Adenoid Cystic Carcinoma Arising in the Intrapulmonary Bronchus

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A 63-year-old male was admitted to our hospital because of a coin lesion in the intrapulmonary bronchus. The patient had no symptoms such as cough or sputum. Imaging studies showed that the tumor was present in the S10b,c of the right lung. There was no metastasis or salivary gland tumor. Transbronchial tumor biopsy revealed a primary adenoid cystic carcinoma. The patient underwent a lobectomy, and has been well since the operation. This is an uncommon case of primary adenoid cystic carcinoma arising from the intrapulmonary bronchus.

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Introduction

Adenoid cystic carcinoma is a distinctive type of malignant epithelial neoplasm, which commonly arises in salivary glands (1). However, it has also been described in the breast, skin, uterine cervix, upper aerodigestive tract, and lung (2). Primary adenoid cystic carcinoma in the lung is rare, accounting for 0.2% of all cases of primary lung cancer (3). Because of its slow growth and absence of metastases until the late stage of the tumor, this tumor is regarded as a low grade malignancy. Most occur in the extrapulmonary bronchi, originating from bronchial glands. Here, we describe a rare case of adenoid cystic carcinoma arising in the intrapulmonary bronchus.

Case Report

A 63-year-old man was admitted to our hospital in April 1995, because of a coin lesion in the right lung. The patient had no symptoms such as cough or sputum. He had no previous medical problems. Upon admission, his height was 161 cm, weight 60 kg, blood pressure 154/70 mmHg, and heart rate 64/min. There was no swelling of the salivary glands. Laboratory examinations demonstrated a white blood cell count of 10,500/μl, hemoglobin of 17.0 g/dl, and a platelet count of 27.5x10⁴/μl. The erythrocyte sedimentation rate was 3 mm/h and C-reactive protein (CRP) was 0.0 mg/dl. The total protein level was 7.1 g/dl with a normal protogram. Liver function and renal function tests were both within normal ranges. The levels of tumor markers were not elevated (SCC: 0.8 ng/ml, SLX: 34 ng/ml, NSE: 5.9 ng/ml, carcinoembryonic antigen (CEA): 5.0 ng/ml, carbohydrate antigen 19-9: 22 U/ml). A chest X-ray showed a mass shadow 2 cm in diameter in the right lower lobe (Fig. 1). Chest computed tomography (CT) revealed a round shape and a well circumscribed mass without lymph node swelling (Fig. 2). The mass was enhanced just around the margin. An abnormal accumulation of ⁶⁷gallium was recognized in the right lower lung field, but not in other regions. Fiberoptic bronchoscopy revealed a tumor with a smooth surface in the lumen of B¹⁰ of the right lung (Fig. 3). Transbronchial tumor biopsy disclosed a primary adenoid cystic carcinoma. No metastasis was evident by imaging studies. A lobectomy was then carried out on May 8, 1995. The tumor was well circumscribed and located in the right lower segment (S10b,c). The resected tumor was 3.0 x 2.8 x 2.5 cm in size, white-yellow in color, and elastic hard in consistency (Fig. 4a). Histologically, the tumor arose from the submucosa of the intrapulmonary bronchus (Fig. 4b). The bronchial lumen was covered with normal ciliated cylindroform cells. But, a part of the tumor appeared in the bronchial lumen. The tumor cells were small and uniform with round or ovaloid nuclei and a cribriformed pattern. The center of the cylinder or tubule was filled with PAS and Alcian blue stained mucin (Fig. 4c). The stroma was also stained by Alcian blue. No metastasis was noted in the lymph nodes. These histologic findings are typical of adenoid cystic carcinoma. The patient has been well and has had no recurrence of the tumor since the operation.

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Figure 1. Chest radiograph showing a mass shadow in the right lower lobe.

Figure 2. Chest computed tomography revealing a round shape and a well circumscribed mass without lymph node swelling.

Figure 3. Fiberoptic bronchoscopy revealing a tumor with a smooth surface in the lumen of B10 of the right lung (arrow).

Discussion

It is known that primary salivary gland-type tumors rarely arise in the lung. Cases of primary adenoid cystic carcinoma in the lung account for approximately 0.2% of all cases of primary lung cancer.

Adenoid cystic carcinoma in the lung usually originates from the bronchial glands which are found almost exclusively in the extrapulmonary bronchi. Bronchial glands are present from the first to the fourth order bronchi, and they are found in 79% of the fifth order bronchi, but in only 11% of the sixth order bronchi. Therefore, this tumor commonly arises in the extrapulmonary bronchi. This case is quite different from those discussed in previous reports since the tumor arose in the intrapulmonary bronchus (B10), which is extremely rare for primary adenoid cystic carcinoma.

It has been believed that adenoid cystic carcinoma in the salivary glands has a better prognosis than other adenocarcinomas or squamous cell carcinomas in the salivary glands, because of its slow growth and the absence of metastasis until the late stages of the tumor. However, a recent report has described that the lesions in the lung do not necessarily behave indolently and may occasionally follow a much more aggressive course than that generally ascribed to their salivary gland counterparts. It is reported that the clinical stage at the time of operation represents the most important prognostic parameter, especially in cases arising in the extrapulmonary bronchus. In the present case, it was thought that the patient's good prognosis after the operation was due to the origin of the tumor from the intrapulmonary bronchus and the Ib clinical stage (T2N0M0).

Histologically, three main growth patterns are identified admixed in various proportions: cribriform (cylindromatous), tubular, and solid, as described in other locations. The most frequent and predominant pattern is the cribriform
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Figure 4. a) The resected tumor was well circumscribed, 3.0 × 2.8 × 2.5 cm in size, white-yellow in color, and elastic hard in consistency. b) Cross-section of the tumor (HE stain, ×1) showing that the tumor arose from the submucosa of the bronchial lumen (rectangle A). c) Enlarged view (HE stain, ×10) of the rectangle A in the Fig. 4b showing that the bronchial lumen was covered with normal ciliated cylindriform cells and the tumor cells were small and uniform with round or ovaloid nuclei and a cribriform growth pattern.

References