Clinical Case Reports International

Case Report
Published: 02 Mar, 2024

Misdiagnosis of Multiple Mucinous Cystadenomas in Both Lungs in Adults as Cryptococcal Infection: A Case Report

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Abstract

Pulmonary Mucinous Cystadenoma (PMCA) is an extremely rare benign tumor of the lungs. We reported a 58-year-old woman with incidental discovery of multiple nodules in both lungs on routine chest CT. Based on pathological and immunohistochemical findings, the final diagnosis was PMCA. Thoracic and superior abdomen Computed Tomography (CT) scan with contrast discovered multiple nodules in both lungs, with the largest nodule located in the lateral basal segment of the right lower lobe, adjacent to the pleura, measuring approximately 17 mm × 15 mm. About 3 months after the operation, the patient is in good condition with no recurrence or metastasis and is undergoing long-term follow-up.

Keywords: Pulmonary mucinous cystadenoma; Rare benign lung tumor; CT manifestations; Pathological manifestations

Introduction

Mucinous Cystadenoma (MCA), characterized by the accumulation of mucus pools within the cystic space and lined by columnar epithelium. These mucinous cystic tumors can be benign (mucinous cystadenoma), malignant (mucinous cystadenocarcinoma), or borderline malignant [1-3]. Few cases of Pulmonary Mucinous Cystadenoma (PMCA) have been reported worldwide [4]. Since the first description by Eck in 1969 [5] and a subsequent description by Sambrook Gower in 1978, PMCA of the lung has been regarded as a separate entity [6]. This case report summarizes the characteristics of a PMCA patient diagnosed and treated at our hospital.

Case Presentation

A 58-year-old female patient was diagnosed with pulmonary nodules one month before admission to our hospital. The pulmonary nodules were discovered incidentally on a routine chest CT (Figure 1), and she exhibited no respiratory discomfort such as coughing and sputum, and dyspnea. She had a history of hypertension for 5 years and used hydrochlorothiazide antihypertensive drugs regularly. Her blood pressure was well controlled. The patient has no smoking history, no long-term exposure to industrial dust, poisons and radioactive substances, and no family history of cancer-related genetic diseases. No special problems found during physical examination. Therefore, clinicians do not make special treatment for this.

Following the ineffectiveness of anti-infective therapy, the patient underwent single port thoracoscopic wedge resection of the right lung. Immunohistochemical analysis of the resected specimen revealed positive staining for p63, Thyroid Transcription Factor-1 (TTF-1), and Ki-67 (3%) (Figure 2).

At 3 months post-surgery, the patient had no tumor recurrence or metastasis and is undergoing regular follow-up every 3 months. If there is no metastasis or recurrence of the disease after one year, the chest CT should be reviewed annually.

Discussion

According to previous case reports and reviews, the incidence ratio of females to males is 7:5. Most cases of PMCA do not show any clinical symptoms and are only found by routine imaging. Case reports suggest that PMCA appears on CT images as a solitary mass located in the periphery of the lung with well-defined borders and occasionally with inflammatory manifestations of adjacent lung tissue. To date, there is only 2 cases reporting the presence of mucinous cystadenoma in multiple

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Received Date: *12 Feb 2024*
Accepted Date: *27 Feb 2024*
Published Date: *02 Mar 2024*

Citation:
Huang J, Yu H, Feng X. Misdiagnosis of Multiple Mucinous Cystadenomas in Both Lungs in Adults as Cryptococcal Infection: A Case Report. Clin Case Rep Int. 2024; 8: 1663.

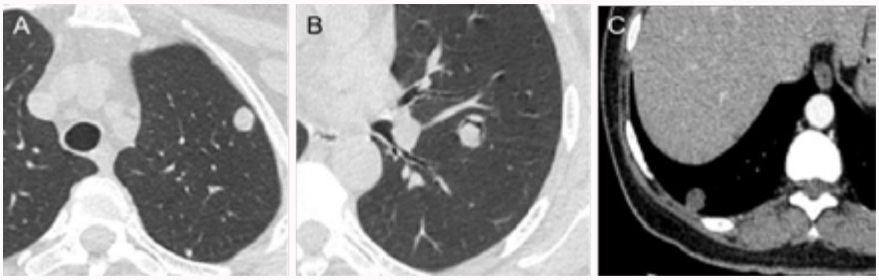

Figure 1: Coronal and sagittal CT images of the chest depict multiple well-defined bilateral pulmonary nodules in predominately peripheral locations. A) The larger nodules were in the lateral basal segment of the right lower lobe. B) The left lung mass with an "air crescent sign" can be seen running through the largest mass in the lateral basal segment of the right lower lobe. C) Contrast-enhanced CT scan in the arterial phase shows the right lower lobe nodule with marginal enhancement in the mediastinal window.

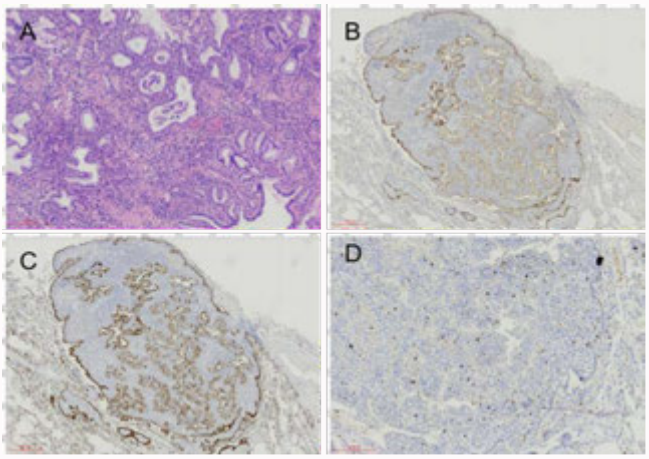

Figure 2: A) HE stains showed that the lesions were multiple cystic lesions, some areas were multilocular cystic structures (HE, ×200). B) Immunohistochemical detection revealed positive staining for p63 (×400). C) Immunohistochemical detection revealed positive staining for Thyroid Transcription Factor-1 (TTF-1) (×400). D) Ki-67 (3%) (×400).

lungs [1,7]. Ours is the third reported case of multifocal PMCA. Our postoperative pathology confirmed that all nodular lesions in the right lung were PMCA. Although there were cavitary shadows present in the left lung lesions, and solid components were seen adjacent to the cavity, this imaging manifestation is often considered a typical sign of pulmonary *Aspergillus* infection. Nevertheless, as the imaging manifestations of contrast-enhanced CT were the same as those of the right lung lesions, we consider the left lung lesions very likely to be PMCA. We have not identified patients with associated elevated tumor markers. On reviewing the reported cases of PMCA, we found that the images of PMCA show certain characteristics. The cysts can be single or multiple in an image and are mostly located in the peripheral zone of the lung. The presence of characteristic mucous substances in the lesion makes the determination of enhancement of PMCA through imaging not obvious. Our case only showed mild marginal enhancement. However, note that in our case it was hard to differentiate PMCA from pulmonary *Aspergillus* infection. The same diagnostic difficulty has also been reported with bronchial cysts, congenital adenomatous malformations, or post-infectious bronchial cysts, metastases, pulmonary tuberculosis, etc. [15,16].

Fiberoptic bronchoscopy and fine-needle aspiration biopsy usually do not show any definitive abnormal findings and cannot be used for a definitive diagnosis. All the reports on PMCA cases rely on histopathology as a means of definitive diagnosis. Pulmonary mucinous cystadenoma is a benign tumor with low tumor heterogeneity, few mitoses, mild biological behavior (degree of differentiation, lymph node metastasis, etc.) and other pathological features of benign tumors. Reports have presented PMCA as a single- or multi-housed cystic lesion filled with mucus. It has a single layer of columnar or cuboidal epithelium covering the cyst wall and shows no cellular anisotropy and only rare nuclear fission images microscopically [1,3,8-10,13,17]. Slight cellular nuclear anisotropy [4,12,14], with localized multinucleated giant cells has also been reported [10,11]. In our case, when we observed the papillary or adenoid structure under the microscope, we found the local epithelium to be mildly eosinophilic. These characteristics are consistent with the previously reported case of multifocal PMCA [1] and thus lend support to our case as a benign lung tumor. However, Moneke et al. [4] reported that though they initially diagnosed PMCA, the histological examination showed that there were focal singlet cells in the mucus, and was positive for CEA. Their final diagnosis was local invasive cancer. In the literature review, we came across only 1 case of invasive cancer in PMCA lesions [4]. Thus, strict and accurate sampling and pathological immunohistochemical examinations are essential for evaluating the biological behavior of PCMA. We also believe that long-term follow-up is necessary for an accurate prognosis. Moneke et al. suggest that KRAS driver mutations (mutations typical of mucinous lung cancer) may also be a mechanism by which PCMA causes mucinous cystadenocarcinoma. Note that in our case, the immunohistochemical staining was positive for Thyroid Transcription Factor (TTF-1), and also for P40 and P63, which were not detected in previous reports, to exclude the possibility of lung squamous cell carcinoma.

The treatment of PMCA usually involves surgical resection. Our patient underwent right lung wedge resection under video-assisted thoracoscopy, and PMCA was considered from both intraoperative and postoperative pathology. We did not perform further surgical and postoperative treatment for the lesions of the left lung and only informed the patient to review every three months. Everything works fine so far. If there is no metastasis or recurrence of the disease after one year, the chest CT should be reviewed annually. The prognosis of the cases reviewed in the literature was good. Except for one case of PMCA reported by Matsuo [14], which recurred 20 years after the initial operation, the other cases had no tumor recurrence, malignant transformation, or metastasis. Therefore, we can assume that PMCA has a good prognosis.

In conclusion, case reports of PCMA are very rare, and our current understanding of PMCA occurring in the lung is insufficient. Though

the location and characteristic mucous content impart a certain characteristic on imaging and can be used for assumption, it is very difficult to make a preoperative diagnosis on this basis. Our diagnosis of PMCA relied on histopathological and immunohistochemical features. Although the cases we reviewed showed benign biological behavior, there were also case reports of malignant transformation. Thus, a regular follow-up after surgery is very necessary. We will track the prognosis of our case for a long time, and if there are special findings, we will report and communicate again.

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