INTRODUCTION

Polymorphous low-grade adenocarcinoma (PLGA) is a low-grade malignant infiltrative tumor of the minor salivary gland. The term *polymorphous low-grade adenocarcinoma of the minor salivary glands* was coined in 1984 by Evans and Batsakis (1), to reflect this tumor's morphologic appearance and clinical behavior. PLGAs are thought to be indolent tumors that are preferentially localized to the palate, and they affect the minor salivary glands almost exclusively. Until now, two cases of distant metastases to the lung have been reported in the English literature. To the best of our knowledge, only one case of PLGA of minor salivary gland-type of the lung without evidence of a previous oropharyngeal primary tumor has been reported in the English literature. But the case was not a single lesion; it was bilateral tumors accompanied by tumors of the cervical lymph nodes. We report here the first case of a single primary PLGA of the minor salivary gland-type of the lung, which was successfully treated by sleeve bronchial resection of right upper lobe.

CASE REPORT

A 66-yr-old woman was admitted to the department of pulmonology due to her chronic and recently aggravated dyspnea. She had been treated for three years at another hospital under the diagnosis of asthma. The patient was referred to the department of pulmonology at our hospital. The finding of collapse and consolidation of the anterior segment of the right upper lobe was noted on her chest radiography (Fig. 1A). The bronchoscopic examination revealed a cystic fungating mass that nearly obstructing the inlet of the right main bronchus (Fig. 1B). A portion of the mass was hyperemic, and the biopsy from the periphery of the mass revealed a few atypical spindle cell proliferations in the stroma. The tentative diagnosis did not exclude the possibility of a spindle cell neoplasm. The patient was referred to the department of thoracic and cardiovascular surgery for surgical intervention. Preoperative chest CT scanning revealed about a 1.8-cm sized slightly lobulated and mildly heterogeneously enhancing endobronchial mass at the right main bronchus and the upper lobar bronchus with resultant postobstructive pneumonia in

---

**Key Words** : PLGA; Lung; Primary Tumor; Sleeve Lobectomy
the right upper lobe and right middle lobe. The tracheobronchial lymph nodes and the subcarinal nodes were also noted to be enlarged. The patient was in the high risk group for pneumonectomy and the parameters of the lung function test were as follows; forced vital capacity (FVC) of 1.04 L and 47% of the predicted value, forced expired volume in 1 second (FEV₁) of 0.76 L and 48% of the predicted value, residual volume (RV) of 2.49 L and 177% of predicted value, and diffusing capacity for carbon monoxide (DLCO) of 7.1 mL/mmHg/min and 51% of the predicted value. The operation of sleeve right upper lobectomy and mediastinal lymph node dissection was performed. The mass was found to have originated from the right upper lobar bronchial inlet, and it was about 1.0 × 0.7 cm in size upon fixation with formalin. The mass was lobulated, and the cut surface of the tumor was soft.
Primary PLGA of Minor Salivary Gland-type of the Lung

375

Afflicted PLGA of Minor Salivary Gland-type of the Lung often after a considerable period of years since the diagnosis undergone differentiation to a high-grade carcinoma, and there are also a few reported cases where the tumor has that are malignant (7). Despite its bland morphologic appearance, there are 200 cases of this tumor have been reported. PLGA comprises described by Evans and Batsakis in 1984 (1). Since its origi-

nal description and acceptance as a separate entity, more than 200 cases of this tumor have been reported. PLGA comprises 7.4% of the minor salivary gland tumors and 19.6% of those that are malignant (7). Despite its bland morphologic appearance, there are also a few reported cases where the tumor has undergone differentiation to a high-grade carcinoma, and often after a considerable period of years since the diagnosis.
aged by bilateral wedge resection of the lung. The patient underwent postoperative clinical examination including the laryngoscopy, CAT scans of the head, neck, chest and abdomen, and a whole-body PET scan, which did not reveal any primary lesion. However, the authors discussed a possibility that those lung tumors might have represented metastasis of unknown origin rather than primary lung tumors. In our case, the pathological features were typical of PLGA, where the tumor cells displayed a variety of growth patterns and they had a bland cytologic appearance, which was similar to its counterpart in the minor salivary gland. The diagnosis of PLGA was based on both the typical morphologic appearance and the immunohistochemical findings. There was no evidence of primary tumor outside the chest on the detailed head and neck examination including the laryngoscopy, chest CT scanning, and whole body PET-scanning.

The specific operative technique of bronchial sleeve resection was first performed by Price Thomas (10). It avoids the need for pneumonectomy and it spares the healthy lung tissue distal to the pathologic lesion. Benign or malignant tumors confined to the large bronchus can be successfully managed by sleeve lobectomy. Because the surgical risk is not higher than that for pneumonectomy, and a significantly better postoperative quality of life and physical activity is ensured by the preservation of the functioning and intact lung tissue, we regard this type of tumor with its low-grade malignant potential as being well suited for sleeve bronchial resection.

In conclusion, PLGA is a rare minor salivary gland-type tumor that has been thought to occur almost exclusively in the minor salivary glands in the oral cavity and the neck. However, PLGA should be considered in the differential diagnosis of patients presenting with low-grade gland-forming tumors of the lung. When this tumor originates from the large bronchus, it can be safely resected by the technique of bronchial sleeve resection.

REFERENCE

1. Evans HL, Batsakis JG. Polymorphous low-grade adenocarcinoma of minor salivary glands. A study of 14 cases of a distinctive neoplasm. Cancer 1984; 53: 935-42.
2. Vincent SD, Hammond HL, Finkelstein MW. Clinical and therapeutic features of polymorphous low-grade adenocarcinoma. Oral Surg Oral Med Oral Pathol 1994; 77: 41-7.
3. Gnepp DR, Chen JC, Warren C. Polymorphous low-grade adenocarcinoma of minor salivary gland. An immunohistochemical and clinicopathologic study. Am J Surg Pathol 1988; 12: 461-8.
4. Tanaka F, Wada H, Inui K, Mizuno H, Ike O, Yokomise H, Fukuse T, Hitomi S, Shoji K, Nakashima Y. Pulmonary metastasis of polymorphous low-grade adenocarcinoma of the minor salivary gland. Thorac Cardiovasc Surg 1995; 43: 178-80.
5. Hannen EJ, Bulten J, Festen J, Wienk SM, de Wilde PC. Polymorphous low grade adenocarcinoma with distant metastases and deletions on chromosome 6q23-qter and 11q23-qter: a case report. J Clin Pathol 2000; 53: 942-5.
6. Lee VK, McCaughan BC, Scolyer RA. Polymorphous low-grade adenocarcinoma in the lung: a case report. Int J Surg Pathol 2004; 12: 287-94.
7. Ellis LG, Auclair Paul L. Tumors of the Salivary Glands. In atlas of tumor pathology. Third series. Fascicle no.17. Armed Forces Institute of Pathology, Washington, DC. 1996
8. Simpson RH, Reis-Filho JS, Pereira EM, Ribeiro AC, Abdulkadir A. Polymorphous low-grade adenocarcinoma of the salivary glands with transformation to high-grade carcinoma. Histopathology 2002; 41: 250-9.
9. Castle JT, Thompson LD, Frommelt RA, Wenig BM, Kessler HP. Polymorphous low-grade adenocarcinoma: a clinicopathologic study of 164 cases. Cancer 1999; 86: 207-19.
10. Thomas PC. Conservative resection of the bronchial tree. J R Coll Surg Edinb 1956; 1: 169-86.