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Case report

A unique case of a huge mixed squamous cell and glandular papilloma of non-endobronchial origin with a peripheral growth

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ABSTRACT

We report a case of a huge solitary non-endobronchial pulmonary tumor in a 76-year-old male smoker. The tumor measured 11 × 10 × 8 cm. It was ill-defined, and it was located periphery of the right lower lobe with the subpleural cystic spaces. He underwent right lower lobectomy with mediastinal lymph node dissection and is free from tumor 30 months after surgery. Microscopically, it was composed of a proliferation of squamous and ciliated columnar epithelial cells with a few mucous cells. These cells were arranged in a papillary growth fashion extending along the fibrously thickened alveolar septa together with metaplastic bronchiolar and squamous epithelia displaying an usual interstitial pneumonia-pattern. Although the histologic features of the tumor were that of a mixed squamous cell and glandular papilloma (MSCGP), it was peripherally located and showed a lepidic growth, and it was much larger than previously reported MSCGPs. It is possible that the tumor developed in association with bronchial metaplasia in the periphery of the lung, and then extended along the surface of the reconstructed air spaces, which resulted in its unique histologic appearance. Further investigations of respiratory papilloma are needed to clarify the pathogenesis of these lesions.

1. Introduction

Solitary endobronchial papilloma, a rare benign pulmonary neoplasm, accounts for < 0.5% of all lung tumors [1]. It usually originates from within the trachea or main-stem bronchi. Only a few exceptional cases are located peripherally [2–4]. Pulmonary papilloma is divided into three subtypes in the WHO classification: squamous cell papilloma, glandular papilloma, and mixed squamous cell and glandular papilloma (MSCGP) [5]. MSCGP, the rarest subtype of pulmonary papilloma, is usually < 2.5 cm in size and comprises a mixture of squamous and pseudostratified, ciliated and non-ciliated cells, admixed with few mucin-containing columnar cells, arranged in a papillary growth fashion. The association between papilloma and interstitial pneumonia has not been documented. We describe a unique case of a huge non-endobronchial pulmonary tumor with features of MSCGP that extended widely along the alveolar septa in association with interstitial pneumonia.

2. Case report

2.1. Clinical presentation

A 76-year-old Japanese man with a smoking history of 20 cigarettes daily for 40 years (equivalent to a 40 pack-year smoking history) was admitted to our hospital to evaluate a mass in the right lung, detected by chest radiograph during a physical examination. He had no respiratory symptoms. Serum tumor markers, including squamous cell carcinoma antigen [24.5 ng/mL (normal, < 1.5)], cytokeratin 19 fragment [10.3 ng/mL, (< 3.5)], carcinoembryonic antigen [7.3 ng/mL, (< 3.5)]. Serum surfactant protein D [299 ng/mL, (< 110)], which is a biomarker of interstitial lung disease, was also elevated. Chest radiograph revealed an irregular opacity in the right lower lobe (Fig. 1a). A subsequent chest computed tomography (CT) scan demonstrated a large area of consolidation with air bronchogram in the right lower lobe, and reticular opacities with honeycombing in both lungs (Fig. 1b and c).

Abbreviations: MSCGP, mixed squamous and glandular papilloma; CT, computed tomography; FDG-PET, fluorodeoxyglucose positron emission tomography; CK, cytokeratin; RRP, recurrent respiratory papillomatosis; HPV, human papilloma virus; CMPT, ciliated muconodular papillary tumor

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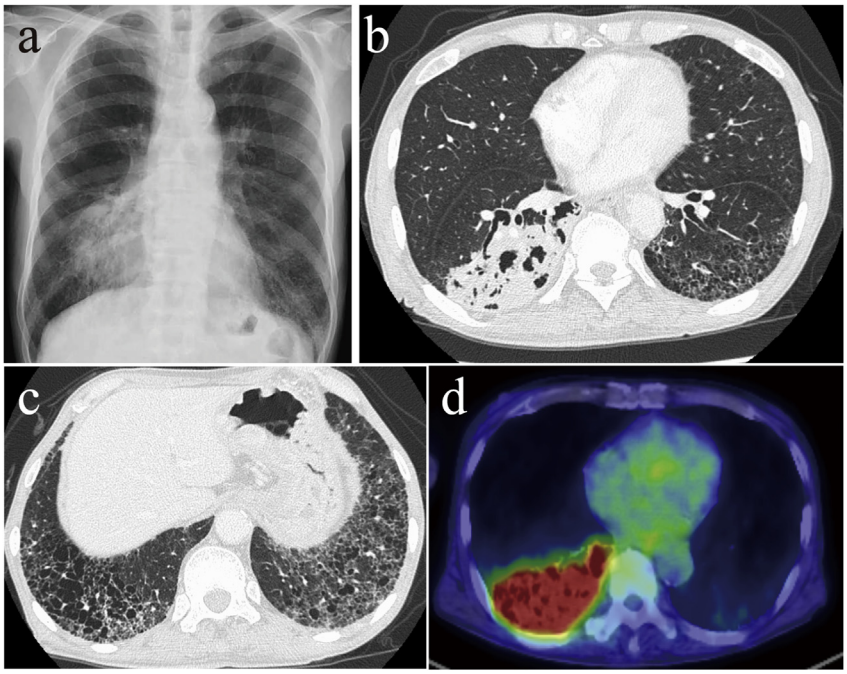

Fig. 1. Chest radiograph shows an irregular opacity in the lower lobe of the right lung (a). Chest computed tomography (CT) scan shows a large area of consolidation with air bronchogram in the right lower lobe and reticular opacities with honeycombing in both lungs (b, c). 18F-fluorodeoxyglucose positron emission tomography (FDG-PET)/CT shows increased uptake (maximum standardized uptake value of 13.03) in the right lung lesion (d).

The right lung lesion showed high uptake (maximum standardized uptake value at 1 hour and 2 hours 9.38 and 13.03, respectively) in the 18F-fluorodeoxyglucose positron emission tomography (FDG-PET)/CT (Fig. 1d). We did not observe any laryngeal or endobronchial lesion by bronchoscopic examination. We performed transbronchial lung biopsy, bronchial brushing, and bronchoalveolar lavage. Squamous cell carcinoma was suspected based on the cytological examination of the specimen. He underwent right lower lobectomy with mediastinal lymph node dissection and is free from tumor 30 months after surgery.

2.2. Pathological findings

The cytological smear of specimens obtained by bronchial brushing and washing showed high cellularity. There were many clusters of squamous epithelial cells in a sheet-like or papillary arrangement, and singly scattered cells, including keratinizing cells characterized by thick and deeply eosinophilic cytoplasm on a hemorrhagic or inflammatory background (Fig. 2a). The cells had centrally located and mildly hyperchromatic nuclei with prominent nucleoli. Therefore, the cytological findings were interpreted as squamous cell carcinoma. Additionally, there were singly scattered or clusters of ciliated epithelial cells and portions of mucous columnar epithelium without nuclear atypia (Fig. 2b and c).

Macroscopic examination of the right lower lobectomy specimen revealed that the tumor was located at the periphery of the lung It was ill-defined, measuring 11 × 10 × 8 cm in size, with a yellowish-to-whitish cut surface (Fig. 3). Variable-sized cystic spaces were seen mainly at the subpleural areas in the non-neoplastic pulmonary tissue, and in the tumor. The inferior lobar bronchus was free from the tumor. Microscopically, the tumor was composed of inflamed fibrovascular cores and papillomatous fronds covered by squamous and glandular epithelia (Fig. 4a). The squamous components were predominant (approximately 60% of the tumor) and showed several transitions from the ciliated columnar cells. The squamous epithelium was acanthotic and focally keratinizing. It coexisted with the glandular epithelium, which comprised ciliated or non-ciliated pseudostratified columnar cells and a few mucous columnar cells (Fig. 4b–d). No cell had significant nuclear atypia; stromal or vascular invasion was not evident. The tumor

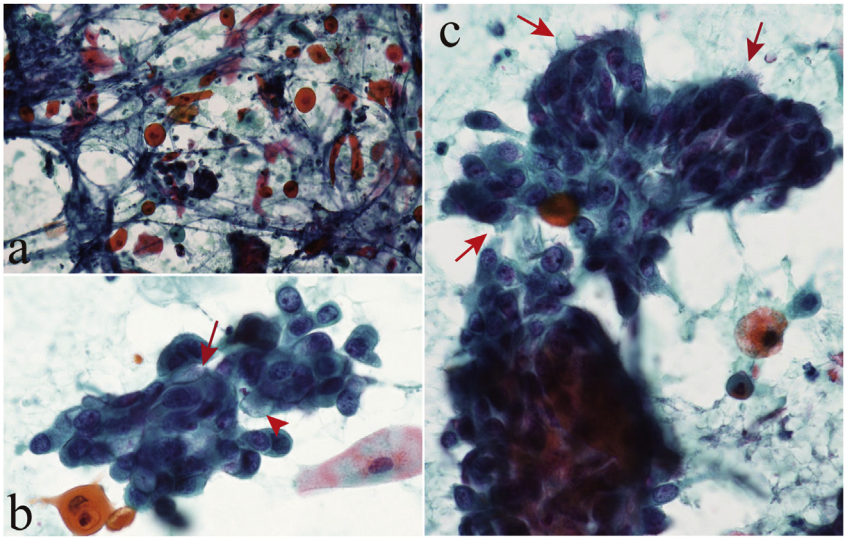

Fig. 2. Cytological findings of the tumor. The smear preparation shows clusters of squamous cells with abnormal keratinization on a hemorrhagic or inflammatory background (a), many clusters of ciliated epithelial cells (arrows), and some portions with columnar epithelium containing mucin (arrowhead) without nuclear atypia (b, c).