Tracheal Adenoid Cystic Carcinoma Treated by Repeated Bronchoscopic Argon Plasma Coagulation as a Palliative Therapy

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Primary tracheal tumors are uncommon, making up only 0.2% of all respiratory malignancies. Adenoid cystic carcinoma (ACC) is the most common tumor, accounting for about 30% of primary tracheal tumors. It is often difficult to manage these tumors surgically, due to its expansion and submucosal invasion, and furthermore, due to the patient’s condition. Thus, it is essential to perform palliative treatment in order to maintain the airway through and to control the progress. Herein, we report a case of ACC treated by repeated bronchoscopic argon plasma coagulation (APC) as palliative therapy.

A 71-year-old Japanese male was referred to our hospital’s emergency department for dyspnea. Bronchoscopic examination revealed expanded intraluminal obstructive bronchial tumors from the left bronchus to right. The patient had undergone bronchoscopic APC treatment several times. The obstruction was improved, and no worsening was seen for 26 months. Repeated bronchoscopic APC as a palliative therapy is promising and useful therapy without heavy adverse reactions to control the tumor growth.

Keywords: adenoid cystic carcinoma (ACC), primary tracheal tumor, argon plasma coagulation (APC), obstructive pneumonia, palliative therapy

Introduction

Primary tracheal tumors are very uncommon and adenoid cystic carcinoma (ACC) is about 30% of tracheal tumors in worldwide.1 Due to its slow growth, ACC is rarely discovered until the late stage.2 Thus, it sometimes expands too wide to perform surgical resection. To remove the obstruction of the trachea, we often need palliative treatment to control the tumor. Argon plasma coagulation (APC) is considered to be a safer method to control the tumors than other methods, such as electrocautery or a neodymium-yttrium aluminum garnet (Nd-YAG) laser.3

Herein, we show a case of ACC treated by repeated bronchoscopic APC as a palliative therapy.

Case Report

A 71-year-old Japanese male was referred to our hospital’s emergency department, because of dyspnea. He had no history of lung disease though he had smoked half a pack of cigarettes for 10 years. Serum laboratory tests showed an elevated WBC (13,510/µL) and C-reactive protein (12.46 mg/dL). Chest x-ray (Fig. 1A) revealed left lower lobe pneumonia, and he was treated with ceftriaxone 2g-q 24 HR. The following chest computed tomography (CT) showed a left, lower consolidation and
tumors in the left bronchus, suggesting that it was obstructive pneumonia (Fig. 1B). The tumor derived from the left bronchus, expanding to the right (Fig. 1C). The pulmonary function test showed an obstructive pattern with plateau in the peak flow volume curve (FEV$_{1.0}$ 1.10 L, FEV$_{1.0%}$ 59.8%) (Fig. 1D). The follow bronchoscopy on the fifth day revealed a tumor with a telangiectatic surface protruding from the left bronchus to the right (Fig. 1E and 1F). Pathological analysis showed a small, glandular cribriform pattern and mucus-secreting cells (Fig. 1G). The following immunostaining was positive for actin, S100, and p63. These results, taken together, suggested that the tumor was ACC. Whole body CT, brain magnetic resonance imaging (MRI) and bone scintigraphy showed no metastatic lesions. There is a consensus that for ACC, surgical resection is preferable to other therapies. However, after consulting with surgeons, we decided that the surgical approach was too high-risk to be performed in this patient of advanced age because the tumor had expanded from the left to right bronchus, invading the trachea. For these considerations, we tried to control the tumor by bronchoscopic coagulation using hot biopsy and APC. We used a flexible bronchoscope (BF-1T, Olympus Corporation, Tokyo, Japan), ICC350 (ERBE Elektromedizin GmbH, Tubingen, Germany) as the device; APC300 (ERBE, Germany) with a flexible probe (ERBE, Germany). The starting power setting of 20W and an argon gas flow of 0.5 L/min were used. The first coagulation treatment was performed to improve the stenosis by the tumor, as we passed the bronchofiber through the left bronchus (spending time for therapy: for about 2.5 hours) (Fig. 2A). Five days later, a second treatment was performed to improve the stenosis in the left bronchus (for about 2 hours) (Fig. 2B). One month later, a third treatment was performed to expand the right bronchus (for about 1.5 hours) (Fig. 2C). After this treatment, we evaluated the effect of therapy with CT, indicating that the obstructive pneumonia of left lower lobe was cured and the tumor size had decreased (Fig. 3A). One month later, a fourth treatment (Fig. 2D) and, one month after that, a fifth treatment (Fig. 2E) were performed (each for about 1 hour). 3D-CT revealed further decreasing tumors and no stenosis of the bronchus (Fig. 3B). Three months later (8 months after the first admission), there was no stenosis in bronchus and only an abnormal appearance in the wall of the trachea and bronchus (Fig. 2F and 2G). He rejected additional chemotherapy and radiotherapy, thus he was only followed up by observation with CT every 4 months. There has been no worsening or increase in tumor size during the 26 months from the first admission (Fig. 3C). Bronchoscopy showed no worsening (Fig. 2H-2J). The pulmonary function test was normal (Fig. 3D).

**Discussion**

Primary tracheal tumors are uncommon, accounting for 0.2% of lung carcinomas; ACC is more common,
accounting for about 30% of tracheal tumors. ACC usually arises from salivary glands and can also arise from skin, breast, uterine cervix, upper aerodigestive tract, trachea, and lung. ACC is classified into 4 types via the type of expansion and invasion (Fig. 3E). Our case was classified Type III: expansive infiltrating type. The tumor in our case actually expanded from the left lower bronchus to the right.

Tumors located in the trachea or bronchus cause progressive airway obstruction and have a variable time course and nonspecific symptoms, because ACC is one of the low-grade malignant tumors. As a result, diagnosis is often delayed. The duration of symptoms alone predicts neither malignant potential nor resectability of
a tumor. The best way to cure patients with ACC is surgical treatment. The 5- and 10-year survival rate is 91% and 76% for resected, and 40% and 0% for unresected, respectively. The estimation of tumor size, length and depth influences the assessment of resectability. Although surgical treatment is firstly recommended to treat ACC, 25% of patients with ACC had unresectable disease. Tumor length in 68% of these patients was the most common reason for which resection was declined. The aim of treatment in unresectable malignant tumors is to maintain airway flow and to slow progression of disease. Effective local therapy, other than resection, may provide meaningful palliation, either by regional radiation or bronchoscopic treatment. In our case, we chose bronchoscopic APC treatment. During 26 months, no worsening had been detected.

APC was first used in open surgery for haemostasis in parenchymatous organs. The APC working principle is that high-frequency electrical current is fed from the tungsten electrode in the probe tip through the ionized and therefore electrically conductive, argon plasma without contact with the tissue. APC treatment is safer than the other treatments for several reasons: (1) low smoke and vapor production, (2) controllable depth of coagulation (maximum 3 mm to 4 mm), (3) effective and safe coagulation, especially of larger areas, with shallow but uniform coagulation depth, and (4) conical coagulation in an axial, as well as lateral direction. A disadvantage of APC is distention of mucosa due to argon gas insufflation. Major complications are emphysema and gas embolism.

In conclusion, as a palliative therapy, we treated ACC that obstructed bronchus and expanded bilateral bronchi, by repeated bronchoscopic APC. Although further studies on a larger scale are required to establish the indications and limitations in the palliative treatment of ACC, repeated bronchoscopic APC is a promising and useful therapy without heavy adverse reactions in the control of tumor growth.

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Disclosure Statement

None declared.

References