Benign Myoepithelioma in the Intrathoracic Trachea

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Abstract

A 67-year-old woman who was followed as a patient with bronchial asthma for 1.5 years visited our hospital with progressive dyspnea. Although the chest radiography findings were normal, a chest computed tomography scan revealed a mass obliterating the intrathoracic tracheal lumen. The patient’s symptoms disappeared immediately after tumor excision, and no recurrence was observed during a 1.5-year follow-up period. Microscopically, the tumor was composed of densely packed polygonal-, oval- and spindle-shaped cells that were positive for pan-cytokeratin, α-smooth muscle actin and p63. These pathological findings confirmed the diagnosis of benign myoepithelioma. Chest physicians should recognize that benign myoepithelioma can develop in the trachea, although it is very rare.

Key words: myoepithelioma, benign, trachea

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Introduction

Myoepithelioma accounts for only 1-1.5% of all salivary gland tumors (1, 2). Although the tracheobronchial submucosal glands are considered to be counterparts of the salivary glands (3), myoepithelioma of the respiratory system is extremely rare, and only two cases of tracheal myoepithelioma have been reported (4). We herein report a case of intrathoracic tracheal myoepithelioma that was followed as bronchial asthma for 1.5 years.

Case Report

A 67-year-old woman presented with a dry cough in August 2010. She was a never-smoker with a history of appendicitis and had been diagnosed with bronchial asthma. Treatment with anti-asthmatic drugs, including a bronchodilator and steroid inhaler, was initiated; however, it did not produce the desired effect.

The patient developed dyspnea in December 2011 and visited our hospital in January 2012. Upon a physical examination, inspiratory and expiratory wheezing was detected. Although a chest radiography showed only overinflated lungs, a chest computed tomography revealed a mass in the intrathoracic trachea (Fig. 1A, B). Pulmonary function tests demonstrated a normal forced vital capacity (FVC) of 2.62 L (111% of predicted), a normal forced expiratory volume in 1 second (FEV1) of 1.74 L (96.2% of predicted) and a FEV1 to FVC ratio of 71.6%. The peak expiratory flow rate (PEFR) decreased to 2.66 L (47.7% of predicted). The flow-volume curve was characteristic for central airway obstruction, with a plateau in both the inspiratory and expiratory limbs (Fig. 2A). Emergent flexible bronchoscopy performed under local anesthesia revealed a solid, movable tumor with a small pedunculated base attached to the lateral wall that obliterated 80% of the tracheal lumen (Fig. 1C).

The entire mass was removed using a snare followed by cautery. The patient’s symptoms, including coughing, dyspnea and wheezing, disappeared immediately after tumor removal. A pulmonary function test showed that the PEFR had increased up to 5.32 L (95.3% of predicted) and the
plateauing of the flow-volume curve had disappeared (Fig. 2B). As of August 2013, no local recurrence had been observed.

A macroscopic examination of the specimen revealed a homogeneous 20 mm × 18 mm × 12 mm yellowish-white polypoid mass (Fig. 3A). The tumor appeared to be well circumscribed with a thin fibrous capsule (Fig. 3B). Microscopically, the lesion was composed of densely packed polygonal-, oval- and spindle-shaped cells, with occasional eosinophilic collagenous crystalloids (Fig. 3C). No vascular or lymphatic invasion was found. In addition, the lack of mitosis and necrosis and the low (4%) proliferative activity, measured using Ki-67 antigen immunoreactivity, confirmed that the tumor was benign. Immunohistochemistry showed the tumor to be positive for the epithelial cell marker pan-cytokeratin (Fig. 3D), the myoepithelial cell marker α-smooth muscle actin (Fig. 3E) and the basal cell marker p63 (Fig. 3F). The tumor was also positive for S-100 protein, glial fibrillary acidic protein and CD10 (data not shown). Based on the immunostaining findings and morphology of the myoepithelial cells, the patient was diagnosed with benign myoepithelioma.

**Figure 1.** (A) Chest radiography shows overinflated lungs. (B) Chest computed tomography (CT) demonstrates a tumor located in the distal trachea. (C) A fiberoptic bronchoscopy shows a polypoid tumor in the lower trachea that occludes the tracheal lumen.

**Figure 2.** (A) Flow-volume loop reveals flattening of the inspiratory and expiratory limbs. (B) Flattening of the inspiratory and expiratory limbs disappeared after removing the tumor.
Discussion

In this report, we described a case of an intrathoracic tracheal tumor obliterating the tracheal lumen. The tumor morphology and immunohistochemical testing showed that the tumor was a benign myoepithelioma. Currently, myoepithelioma is a vague and confusing term used to describe a pulmonary lesion of myoepithelial origin, particularly in the respiratory system (5). Because some myoepitheliomas have been shown to be malignant (6, 7), the use of the term “benign myoepithelioma” is recommended to prevent confusion between benign and malignant myoepithelial lesions (2, 4, 5). To the best of our knowledge, there have been only four reported cases of benign myoepithelioma located in the peripheral lungs (5, 8-10) and two cases in the bronchus (2, 11). Furthermore, only two cases of benign myoepithelioma in the trachea have been reported. The tumors were detected while investigating productive coughing (4) and neck mass (12, 13) and were successfully resected without recurrence. However, the follow-up periods after resection were only eight months (12, 13) and not provided (4) and thus were not long enough to confirm the benignity of the tumors. Our patient exhibited a good clinical course for 1.5 years after undergoing resection, although it took 1.5 years to diagnose the myoepithelioma. Both the pathological findings and clinical course of the present case confirm the benign nature of the tumor.

In conclusion, we herein reported a case of tracheal benign myoepithelioma that was diagnosed and followed as bronchial asthma. Chest physicians should recognize that benign myoepithelioma can develop in the trachea, although it is very rare.

The authors state that they have no Conflict of Interest (COI).

References

4. Chand M, Mann JM, Sabayev V, et al. Endotracheal myoepithe-

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http://www.naika.or.jp/imonline/index.html