A Salivary Gland-Type Monomorphic Adenoma with Trabecular Proliferation in the Lung

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Salivary gland-type adenoma, especially monomorphic adenoma in the lung is very rare. We report a 66-year-old previously healthy woman who developed cough, hemoptysis, and fever. Bronchoscopy revealed an endobronchial mass in the proximal portion of the left lower lobe bronchus. Following left lower lobectomy, the pathological diagnosis was an unusual monomorphic adenoma of the salivary gland-type with trabecular proliferation.

Key words: endobronchial adenoma, salivary gland tumors

Introduction

Endobronchial tumors form a heterogeneous group accounting for between 1 and 10% of all primary lung tumors (1, 2). Carcinoids account for 90% of these tumors (1, 2). The remainder consists of various tumors of salivary gland-type tumors, including adenoid cystic carcinomas, mucoepidermoid carcinoma, and truly benign adenomas of the bronchial gland (1). About two-thirds of the salivary gland-type tumors are adenoid cystic carcinomas and most of the remainder are mucoepidermoid carcinomas (2). Therefore, truly benign bronchial gland adenomas are very rare. Tumors bearing the features of so-called mixed tumors (pleomorphic adenoma) are rarely recognized in the lung, despite being the most common histologic form of salivary gland neoplasm (3). However, some examples of this type of lesion have been described (4). Although pulmonary oncocytoma and epithelial-myoepithelial tumors are other rare variants (3, 5), monomorphic adenoma is even more rare compared to pleomorphic adenoma. We describe a 66-year-old woman with a monomorphic adenoma of the salivary gland type showing a trabecular pattern who developed cough, hemoptysis, and fever due to airway obstruction of the left lower lobe bronchus. To our knowledge, no previous report of this type of monomorphic adenoma of the salivary gland-type has been reported.

Case Report

A 66-year-old female nonsmoker presenting cough, fever, and hemoptysis visited her local physician. There was no remarkable past history or family history. A chest X-ray film revealed infiltrates on the left lower lobe. Bronchoscopy showed an endobronchial mass in the left lower lobe bronchus. She was referred to Jichi Medical School Hospital for further evaluation and therapy. The physical examination showed only fine crackles at the left lower lung field. Laboratory studies revealed the following: leukocyte count, 3.4x10^9/l; C-reactive protein, 0.029 mg/dl; erythrocyte sedimentation rate, 13 mm in the first hour; normal results of hepatic and renal function test. Arterial blood gas analysis on room air disclosed a pH of 7.414, PaO_2 of 80.4 mmHg, and PaCO_2 of 43.8 mmHg. Pulmonary function testing revealed VC of 123%, FEV_1 of 67.5%, and DLCO of 67.5% showing a slight obstructive respiratory dysfunction and a slightly decreased diffusing capacity. There was no elevation of tumor markers including squamous cell carcinoma antigen, carcinoembryonic antigen, and neuron-specific enolase.

A thoracic computed tomography showed a round tumor which occupied most of the intralumen of the proximal portion of the left lower lobe bronchus (Fig. 1). Bronchoscopy revealed reddish, smooth, regular mass with stem originating at the second carina (Fig. 2). No tumor tissue was obtained by transbronchial biopsies. Polypectomy was also unsuccessful. A left lower lobectomy with lymphadenectomy was performed, since the possibility of carcinoid tumor was suggested by the frozen section biopsy during operation. Postoperative examination revealed trabecular or cord-like proliferation of monomorphic oval cells in a hyaline-vascular stroma (Fig. 3).
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Figure 1. Thoracic computed tomography shows the round tumor which occupied most of the intralumen of the proximal portion of the left lower lobe bronchus.

Figure 2. Bronchoscopic view of the second carina revealing a reddish, smooth, regular mass with stem. No cribriform pattern, alveolar pattern, tubular formation or cornification was noted. The tumor cells had scanty, mucin-negative cytoplasm. No chondromatous area was observed. Immunohistochemically, the tumor cells were positive for cytokeratin and epithelial membrane antigen, and negative for chromogranin, neuron-specific antigen, S-100 protein, and HHF-35 (smooth muscle actin). According to the microscopic appearance and the immunohistochemical features of the tumor cells, a diagnosis of monomorphic adenoma of the salivary gland type with trabecular proliferation was obtained. No lymph node metastasis was found. The patient had an uneventful postoperative course and was performing normal activities, with no roentgenographic abnormalities.

Discussion

The term “bronchial adenoma” was introduced by Kramer (6) in 1930 to differentiate a group of primary endobronchial tumors characterized by slow growth, low grade malignant potential and long-term host survival. Pathologically, “bronchial adenomas” are either bronchial carcinoids or salivary gland-type tumors (1).

Salivary gland tumors are subdivided into adenoid cystic carcinomas, mucoepidermoid carcinoma, acinar cell tumor, polymorphous adenocarcinoma of low degree, mixed tumor, oncocytoma, and several types of monomorphic adenomas (1, 5).

Truly benign bronchial gland adenomas are very rare. These tumors are divided into pleomorphic adenomas and monomorphic adenomas. Pleomorphic adenomas are the most common tumors of the major salivary glands, accounting for between 70 and 80% of parotid, and between 60 and 70% of submandibular neoplasms (7). Pleomorphic adenomas in the bronchial gland are very rare, however, some examples of this type of lesion have been described (3, 4). Monomorphic adenomas are rare, accounting for only 1 or 2% of all parotid tumors (7). Adenolymphoma is the commonest, accounting for over 70% of all monomorphic adenomas originating at the salivary glands. The oncocytoma (oxyphilic adenoma) is another distinctive type of tumor. The rest of the monomorphic adenomas are grouped together as ‘other types’. Although oncocytoma in the lung have only rarely been reported, other types of monomorphic adenomas seem to be much rarer than pleomorphic adenomas in the lung (4, 8).

In the present case, the tumor cells were monomorphic and showed trabecular pattern. The histopathologic features of the
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The present case differed from adenoid cystic carcinoma, mucoepidermoid carcinoma, pleomorphic adenoma and acinic cell tumor. Carcinoid tumor was also denied by the absence of Grimelius-positive cytoplasmic granule and negative immunoreactivity for chromogranin and neuron-specific enolase. Myoepithelial differentiation of the tumor cell was not identified, since tumor cells were S-100 and HHF-35 negative. The histological characteristics of the present case, i.e., trabecular or cord-like proliferation of monomorphic oval epithelial cells, closely resembled those of trabecular subtype of basal cell adenoma of the salivary gland (9). However, the ultrastructural characteristic of basal cell adenoma has not been obtained. According to the interpretation of the microscopical appearance and immunohistochemical evidence, a diagnosis of monomorphic adenoma of the salivary gland with trabecular proliferation was obtained. The postoperative course was benign, however, further observation is necessary.

References