Int J Clin Exp Pathol 2017;10(1):747-749
www.ijcep.com /ISSN:1936-2625/IJCEP0040890

Case Report

Coexistence of lung alveolar adenoma with bronchogenic cyst: a case report and literature review

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Received September 28, 2016; Accepted October 25, 2016; Epub January 1, 2017; Published January 15, 2017

Abstract: Alveolar adenoma (AA) is extremely rare benign tumor of the lung. This tumor is comprised of proliferative type II alveolar epithelium and septal mesenchyma and shows characterized immunohistochemical patterns. Up to date, there have been only few reports describing this disease. Here we described a case of alveolar adenoma in a 48-year-old woman. This female patient was incidentally found to have a left lower lobe mass by the chest CT scan and went through a left lower lobectomy. A nodule were observed and the pathological and immunohistochemical examinations of the surgically resected nodule revealed this lesion to be an alveolar adenoma. In addition, an isolated bronchogenic cyst was also observed. The postoperative course was uneventful and no recurrence was observed after 5 years. In conclusion, we reported the rare disease of Alveolar adenoma coexisting with bronchogenic cyst. Besides, we also gave a literature review of previously published papers describing alveolar adenoma.

Keywords: Alveolar adenoma (AA), bronchogenic cyst, benign tumor

Introduction

Alveolar adenoma (AA) is a rare peripheral lung tumor, this disease was first described by Yousem and Hochholzer in 1986 [1]. In most of the cases, AA was discovered incidentally during routine chest radiographs. AA is regarded as a benign neoplasm, and cases with recurrence after resection have not been reported [2, 3]. AA is characterized by proliferation of type II alveolar epithelium and septal mesenchyma [4]. Herein, we described a rare case of typical AA coexisting with bronchogenic cyst.

Case representation

A 48-year-old woman with medium incapability of her right lower extremity went to the hospital and was later diagnosed as mild cerebral infraction. This patient was soon admitted to the hospital. During the hospitalization preparation, CT scan revealed a 3.5 × 3.0-cm mass in the left lower lobe (**Figure 1**). This woman then underwent a left lower lobectomy and tolerated the procedure well without any remarkable postoperative course.

Grossly, a diameter of 34 mm bronchogenic cyst (**Figure 2A**) and solid mass with cysts and white cut surface (35 × 33 × 26-mm in size) were observed. Pathologically, this tumor was comprised of epithelial and interstitial cells. The epithelial cells were morphological flat and they lined together to form different sizes of cysts (**Figure 2B**). The cystic spaces were filled with eosinophilic granular material and histiocytes. The interstitial cells distributed in cysts septa were spindle-shaped and showed no heterogenesity. Nuclear atypia and mitosis was rarely observed. Immunohistologically, the epithelial cells of tumor sections were positive for cytokeratin 7 (CK7) (**Figure 2C**), thyroid transcription factor-1 (TTF-1) (**Figure 2D**), epithelial membrane antigen (EMA). Interstitial cells were partially positive for smooth muscle actin (SMA). Additionally, the whole tumor section was negative for P53, CD31, CD34 and D2-40. The overall Ki-67 index was lower than 1%. Histologic and immunohistochemical examinations of the surgically resected solid tissue revealed this lesion to be an alveolar adenoma. The postoperative course was uneventful and no recurrence was observed after 5 years.

Discussion

AA is an extremely rare benign neoplasm of undefined histogenesis with unique gross and

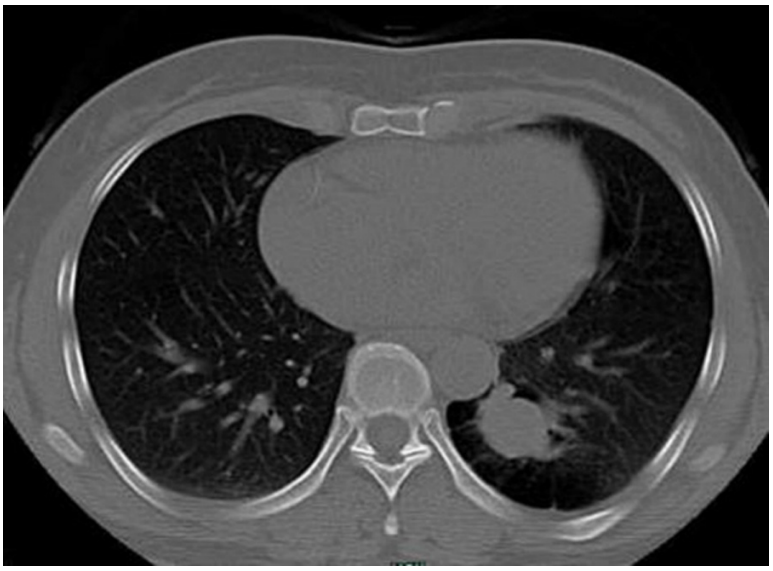

Figure 1. CT scan of the female patient. The CT scan revealed a 3.5 × 3.0-cm mass in the left lower lobe.

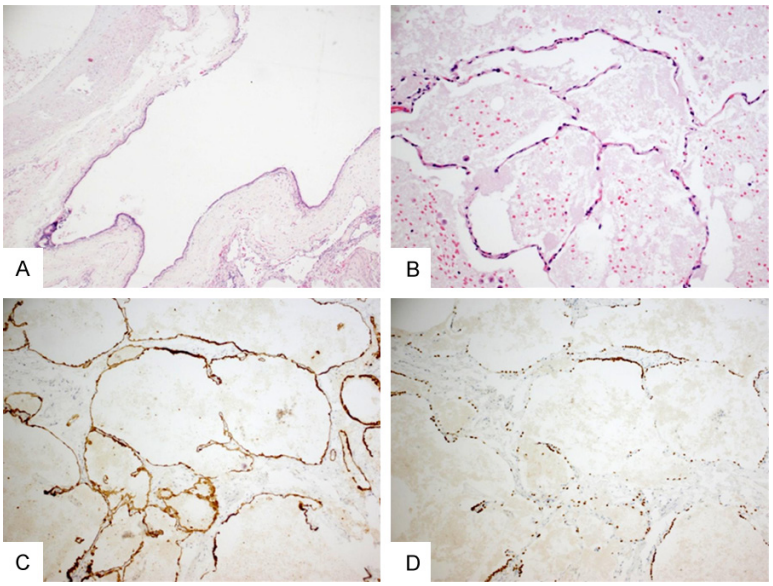

Figure 2. Pathological and immunohistochemical analysis. A. H+E × 50, a bronchogenic cyst was observed; B. H+E × 200, The epithelial cells formed different sizes of cysts and the cystic spaces were filled with eosinophilic granular material and histiocytes; C. The epithelial cells of tumor sections were positive for CK7; D. The epithelial cells of tumor sections were positive for TTF-1.

microscopic features. Until now, approximately 30 cases in English have been reported, whereas the exact number of alveolar adenoma cases was difficult to determine since this disease was sometimes confused with other rare pulmonary disease [5-7]. Summarized from these articles and review papers, the age range of alveolar adenoma patients varied from 34 to 83 years and was with a slight female predominance [8]. This disease was usually found very incidentally in asymptomatic patients. There was only one previous literature reported an alveolar adenoma patient presenting severe dyspnea [9]. Our patient was also found alveolar adenoma very incidentally through hospitalization regular check. Although AA usually presents of a solitary lung solid nodule, it has no specific imaging features. Therefore, correct diagnosis needs to be combined with morphologic features and immunohistochemical features.

AA represents a benign proliferation of both the alveolar epithelium with no signs of malignance and fibrous tissue originating from septal mesenchyma [3], but occasionally coexists with lung carcinoma and malignant transformation [4, 10]. In this case, nuclear atypia and mitosis of epithelial and stromal cells were rarely observed. Moreover, our patient was found to have bronchogenic cyst additionally to AA. To the best of our knowledge, this is the first case report mentioned the coexistence of AA with bronchogenic cyst. If there is a relationship between two diseases is not clear.

Differential diagnoses like sclerosing pneumocytoma, lymphangioma, atypical adenomatous hyperplasia and bronchioloalveolar adenocarcinoma are sometimes problematic and need to pay attention. Sclerosing pneumocytoma has a typical polymorphic histologic appearance with a mixture of solid, vascular, and sclerotic and papillary patterns whereas AA occurs along the alveolar structure. Moreover,

TTF-1 expression in alveolar adenoma is very important for the discrimination of alveolar adenoma from sclerosing pneumocytoma. Lymphangiomas are characterized by lining cells that positive for endothelial marker and negative for CK. Atypical adenomatous hyperplasias present an atypical bronchoalveolar proliferation, whereas AAs lack cellular atypia. The well-circumscribed growth pattern, lack of lepidic growth and cellucar atypia of AA distinguishes it from the other carcinomas.

The conservative surgery is considered to be the best treatment for AA and no further therapy is needed. It usually has an indolent clinical progression and is absent of recurrence or metastasis after complete resection which indicating its benign behavior. Our patient only received lobectomy and remained health in the 5 follow-up years, no disease recurrence or metastasis was reported.

In conclusion, the present study is the first to report a case of AA coexisting with bronchogenic cyst. AA has been considered as a rare benign disease and can be identified based on its typical histological and immunohistochemical features. Surgical resection is thought to be curative for this tumor and almost all patients recovered very well from this disease.

Acknowledgements

The study participant provided informed consent for the findings of this case to be published.

Disclosure of conflict of interest

None.

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