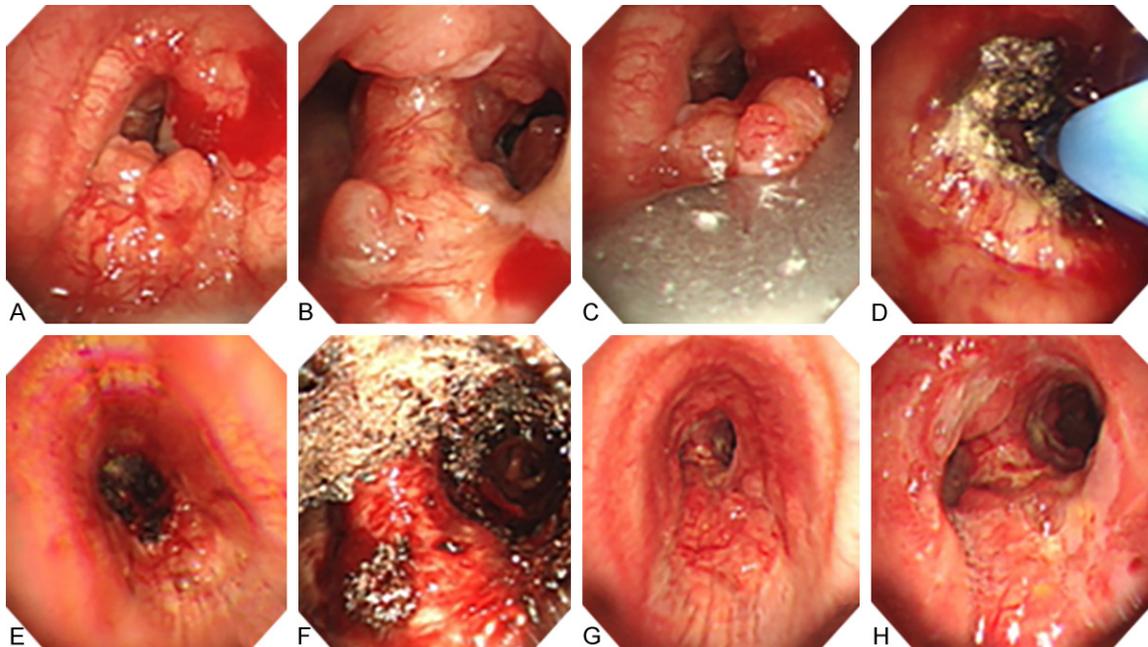


Figure 3. Bronchoscopic bronchopathy of 2018. (A, B) Before microscopic treatment: about 4 cm away from the bulge of the lower part of trachea, cauliflower-like growth and narrows the lumen; the bulge is widened, and the left main bronchus opening shows changes of tumor infiltration; the right main bronchus shows cauliflower-like new growth. Rotary incision (C) and CO₂ cryotherapy (D) are given under rigid bronchoscope; (E, F) after endoscopic treatment, the lumen diameter of the lower trachea was about 15 mm, the diameter of the right main bronchus was expanded to about 10 mm. One month later, the patient received bronchoscopy examination, which shows only a little growth in the lower trachea (G), the carina is widened, and the right main bronchial lumen is just slightly narrowed (H). The stenosis of the main bronchial lumen is significantly improved compare with that before treatment.

bronchus. After treatment, the lumen diameter of the lower trachea was about 15 mm, the diameter of the right main bronchus was expanded to about 10 mm (**Figure 3**). The symptoms of breathing shortness and dyspnea in the patient were significantly relieved. The patient was repeatedly treated with high-frequency electrical snare, APC, laser ablation, and CO₂ cryotherapy due to tumor recurrence in our department within 9 years after the diagnosis of TACC and achieved good results. However, the patient eventually died of distant metastasis and lung infection in May 2019. This study was conducted in accordance with the declaration of Helsinki. This study was conducted with approval from the Ethics Committee of Kunming Medical University. Written informed consent was obtained from all participants.

Discussion

Among the primary malignant tracheal tumors, mucoepidermoid carcinoma is the most common, followed by ACC, pleomorphic adenoma, acinar cell carcinoma, and epithelial-myoepi-

thelial carcinoma [5]. TACC is a low-grade malignant tumor that originates in the trachea and is mainly located in the main bronchus visible by bronchoscopy [6]. Symptoms are usually attributable to the intraluminal component of the tumor causing an obstruction of the airway, potentially leading to stridor, dyspnoea, wheezing, haemoptysis, and cough [7]. The diagnosis of TACC mainly depends on imaging, bronchoscopy and pathological tissue biopsy. Bronchoscopy can directly observe the size and morphology of the tumor. Bronchoscopy also provides the specific information for operative planning, as measurements can be taken to assess whether a tumor is resectable based on its location and size [1].

The preferred primary treatment of ACC is surgical resection with clear margins and tracheal reconstruction when possible [7]. When tumors extensively involve the airway, are combined with distant metastasis, or the patient's physical conditions can't tolerate surgery; radiation therapy and bronchoscopy are common palliative treatment methods. Bronchoscopic inter-

ventional therapy can effectively remove intratumoral tumors and temporarily/permanently relieve the airway obstruction caused by ACC [8]. The bronchoscopic therapies mainly include: endbronchial laser ablation, stent implantation, argon plasma coagulation (APC), CO₂ cryotherapy, rigid bronchoscopic recanalization and so on [9], which can effectively alleviate life-threatening airway obstruction. The patient in this study had life-threatening airway obstruction due to aggravated dyspnea at the time of his first visit to our department. Tumor growth involved a long part of the trachea and can't be treated with surgery, so bronchoscopic intraluminal interventional treatment was the preferred and effective option.

ACC has a good prognosis but is known for late local recurrence and metastasis for which long-term follow up is indicated [10]. Patients can survive with tumors for many years. When airway stenosis recurs after treatment, bronchoscopic intervention can be performed several times to ensure airway patency. The patient in our case was diagnosed with TACC in 2009. Since 2014, this patient was repeatedly treated with various interventional therapies under bronchoscope for airway obstruction caused by local tumor recurrence, all of which achieved good results. The patient survived with the tumor for nearly 10 years. Eventually, he died of distant metastasis and lung infection.

Conclusions

ACC is a rare and slow-growing malignant tracheal tumor, which is less aggressive than squamous cell carcinoma. Nonspecific clinical symptoms are often the reason for late diagnosis or misdiagnosis. The diagnosis of ACC mainly depends on imaging, bronchoscopy, and pathological tissue biopsy. Surgical resection is the first choice for early treatment of ACC. When the tumor is non-resectable, bronchoscopic intervention would be an effective and acceptable option for these kinds of patients to obtain a promising prognosis. ACC is known for late local recurrence and metastasis for which long-term follow up is recommended.

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Disclosure of conflict of interest

None.

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