Removal of Glomus Tumor in the Lower Tracheal Segment with a Flexible Bronchoscope: Report of Two Cases

Yan Shang, Yi Huang, Hai-dong Huang, Chong Bai, Yu-chao Dong, Li-jun Zhao and Qiang Li

Abstract

Tracheal glomus tumor is an extremely rare neoplasm resected mostly by open surgery or through rigid bronchoscopy. We report two cases presenting with polypoid masses arising from the tracheal membrane in the posterior wall of the lower tracheal segment. The tumor was removed by high-frequency electrocautery and flexible bronchoscopic argon-plasma coagulation, and follow-up bronchoscopy and chest CT did not reveal tumor recurrence 12 months after the operation. In patients with tracheal glomus tumor who have poor surgical tolerance or are not willing to receive an open surgery, flexible bronchoscopic tumor removal can be a good alternative to relieve the airway obstruction symptoms.

Key words: glomus tumors, trachea, flexible bronchoscopy


Introduction

As a rare benign neoplasm occurring primarily in the extremities, a glomus tumor in the respiratory system is extremely rare. To date, only a few more than 20 cases of tracheal or bronchial glomus tumors (1) and 11 cases of pulmonary glomus tumors were reported (2). Glomus tumors in the respiratory system are removed mostly by open surgery or rigid bronchoscopy. In this report, we describe two cases of glomus tumors (1.8 and 2.0 cm in diameter respectively) presenting with polypoid masses arising from the tracheal membrane of the posterior wall in the lower tracheal segment and obstructing the airway. The patients underwent high-frequency electrocautery and flexible bronchoscopic argon-plasma coagulation (APC) for tumor removal, and were followed up for 12 months after the operation. Till now no signs of tumor recurrence have been found in these two patients by follow-up bronchoscopy or chest CT scanning.

Case Report

Case 1 A 59-year-old man patient was admitted in mid-
Figure 1. Chest X-ray films of Case 1 showing no abnormal findings.

Figure 2. Chest CT in Case 1 showing a neoplasm in the left wall of the lower tracheal segment, which partially obstructs the tracheal lumen. No organic lesion in the bilateral lungs or lymph node enlargement in the mediastinum is observed.

ameter on the left tracheal wall in the lower segment nearly obliterating the lumen; no other organic lesions in the bilateral lungs, or lymph node enlargement in the mediastinum were found (Fig. 2). Bronchoscopy revealed the presence of a neoplasm at the 5 to 9 o’clock position on the lower tracheal wall 2 cm from the tracheal carina. Mucosal hypereemia at the tracheal carina was found beyond the neoplasm, and the bronchial of the bilateral lobes remained normal (Fig. 3A).

To derive a definite diagnosis, bronchoscopic electrocautery and snaring of the neoplasm was performed in mid-September 2008, and mild bleeding occurred during the operation and 1 : 10,000 noradrenalin and 200 IU thrombin were administered intralumenally. A tissue specimen measuring 2 cm x1 cm x0.5 cm was obtained, which showed pinkish gray cross section and moderate hardness. Pathological examination revealed a lobular structure of the neoplasm with the tumor cells aligned around the blood vessels. The tumor cells were spherical or ovoid with distinct cell boundaries and rich cytoplasm stained in light pink. No nuclear atypia were detected. The interstitial tissue contained rich blood vessels. Immunohistochemistry yielded positive results for vimentin, smooth muscle actin (SMA), CALP, and CD99 (Fig. 4). A pathological diagnosis of glomus tumor was established.

Subsequent consultation with cardiothoracic surgeons suggested the presence of surgical indications for tumor removal in an open surgery, but the patient and his relatives failed to give their consent. Bronchoscopic tumor removal was therefore scheduled. In late-September, the patient underwent high-frequency electrocautery combined with argon-plasma coagulation through a flexible bronchoscopy, and basically total lesion clearance was achieved after the procedure (Fig. 3B). The patient was discharged in late September. Further follow-up was based on CT-scan and bronchoscopy. The airway remains patent without evidence of relapse twelve months after treatment (Fig. 3C).

Case 2 A 22-year-old woman patient was admitted in our department in mid-November 2008 due to episodes of coughing and hemoptysis for one year, and chest tightness and shortness of breath for over 3 months. One year before admission, the patient experienced episodes of coughing without identifiable causes and hemoptysis of about 20 mL fresh blood without blood clots, but no fever, chest pain or shortness of breath was reported. These symptoms were relieved after antibiotic treatment prescribed by a local hospital. The patient reported subsequent symptom relapse after catching a cold, with hemoptysis of about 20 mL blood,
Figure 3. A: Bronchoscopy in Case 1 showing a neoplasm in the 5 to 9 o’clock position in the left tracheal wall with the basal part measuring 2-2.5 cm in length and 2 cm in width. B: Immediately after high-frequency electrocautery and argon-plasma coagulation treatment. C: Twelve months after treatment.

Figure 4. A: Hematoxylin and Eosin staining of the tumor tissue in Case 1 (×40). The tumor cells are spherical or ovoid and contain rich cytoplasm, which is lightly stained. No nuclear atypia is seen. The cells are aligned around the blood vessels. Immunohistochemical staining of the tumor tissue in Case 1 is positive for SMA (B) and vimentin (C) (×100).

Figure 5. Chest CT in Case 2 showing a nodule in the posterior wall of the lower tracheal segment and obvious stenosis of the tracheal lumen without lymph node enlargement in the mediastinum.

which received no further medical attention. Three months before admission, the patient complained of chest tightness and shortness of breath after exercise, which progressively worsened with occasional night sweat but without obvious fever. About 1 month before admission the patient was unable to maintain a supine position, and a CT examination 10 days before admission suggested a neoplasm in the lower tracheal segment. The patient had body weight loss of 2.5 kg within 3 months.

Lung function test suggested severe impairment of the ventilation function, FVC 1.61 L, FEV1 0.56 L, FEV1/FVC 35%, FEF25-75 0.33 L/s, PEF 0.50 L/min, MVV 15 L/min, RV/TCL 28%, normal gas exchange. ECG recording demonstrated sinus rhythm and no abnormalities. The patient underwent CT scanning of the chest, which identified a nodule 1.8 cm in diameter located in the posterior wall of the lower tracheal segment (Fig. 5).

Bronchoscopy found a hard polypoid neoplasm, the surface of which was richly lined with capillaries. The base of the neoplasm resided on the left tracheal membrane and almost totally obstructed the trachea (Fig. 6A). High-frequency electrocautery (40 W) was performed to cut the base of the neoplasm, and a tissue specimen of 1.8 cm ×1.5 cm ×1.4 cm was removed to allow the passage of the bronchoscope (Fig. 6B). Beyond the neoplasm a sharp and moderately movable carina was seen, and the bronchial mucosa
in the bilateral lobes remained normal without lumen obstruction, bleeding, stenosis or other neoplasms. Pathological examination led to the diagnosis of tracheal glomus tumor. Six months and twelve months after resection the patient underwent CT-scan and bronchoscopy. No evidence of relapse was found (Fig. 6C).

Discussion

Glomus tumor, first reported in 1924 by Masson (3), is a benign tumor arising from the glomus body at the arteriovenous anastomosis. Consisting of specialized smooth muscle cells, the glomus body is associated with body temperature regulation (4) and characterized by cell embedment in homogeneous and basement membrane-like tissues, and the presence of a large quantity of pinocytotic vesicles along the cell membrane. The cytoplasm of the neoplastic cells contains myofilament bundles parallel to the longitudinal axis of the cells with macula densa. Glomus tumors accounted for 1.6% of the 500 soft tissue neoplasms identified so far (5), mostly benign in nature, and malignant glomus tumors are extremely rare.

Glomus tumors typically present with painful nodules in the skin, occurring primarily in tissues rich in glomus such as the nail bed and finger tips, but can also be found in the palps, wrists, arms, and feet. Their occurrences have been reported in the digestive system including the oral cavity, stomach, intestines and mesentery, or in the reproductive system such as the vagina and labia, or other locations such as the heart, mediastinum, kneecap, and soft tissues (6). Currently researchers have reached a consensus that glomus tumors can also arise in tissues containing minimal or virtually no glomus. Kim et al (7) reported their finding of structures similar to the glomus in the tracheal membrane adjacent to a glomus tumor, but serial tissue sections failed to confirm a definite correlation between these structures and the tumor.

According to Russell et al (8), the average age of patients with tracheal glomus tumors is 58 years (range 43 to 74 years), with a man to woman ratio of 4 : 1. Most of the cases are characterized by a long disease history with slow progression of the tumor, and have such clinical symptoms as gasping, dyspnea, irritating cough, hemoptysis, chest pain, and hoarse voice, among which gasping is the most common in close relation to the body position. The symptoms show poor response to antibiotics, corticosteroids or bronchodilators, and often result in misdiagnoses as bronchial asthma or chronic bronchitis. In physical examinations, the patients may exhibit inspiratory dyspnea and triple depression sign, and inspiratory wheezing can be heard above the sternal midline or the clavicle. Chest X-ray often has negative result. Chest CT scanning and bronchoscopic examination provide important clues for an accurate diagnosis of tracheal glomus tumor and display the location and volume of the tumor. The tumors can be found as polypoid masses in the tracheal membrane of the posterior tracheal wall, growing by protruding to the lumen. The tumor masses may arise between 1 cm below the glottis and the tracheal carina, but commonly in the lower 1/3 segment of the trachea, probably because of abundant glands and blood vessels in this area. The diameter of the tumor mass ranges from 1.2 to 4.5 cm (mean 2.0 cm). The tumors can be completely encapsulated with a smooth surface and rich blood supply, and seldom give rise to local deep layer invasion or distal metastasis. By appearance tracheal glomus tumors are difficult to differentiate from other airway benign tumors, and a definite diagnosis must be established by a pathological examination.

Pathologically, glomus tumors with varying diameters consist of rich blood vessels surrounded by clusters of spherical or polygonal cells with homogeneous sizes and well defined cell boundaries. The neoplastic cells often do not contain rich cytoplasm, which is either eosinophilic or transparent. The cell nuclei are round and regular, and the nucleoli can be obscure without nuclear atypia or nuclear divisions. Immunohistochemically, almost all the neoplastic cells are positive for vimentin and SMA (3), and the endovascular cell markers are found almost exclusively in the
vascular endothelium. As with benign tumor with rare recurrence after removal, tracheal glomus tumor is associated with a favorable prognosis. Due to the potential risk of outgrowth of the tumor in the trachea (9), local surgery for tumor resection is the primary choice of treatment (10). Bronchoscopic treatment such as by laser ablation, argon plasma coagulation and stent placement (11) can effectively relieve the airway obstruction and facilitate later surgical tumor removal. So far only a few cases have been reported concerning the exclusive use of flexible bronchoscopic treatment for tracheal glomus tumor (12, 13), and its safety and efficacy still await further evaluation. The two patients reported herein refused conventional surgical resection of the tumor, and subsequently underwent high-frequency electrocautery and bronchoscopic argon-plasma coagulation. As glomus tumors are mostly polypoid neoplasms with a complete capsule and a narrow base, their surgical removal was uneventful. At the time of this writing the two patients have been closely followed up for 12 months, and bronchoscopy and chest CT did not detect signs of tumor recurrence. We therefore presume that flexible bronchoscopic removal can be one of the good options for relieving airway obstruction and to promote the quality of life in tracheal or bronchus glomus tumor patients who cannot or do not wish to undergo conventional surgical tumor resection.

Acknowledgement
We are grateful to Professor Yong-Wei Yu in the Department of Pathology, Shanghai Hospital for kindly providing pathology images.

References


© 2010 The Japanese Society of Internal Medicine
http://www.naika.or.jp/imindex.html