CASE REPORT

Pleomorphic Adenoma with an Endobronchial Resection

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Abstract

A case of bronchial pleomorphic adenoma is herein presented. The patient came to the hospital for a detailed examination of a bronchial polyp that was detected by computed tomography. Chest computed tomography revealed a bronchial tumor which was located at the distal end of the left main bronchus. The patient refused surgical resection. An electrosurgical snare was performed two times and the patient received several rounds of argon plasma coagulation with a flexible bronchoscope. The diagnosis of a pleomorphic adenoma was made following examination of the resected specimens. No recurrence has been observed by biopsy at the resected site.

Key words: pleomorphic adenoma, bronchial tumor, endobronchial resection, electrosurgical snare, flexible bronchoscope


Introduction

Although pleomorphic adenoma (also called mixed tumor) is the most common type of tumor in the salivary glands, a tracheobronchial pleomorphic adenoma is an extremely rare airway tumor (1). Tumor resection and airway reconstruction is considered to be the best choice for the management of a tracheobronchial pleomorphic adenoma. However, not all patients can undergo surgery due to medical and/or personal reasons. Recently the effectiveness and safety of a flexible bronchoscope for endobronchial therapy by has been widely accepted. Therefore, interventional pulmonology has become an available therapeutic choice in many institutes.

An electrosurgical snare and argon plasma coagulation (APC) has been widely used and accepted for the management of benign and malignant airway stenosis. This report describes a successful endobronchial resection using an electrosurgical snare and APC in a patient with a bronchial pleomorphic adenoma who refused a surgical resection.

Case Report

A 71-year-old man came to the hospital to undergo a detailed medical examination of a bronchial polyp that had been detected by chest computed tomography (CT). He had no respiratory symptoms. He did not smoke and had no history of salivary gland tumors. Multi-planar reconstruction of the CT revealed an endobronchial tumor at the distal end of the left main bronchus (Fig. 1). Subsequent flexible bronchoscopy revealed that the tumor had a relatively wide sessile base and grew from the anterior wall of the left main bronchus (Fig. 2A). The surface of the tumor was lustrous and engorged branching vessels were observed by bronchoscopy (Fig. 2A). Because the patient refused a surgical tumor resection, an electrosurgical snare using a flexible bronchoscope was employed to remove the tumor. The endobronchial electrosurgical snare (and APC) procedure was performed using a flexible bronchoscope. Local anesthesia and conscious sedation with intramuscular injection of pethidine hydrochloride and atropine was administered be-

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before the procedure. Topical anesthesia was achieved by the endobronchial instillation of 2% lidocaine. The pulse oximetry saturation, resting pulse and electrocardiograms were monitored during bronchoscopy. The electrosurgical snare was performed using an ICC200 (Erbe Elektromedizin; Tuebingen, Germany). To remove the tumor, a surgical snare (SD-7P-1, Olympus, Tokyo) was placed into the working channel of the flexible bronchoscope (BF IT-260, Olympus). Two endobronchial resections were performed using the electrosurgical snare and the resected tumor was removed by the grasping forceps (FG-42 L-1, Olympus). Most of the tumor was resected by this procedure. Furthermore, several short 1-2 seconds bursts of APC were administered at the sites of the resection to provide hemostasis and further coagulation for the residual tumor. A flexible-monopolar Teflon tube with a 1.5-mm diameter, 150-cm length fitted into the working channel of the flexible bronchoscope was used to administer the APC.

The lesion was diagnosed to be a pleomorphic adenoma based on the examination of the pathological specimen (Fig. 3A). The tumor was composed of glandular structures that consisted of squamous to cuboidal cells in one portion and spindle or stellate shaped cells in a hyaline or myxoid stroma in another portion. Immunohistochemical staining with polyclonal anti-S-100 protein and anti-glial fibrillar acidic protein was positive (Fig. 3B, C). The resected margin was diagnosed to be positive, so the APC procedure was administered two more times (once per month) in order to coagulate the residual tumor. No oxygen supply was used during the bronchoscopy. Surveillance bronchoscopy six months after the tumor resection showed that the resected site and bronchus remained patent (Fig. 2B) and no recurrence or metastasis was detected by chest CT.

**Discussion**

This report describes the use of an electrosurgical snare and APC to perform an endobronchial resection of a bronchial pleomorphic adenoma. These endobronchial therapeutic modalities provided the safe restoration of airway patency in a patient with a bronchial tumor who refused a surgical resection.

Tracheal tumors in general are uncommon, accounting for only 0.2% of all respiratory malignancies in the United States (2). In Japan, the frequency should be equivalent; however, at present the available data are still insufficient. Gaissert and Mark reported 164 cases of salivary gland tumors of tracheobronchial origin during a 34-year period from 1972 to 1995: 137 of them (83.5%) were adenoid cystic carcinomas, 21 (12.8%) were mucoepidermoid carcinomas, and only three (1.8%) were pleomorphic adenomas (1). In 2007, Aribas et al (3) reported that only 33 cases (range 26-71years) of tracheobronchial pleomorphic adenoma were reported in the literature, and two additional 8- and 15-year-old cases were found in the current study (4, 5). Including the present case and Japanese language case reports, up to 50 cases have presently been reported (3-11).

Recently endobronchial intervention using a rigid and flexible bronchoscope is widely performed in many countries. An evaluation of ‘interventional pulmonology’ tech-
Figure 3. A: The endobronchial resected specimen revealed both epithelial and stromal elements. Hematoxylin and Eosin staining (original magnification 25×). B, C: Immunohistochemical staining demonstrated positive reactions with polyclonal anti-S-100 protein (B) and anti-glial fibrillary acidic protein (C).

Technical skills have been presented by the European Respiratory Society and the American Thoracic Society (12). The combination of an electrosurgical snare and APC is one of the techniques of interventional pulmonology. The immediate effect is now widely accepted, and the present study demonstrated the effectiveness and safety of these procedures (13-15). The application of an electrosurgical snare has been used for many decades in the field of gastroenterology, however, over the past two decades it has also gained popularity for use in interventional pulmonology (16). In addition, these devices have been further improved in Japan for easier and safer use when performing bronchoscope including multicenter studies (17, 18). In a multicenter study that was performed in Japan (18), the safety phase of this modality was also discussed. The documented adverse effects included respiratory failure, massive bleeding, pneumonia, airway burning, perforation, fistula, pneumothorax, air embolism, and cerebral bleeding due to increased blood pressure, etc. In one reported case (18), hemostasis could not be achieved despite using APC after a tumor resection using an electrosurgical snare. At present, the use of the electrosurgical snare to treat endobronchial tumors can be safely performed at many institutes.

Although endobronchial intervention with a flexible bronchoscope has been previously performed without intubation at our institution, based on the findings of these reports, a tumor resection performed under intubation, especially for large tumors such as the one observed in our case, may thus have been necessary.

There are also several reports of pleomorphic adenoma that have been resected using a bronchoscope with an Nd-YAG laser (9-11). A surgical resection is not used when the patient is in poor condition (9), refuses surgery (10) or is of advanced age (11). The longest follow-up period is 5 years (11) and no recurrence or metastasis has been reported. The current patient also refused surgical resection and no recurrence and metastasis was seen in his clinical course (6 months). Additive coagulation was administered for the treatment of the residual tumor by APC and the patient did not experience any serious complications during the treatment courses. Recently, in gastroenterology, the adjunct use of APC after polyp resection has been reported to be safe and efficacious (19). This combination therapy appeared to be effective in the present case.

The optimal surveillance period for performing bronchoscopy after a benign tumor resection has not yet been determined. In the case of benign tracheobronchial tumor, strict survey might not be needed, especially when the resected margin is negative, however, if the resected margin is positive with or without additional APC, then we believe that surveillance biopsy is necessary, at least within six months regardless of the luminal surface appearance.

Because of the rarity of this type of tumor, the optimal therapeutic choice is unknown. However, previous reports indicate that surgical tumor resection and airway anastomosis seems to be the best therapeutic choice (1, 3-8). Furthermore, there are two reports of malignant pleomorphic adenomas (7, 8). Many of the reports (3-6) regarding pleomorphic adenoma stated that surgical resection is the best therapeutic choice for this disease based on those previous reports (7, 8). However, one of the studies addressing malignant pleomorphic adenoma (8) reported that metastasis occurred 11 years after the complete resection of the primary tumor. This indicates that a complete resection is not always a safe modality for patients with this disease. Therefore, endobronchial therapy is an effective therapeutic choice, especially in patients with a poor condition, advanced age or refusal of surgery.

In summary, an endobronchial tumor resection was successfully performed in a case of bronchial pleomorphic adenoma. Although surgical resection seems to be the first choice for the management of this disease, endobronchial therapy is an effective alternative procedure for tumor resection at least in selected patients who are inoperable due to either medical or personal reasons.

References

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