

# Alveolar adenoma with poor imaging: a case report

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## Abstract

Alveolar adenoma is an isolated, well-defined peripheral lung tumor that originates from type II alveolar cells. The tumor consists of a network of simple, low-cubic, epithelium-coated lacunae with varying amounts of fine and inconspicuous-to-thick spindle cells that sometimes contain mucus sample matrix. Few cases of alveolar adenoma have been reported. These tumors are usually detected by imaging examinations where the alveolar adenoma typically presents as a peripheral, solitary cystic nodule in the lung. The presentation may mimic that of other types of lung tumors, consequently leading to difficulties in the differential diagnosis of this condition. Thus, accurate diagnosis of alveolar adenoma is based on a combination of pathological sections and immunohistochemistry. This study describes an alveolar adenoma in a 59-year-old female patient. Chest X-ray imaging and chest computed tomography identified malignant lesions in the right upper lobe. The patient subsequently underwent a thoracoscopic right upper lobectomy. The diagnosis of alveolar adenoma was confirmed after pathological examination of the excised postoperative tissue. The disease course was stable, and there was no recurrence of pulmonary lesions during 3 years of postoperative patient follow-up. Herein, we report the case of a patient with benign alveolar adenoma with poor imaging and pathological results.

## Keywords

Alveolar adenoma, pulmonary nodules, poor imaging, peripheral lung tumor, pulmonary lesions, benign

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## Background

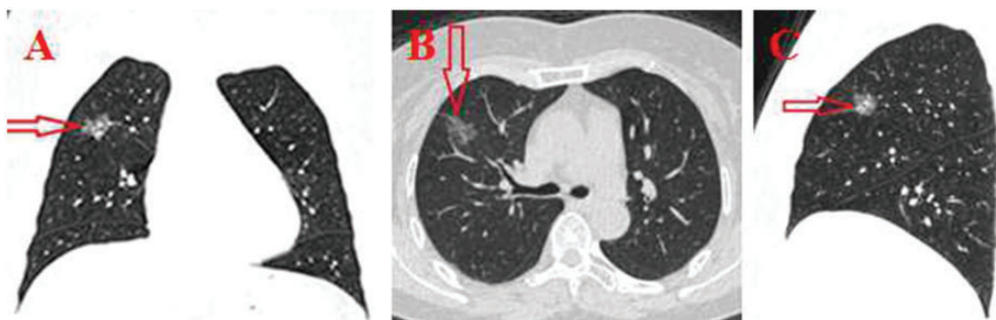
Alveolar adenoma is a peculiar pulmonary disease with uncertain histogenesis. In 1986, Yousem et al.<sup>1</sup> first reported the disease, and few cases have been reported since the initial report. This type of tumor is benign, based on histological findings; it does not recur after resection and is usually detected as a clear solitary pulmonary nodule on chest X-ray images or computed tomography (CT) scans.<sup>2-4</sup> We describe here the case of a patient with alveolar adenoma to facilitate the differential diagnosis of this tumor from other peculiar lung tumors.

## Case presentation

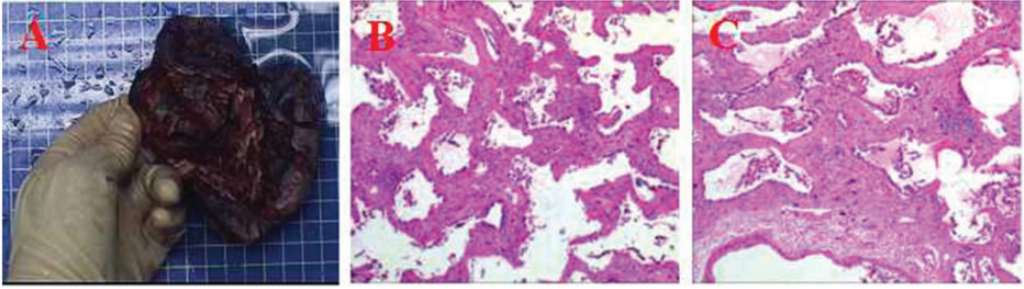
On November 26, 2015, a 59-year-old female patient was hospitalized because of a month-long history of coughing and expectoration. A routine preoperative chest CT examination revealed a patchy ground-glass density in the right upper lobe that measured about  $2 \times 1.8 \text{ cm}^2$ , suggesting the possibility of lung cancer (Figure 1). A bone scan and brain CT detected no abnormalities. On November 30, 2015, the patient underwent a thoroscopic lobectomy (right). During intraoperative examination, a mass in the anterior segment of the right upper lobe was detected. The mass measured approximately  $2.4 \times 1.5 \times 1.9 \text{ cm}^3$ , the image was of

medium texture, and the tumor's position was fixed. There was no obvious swelling of the hilar lymph nodes, and no other lumps were detected in the rest of the lungs. Samples of the lung lesions were taken for pathological examination.

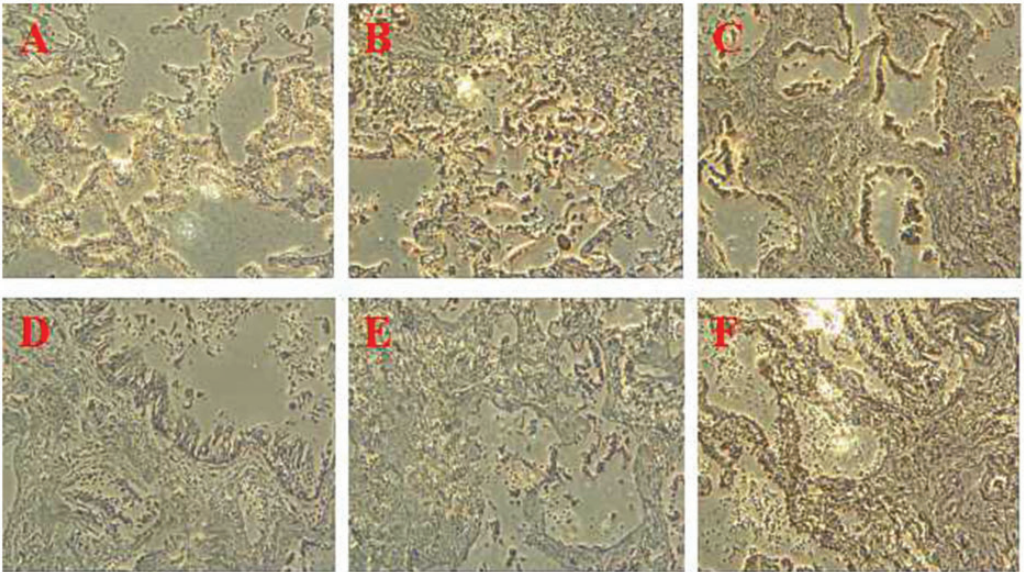
Histological samples were assessed. Grossly, the tumor contained gray-red cystic nodules, measuring approximately  $7.4 \times 5.5 \times 4.9 \text{ cm}^3$  (Figure 2a), the image was of medium texture, the tumor had no capsule, and the boundary between the mass and the surrounding lung tissue was unclear. Microscopically, after hematoxylin and eosin (HE) staining, the lesions were identified as polycystic. The capsule walls were covered with flat epithelium and cubic epithelium (type II alveolar epithelial cells), and eosinophilic granular material could be seen in the capsules. The cystic dilatation was similar to that of the alveoli, and part of the wall was damaged because of the formation of larger cystic expansion. Inter-wall cells containing thicker spindle cells were visible between the terminal pulmonary artery and vein. There was also a small amount of lymphoid and macrophage infiltration. No hemorrhagic or necrotic areas were seen in the lesion tissues. In addition, no lepidic growth was seen in the histiocytes and infiltrating inflammatory cells (Figure 2b,c). Immunohistochemical examination was performed (Figure 3),



**Figure 1.** a-c. CT images of the coronal, transverse, and sagittal directions. A patchy ground-glass density of about  $2 \times 1.8 \text{ cm}^2$  is visible in the right upper lobe.



**Figure 2.** Histological observations. a Macroscopic view of the mass: the gross organization is about  $7.4 \times 5.5 \times 4.9 \text{ cm}^3$  and comprises gray red cystic nodules. b,c Hematoxylin and eosin (HE) staining: No lepidic growth was seen in the histiocytes and infiltrating inflammatory cells (HE staining: magnification  $\times 100$ ).



**Figure 3.** a-f. Immunohistochemical (IHC) examination: Positive staining of type 2 alveolar epithelial cells for thyroid transcription factor-1 (TTF-1), cyto-keratin (CK), and NaspinA. Ki-67 and S-100 were negative. For VIM, interstitial cells were positive and epithelial cells were negative (IHC staining: magnification  $\times 100$ ).

and the results were positive for pan-cyto-keratin (CK), thyroid transcription factor-1 (TTF-1), and naspinA, but the results were negative for S-100 and Ki-67 (5%). Interstitial cells tested positive, while epithelial cells tested negative, for VIM. The final pathological diagnosis was alveolar adenoma.

## Discussion

Alveolar adenoma is a very rare benign pulmonary neoplasm lung tumor that originates from type II alveolar epithelial cells.<sup>2,4,5</sup> The age of onset is 39–74 years, and it is typically more common in middle-aged and older women.<sup>5</sup> Since the first report by

Yousem et al. in 1986, only a few cases have been reported.<sup>1</sup> Lumps are often present in the left lower lobe as an isolated nodule or mass located in the peripheral part of the lung. The tumor boundary is clear and often the boundary is irregular. Most of the tumors remain stable in size over a long period of time, although a few cases have shown an increase in size in the short term.<sup>6,7</sup> Common methods, such as chest CT, electronic fiber bronchoscopy, sputum examination, and other detection methods have not been useful for identifying the disease. Unfortunately, percutaneous biopsy may pose a potential danger because it is difficult to avoid malignant transfer by the needle. Therefore, postoperative pathological examination may be the only confirmed method for a definitive diagnosis.<sup>5</sup> Usually, chest X-ray imaging or a chest CT scan can detect significant solitary pulmonary nodules. Imaging of pulmonary alveolar adenoma typically detects a clear boundary between the mass and normal tissue. The current mass can be seen in the “vacuole sign” and on both the chest X-ray and CT scan (Figure 1). In this Figure, the upper lobe of the anterior segment has an irregular shadow and the boundary is unclear, with a “spicule sign” around the lesion location, near the pleural adhesions, and without a “vacuole sign”. The mediastinal window is also unclear.

We have reported this case to provide a new approach for the diagnosis of solitary pulmonary nodules with poor imaging performance.

Immunohistochemical examination revealed positive results for CK, TTF-1, and NaspinA in the epithelial lining, with negative results for Ki-67 and S-100. Additionally, epithelial cells tested positive for type II lung cell markers, indicating that the epithelial layer is derived from type II lung cells. In this case, it was difficult to distinguish between alveolar adenoma and adenocarcinoma through routine chest CT examination. In addition, both

tumors exhibited positive immunoreactivity for TTF-1, pan-CK, and NaspinA. In the future, immunohistochemical staining and overall structural analysis should be used to make a definitive diagnosis of alveolar adenoma because it has a polycystic histology and it is similar to the normal lung parenchyma.<sup>5</sup> Resection of the entire tumor with subsequent pathological analysis is ultimately necessary.

## **Conclusions**

The best way to diagnose alveolar adenoma is to analyze the pathological tissue immunohistochemically. Surgical resection seems to be the only effective method for treatment, and recurrence after complete resection of the tumor has never been reported. In this case, recurrence did not occur after 3 years of follow-up.

## **Abbreviations**

CT, computed tomography; TTF-1, thyroid transcription factor-1; HE, hematoxylin and eosin; IHC, immunohistochemistry; CK, cyto-keratin

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## **Authors' contributions**

XW, BY, and XJY collected all the data and wrote the manuscript. JCL performed the surgical procedure and contributed to analyzing the data and revising the manuscript. JCL also contributed to the revising the manuscript. LTH revised the manuscript repeatedly. All authors have read and approved the final manuscript.



### Availability of data and materials

The availability of the data and material section concerning the case report is related to all the diagnostic examinations that the patient underwent during their hospitalization. Publication of all these data has been authorized by the First Affiliated Hospital of Nanchang University.

### Consent for publication

Consent for publication of the manuscript and the related images from the patient and/or their relatives has been provided by the First Affiliated Hospital of Nanchang University.


### Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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