CASE REPORT

Basal cell adenoma of the lung identified with obstructive pneumonia

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Abstract

The seromucinous glands of the bronchi can give rise to tumors resembling those of the salivary glands. Basal cell adenoma (BCA) has not been reported in salivary gland-type tumors of the lung. Here, we report a case of an 86-year-old Asian man who suffered from pneumonia in the left upper lung field that may be associated with an endobronchial tumor occluding B1^+^2a+b. The bronchoscopic biopsy of the tumor revealed that the basaloid cells, which constituted a major component of the tumor and were positive for p40/p63 immunohistochemistry, exhibited a cord-like structure. The Ki-67 (MKI67) staining was less than 1% positive. These findings suggest that the endobronchial tumor was BCA of the lung, which caused obstructive pneumonia in the patient.

INTRODUCTION

The seromucinous glands of the trachea and bronchi can give rise to tumors resembling those of the salivary glands, which form polyoid endobronchial masses covered in bronchial epithelium [1]. Most of these tumors are located within the trachea or the main cartilaginous bronchi [2]. According to the latest 2015 World Health Organization classification of lung tumors [3], salivary gland-type tumors of the lung are separated into the following four categories: mucoepidermoid carcinoma, adenoid cystic carcinoma (ACC), epithelial–myoepithelial carcinoma and pleomorphic adenoma (benign mixed tumor). ACC is the most common salivary gland-type tumor of the lung and accounts for 75–80% of all cases but less than 0.3% of all tracheobronchial tumors [2, 4].

Salivary gland neoplasms are uncommon and constitute 3–4% of all head and neck neoplasms. Most (70%) salivary gland tumors arise in the parotid gland and are generally benign [5]. Pleomorphic adenoma is the most common type of salivary neoplasms and constitutes 45% of cases with salivary neoplasms, while ACC accounts for only 10% of the cases [6]. Therefore, the percentage of the tumor types may differ between the lung and salivary glands although both tumors are histologically indistinguishable from each other [2, 4–6].

Basal cell adenoma (BCA) is an exceedingly uncommon benign tumor of the salivary glands and accounts for less than 0.2% of cases with salivary neoplasms [5, 6]. BCA usually occurs in adult patients, and there is a slight predilection for females. Most cases occur in the parotid, but a few cases have been reported within the periparotid lymph nodes and minor salivary gland sites, such as the upper lip. Similar to other benign neoplasms, BCA presents as a slow-growing, asymptomatic mass [2, 5].

Here, we report a unique case of a patient with an endobronchial mass that may be associated with obstructive pneumonia due to BCA in the B1^+^2 bronchus.

CASE REPORT

An 86-year-old Asian man was referred to our division due to refractory pneumonia of the left upper lung field (Figs. 1a, 2a and b) that had been treated with piperacillin/tazobactam, followed by biapenem. The patient had a 22 pack-years smoking...
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Figure 1: (a) Chest radiograph (postero-anterior view) revealing consolidation in the upper left lung field. Left costophrenic angle is dull, and the gastric air bubble (red arrow) was displaced inferiorly, which is reminiscent of diaphragmatic eventration (black arrows), suggesting left subplumonic effusion. (b) Chest radiograph (postero-anterior view) revealing improvement in the consolidation in the upper left lung field 3 months after bronchoscopy.

Figure 2: (a, b) Axial chest CT scans with lung (a) and mediastinal (b) windows revealing consolidation mainly located in the S1+2a. Red circles show air bronchogram sign of B3c. (c, d) Axial (c) and coronal (d) chest CT scans with lung window revealing an endobronchial tumor located at B1+2. AA: arcus aortae; LUDB: left upper division bronchus; S: superior vena cava; T: trachea; 5: fifth thoracic vertebra; 6: sixth thoracic vertebra.

history. His past medical history was significant for hypertension and diabetes mellitus.

The physical examination of the patient at our office visit was unremarkable. There were no abnormalities in his mouth except for removable partial dentures. His swallowing function was within normal limit.

A computed tomography (CT) scan of the chest showed an endobronchial mass at B1+2 (Fig. 2c and d) that nearly obstructed a left upper lobe apico-posterior segmental bronchus (Fig. 2d). The bronchoscopy confirmed a polypoid endobronchial mass with a smooth surface and capillary telangiectasia that occludes B1+2a+ b (Fig. 3a and b). The tumor was removed by forceps biopsy.

The hematoxylin and eosin (HE) staining of the tumor revealed a well-circumscribed, solid tumor comprising small, rounded, basophilic cells in subbronchial epithelial cells (Fig. 4a and b). The tumor cells were relatively equal in size and exhibited a cord-like and alveolar structure and palisading in part (Fig. 4b). Table 1 summarizes the immunohistochemistry (IHC) results of the pan-keratin antibodies AE1 and AE3 (AE1/3), cytokeratin (CK) 7 (CK7), CK20, thyroid transcription factor-1 (TTF-1), cluster of differentiation 56 (CD56; neural cell adhesion molecule), synaptophysin (major synaptic vesicle protein p38), p40/p63 (different isotypes of homologs for p53), α smooth muscle actin (αSMA) and Ki-67 (MKI67). The p40/p63-positive cells mainly proliferated with CK7- or αSMA-positive cells, suggesting that a major component of the tumor constitutes basal/basaloid cells. The CK7-positive cells were surrounded by p40/p63-positive abluminal basaloid cells (Fig. 5b–e). The Ki-67 staining revealed less than 1% positive staining (Fig. 5f).

As summarized in Table 1, CD56 and synaptophysin (neuroendocrine markers), CK20 (goblet cell marker) and TTF-1 (type II respiratory epithelial cell marker) were negative, while the pan-keratin marker AE1/3 was positive. The cells positive for CK7, which highlighted the ductal cells, were surrounded by p40/p63-positive abluminal basaloid cells (Fig. 5b–d). The alveolar configuration identified by the HE staining corresponds to CK7-positive cells, while the cord-like structure corresponds to p40/p63-positive cells (Fig. 4a and b). These findings indicate that the tumor mainly comprises p40/p63-positive basaloid cells forming a cord-like structure and contains CK7-positive luminal and αSMA-positive myoepithelial cells.

Furthermore, less than 1% of the cells were positive for Ki-67, which is a specific nuclear marker of cell proliferation (Fig. 5f). Given that carcinomas, such as ACC and basal cell carcinoma, contain >20% and 21±4.4% of Ki-67 positive cells, respectively [7, 8], the tumor presented here is most likely benign. In fact, BCA
Figure 3: (a, b) Bronchoscopic images showing a polypoid tumor located at B\textsuperscript{1+2} that occludes B\textsuperscript{1+2a+b} (red arrows).

Figure 4: (a, b) HE staining of a polypoid endobronchial tumor at B\textsuperscript{1+2}. Small, rounded, basophilic cells proliferating in subbronchial epithelial cells. The tumor cells are relatively equal in size and exhibit a cord-like and alveolar structure. (a) Low magnification (×0); (b) high magnification (×40).

Table 1: Summary of IHC in an endobronchial tumor of the bronchus

| AE1/3 | CK7     | CK20     | TTF-1 | CD56 | Synaptophysin | p40/p63* | αSMA | Ki-67 |
|-------|---------|----------|-------|------|---------------|---------|------|-------|
| +     | Partially + | –       | –     | –    | –             | +       | Partly + | <1%   |

*p40/p63 proteins are different isotype homologs of p53 [7].

The patient refused surgical resection of the tumor including laser-assisted mechanical debulking after informed consent of the histopathological results mainly due to his old age. Pneumonia did not relapse ~9 months after the forceps biopsy (Fig. 1b).

DISCUSSION

Salivary gland-type tumors of the lung presumably originate from submucosal bronchial glands and faithfully recapitulate the histologic features of similar tumors arising in the salivary glands [9]. The utility of IHC in distinguishing these tumors from one another might be limited given that most of these tumors share the features of myoepithelial differentiation or a plain epithelial immunophenotype [9, 10]. While it is still believed that conventional histology is the most important tool for the diagnosis of these tumors [9], conventional histology is limited to clearly determining the origin of tumors and degrees of malignancy. In fact, IHC was a powerful tool in determining the main immunophenotype of the tumor and distinguishing BCA from basal cell carcinoma in our case.

When an endobronchial tumor is identified, ACC should be first considered, then mucoepidermoid carcinoma, and other less common tumors such as pleomorphic adenoma (benign mixed tumor), acinic cell carcinoma and oncocytoma could be considered as alternatives [2]. On the other hand, differential diagnoses could first be explored for pleomorphic adenoma, mucoepidermoid carcinoma, ACC and adenocarcinoma arising in the salivary glands [5, 6, 9]. BCA is uncommon in either site of origin.

Interestingly, salivary gland adenomas can be viewed as neoplasms lying on a spectrum separable by the dominance of the participating cell types, the morphologic heterogeneity and organization of the cell types and the type and quantity of
extracellular matrix products. The two extremes of the spectrum, i.e. canalicular adenoma and myoepithelioma, are represented by exclusive luminal cell and myoepithelial cell differentiation, respectively [10]. Therefore, the pathological diagnosis of BCA, which was mainly composed of basal cells accompanied by luminal and myoepithelial cells in our case, was valid and feasible.

Regarding the treatment options for benign or low grade malignancy of the bronchi, laser-assisted mechanical resection in rigid bronchoscopy may be the first option. The technique can recanalize the airways and lead the patient to parenchyma, sparing surgery [11].

In summary, we report a unique case of a patient with BCA of the lung, which caused obstructive pneumonia. To the best of our knowledge, this article is the first to report BCA of the lung.

ACKNOWLEDGEMENTS
The authors thank Dr Yoshitake Takagi, GeneticLab Co., Ltd, for the histopathology and photographs used in this study.

CONFLICTS OF INTEREST STATEMENT
The authors have no conflicts of interest to declare.

FUNDING
There is no source of funding to report for this case report.

ETHICAL APPROVAL
No ethical approval was required as this was a clinical case.

CONSENT
Patient permission was obtained prior to writing this report.

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