Case Report
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Bronchogenic Cyst in an Intradiaphragmatic Location: A Case Report

횡경막 내에 위치한 기관지원성 낭종: 증례 보고

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Bronchogenic cysts are congenital lesions usually observed in the mediastinum, near the tracheal carina and middle mediastinum. Herein, we present an exceedingly rare case of intradiaphragmatic bronchogenic cyst with an infectious complication in a 52-year-old man. Chest CT and three-dimensional volume rendered reconstructed images revealed an oval, cystic mass with multiple nodular calcifications, centered in the left diaphragm crus. CT facilitated documentation of the healing process of this rare entity, revealing decrease in size and increase in internal density.

Index terms
Bronchogenic Cyst
Diaphragm
Multidetector Computed Tomography

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INTRODUCTION

Bronchogenic cysts are congenital abnormalities that arise from the ventral foregut. They occur most commonly within the mediastinum but rarely in the diaphragm. Up to now, there have been few reported cases of intradiaphragmatic bronchogenic cyst reported. On CT, bronchogenic cysts typically manifest as spherical masses of either water or soft-tissue attenuation (1, 2). In this report, we report a 52-year-old man who was admitted to our hospital for evaluation of dyspnea. He had a cystic mass in left diaphragm crus, with inner calcification, and peripheral and septal enhancement on CT scan. Follow up CT scans, revealing changes in the size and density of the mass, were believed to be documentation of the healing process of an infectious bronchogenic cyst.

CASE REPORT

A 52-year-old man was admitted to our hospital emergency room due to sudden onset dyspnea in recumbent positioning. On physical examination, he complained of shortness of breath and left chest pain with breathing motion. On the laboratory test, the oxygen (O_2) saturation of 92.2% (normal range 94–100%). White blood cell count (WBC, 15260/μL, normal range 3500–10000/μL), erythrocyte sedimentation rate (ESR, 26 mm/hr, normal range 0–20 mm/hr) and C-reactive protein (CRP, 10.51 mg/dL, normal range 0–0.5 mg/dL) were elevated.

Chest radiograph revealed blunting of the left costophrenic angle and increased opacity in the left lung parenchyma (Not shown). CT scan was performed using a 128-row detector CT scanner (Somatom Definition AS, Siemens Healthcare, Erlangen, Germany). The CT scan parameters were as follows: 120 kVp tube voltages, 100 mA tube current. Nonionic contrast

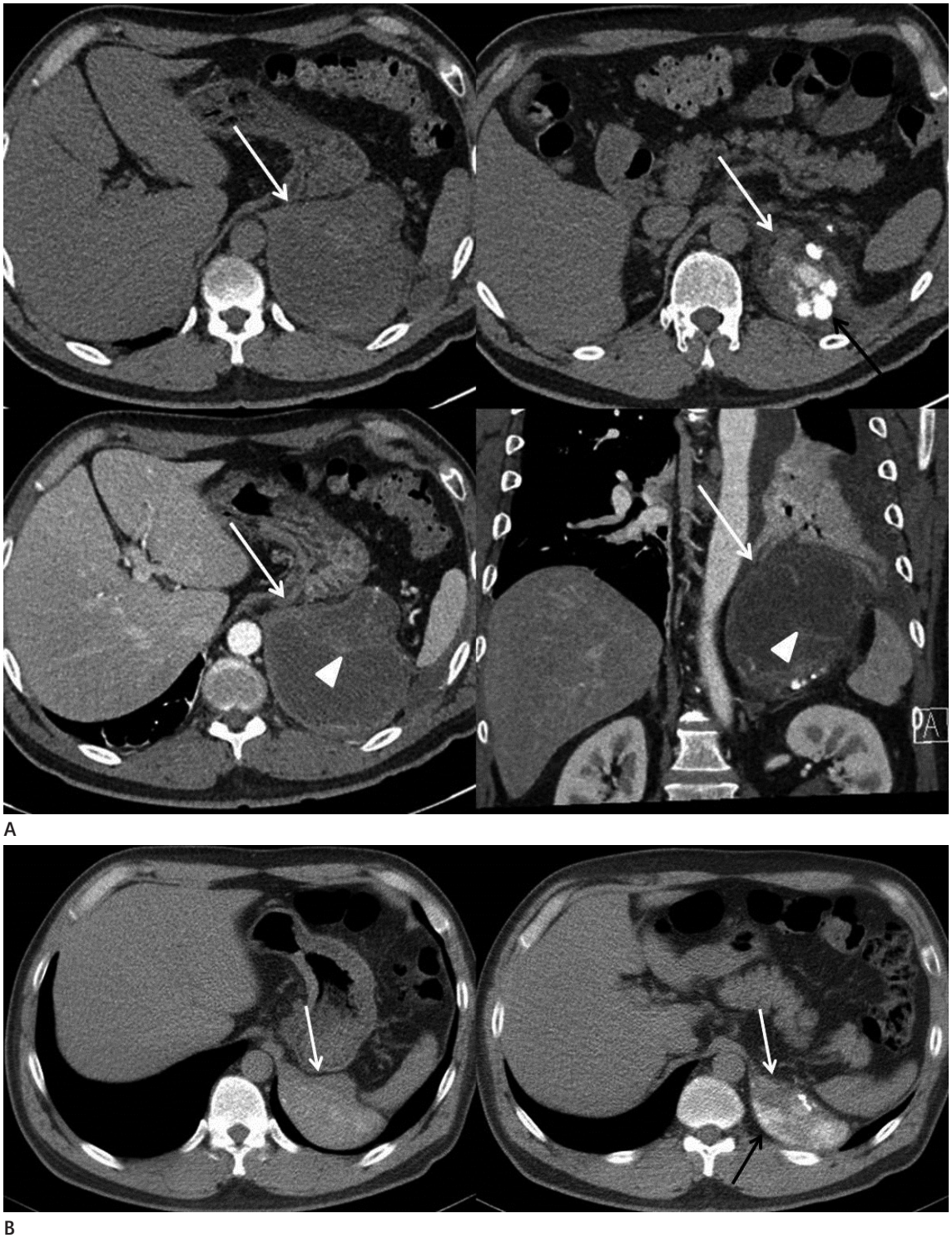

Fig. 1. Bronchogenic cyst in an intradiaphragmatic location in a 52-year-old man with dyspnea.
A. Chest CT shows an about 10 × 9.8 × 10.8 cm sized, oval mass across the left pleural cavity and left retroperitoneum on the axial and coronal reformatted images (white arrows). It has well defined margins and low attenuation of 18–21 Hounsfield unit, which suggests a cystic lesion. The center of the mass is in the left diaphragmatic crus. Contrast enhanced CT reveals peripheral enhancement and multiple enhancing inner septations (arrowheads, left lower and right lower). In the lower portion of the mass, multiple nodular calcifications are seen (black arrow, right upper). There are also pleural effusion and passive atelectasis.
B. On chest CT acquired at another facility approximately 2 years previously, a smooth margined, elliptical and heterogeneous density mass is seen in left diaphragmatic crus (white arrows). Multiple nodular calcifications are seen in the mass (black arrow, right panel).

material (120 mL) was administered at a rate of 2.5mL/s, followed by a 30 mL saline flush. Chest CT revealed a 10 × 9.8 × 10.8 cm sized, well defined, oval shaped, heterogeneous huge cystic mass, centered in the left diaphragm crus (Fig. 1A). The mass contained multiple nodular calcifications and multiple inner septations, which inferiorly abutted the left pleura. After intravenous contrast injection, peripheral and septal enhancement was apparent in the mass. In addition, CT scan revealed left pleural effusion and passive atelectasis of adjacent lung parenchyma, which was thought to be reactive change.

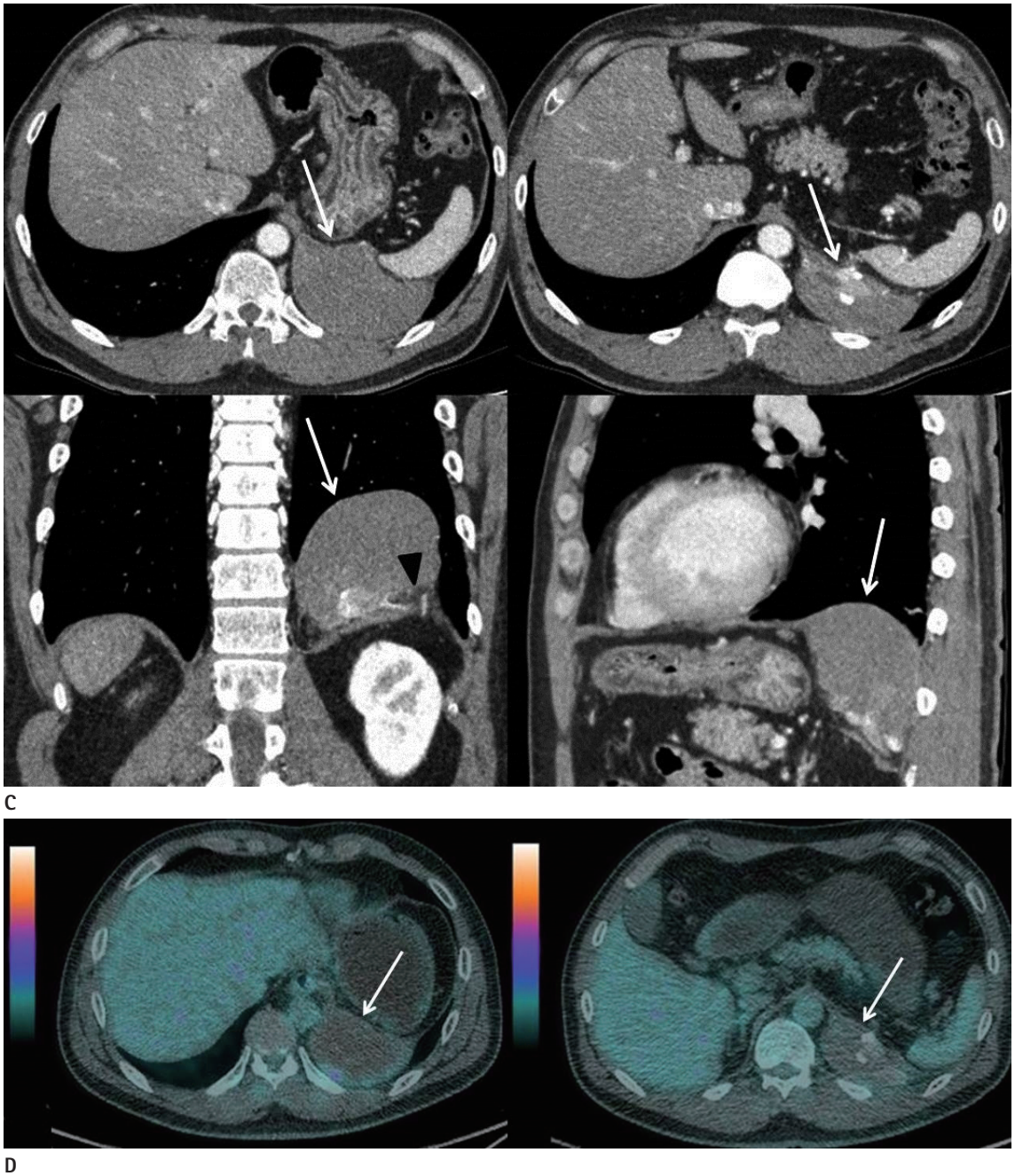

Fig. 1. Bronchogenic cyst in an intradiaphragmatic location in a 52-year-old man with dyspnea.
C. Two-week follow up chest CT scan reveals decreased size of the mass with heterogeneous density and disappearance of the inner cystic lesion (arrows). The mass still has a smooth margin but no significant enhancement. On coronal (left lower) and sagittal (right lower) reconstruction images, the mass splits the left diaphragm and small vessel, penetrating the diaphragm, supplies the mass (arrowhead, left lower). There was no evidence of previous pleural effusion and passive atelectasis in the left lung.
D. Fluorine-18 fluorodeoxyglucose PET/CT shows mild hypermetabolism in the posterior margin and internal metabolic defect in the mass (arrows).

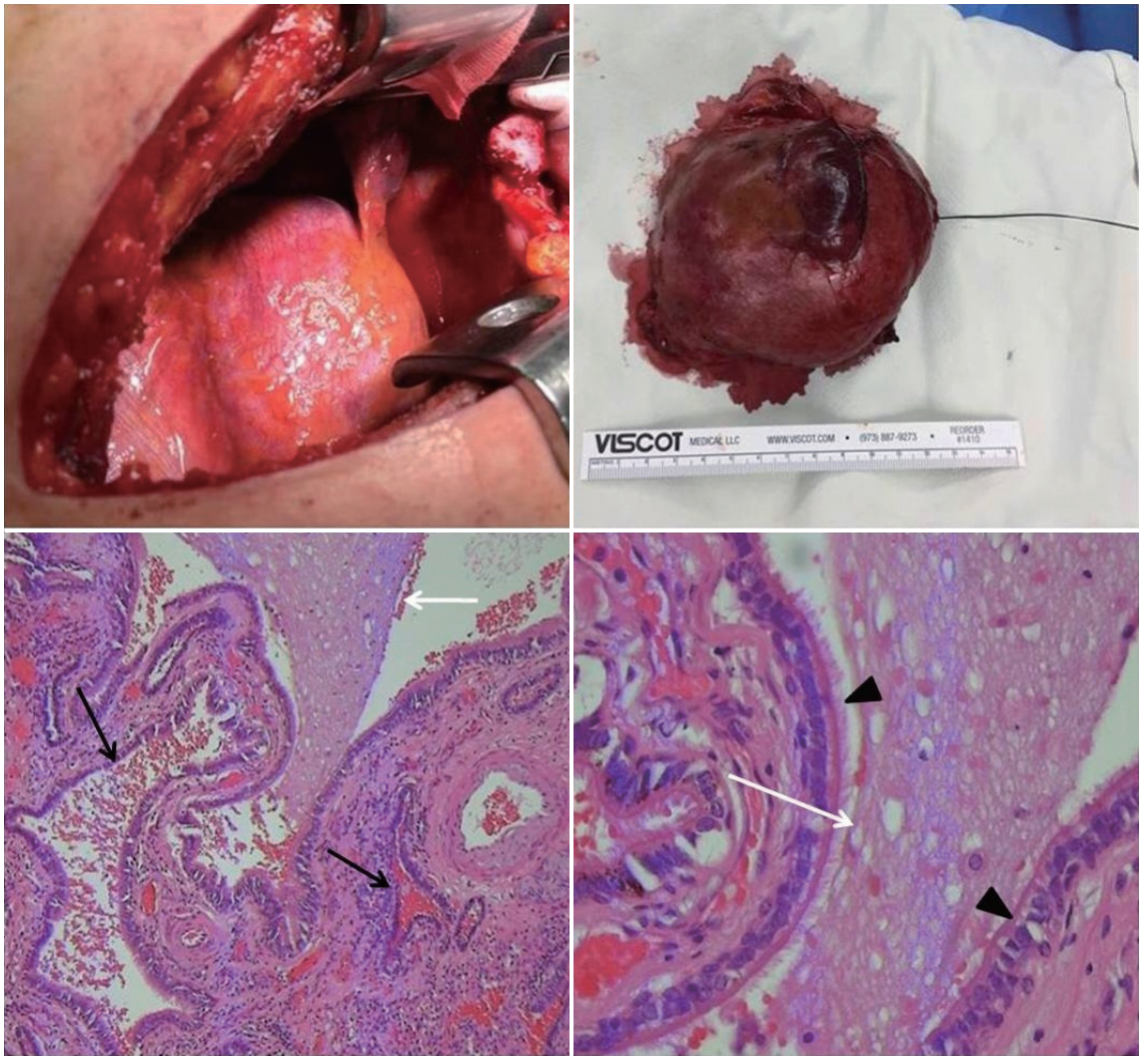

Fig. 1. Bronchogenic cyst in an intradiaphragmatic location in a 52-year-old man with dyspnea.
E. During surgery, the mass was located on posterolateral side of the left diaphragm (left upper). There was no evidence of invasion of the thoracic cavity. The mass was soft and had inner sticky materials greenish in color (right upper). In hematoxylin & eosin stain (× 100, left panel and × 400, right panel), multiple cystic spaces are seen (black arrows, left lower) and filled with gelatinous mucus-like fluid (white arrows) which are small structures in the normal bronchiole. Pseudostratified ciliated respiratory epithelium, which primarily lines the normal respiratory tract, is seen (arrowheads, right lower).

At initial hospitalization, lung abscess was clinically suspected, empirical antibiotics including Piperacillin/tazobactam [Tabaxin (Penmix Ltd., Jeong-dong, Seoul, Korea), 4.5 g/vial, three times per day] and Clindamycin [Fullgram (Samjin Pharm., Seoul, Korea), 600 mg/amp, three times a day] were prescribed for 5 days. On second admission day, laboratory test values were normalized (O_2 saturation: 94%, WBC: 6620/μL, ESR: 15 mm/hr, and CRP: 0.1 mg/dL).

We could receive a previous non-contrast chest CT scan of the patient performed at outside hospital about 2 years ago. This CT scan revealed a smooth, sharply margined, elliptical, homogeneous density mass with multiple calcifications in the same area (Fig. 1B). The size of the mass was significantly smaller than in the initial chest CT in our hospital. Considering the change in size, density and presence of enhancement, ultrasonography guided lung biopsy was performed under the suspicion of malignancy. However, no malignant tumor was detected on pathological analysis. The patient was discharged with a recommendation for two-week follow up chest CT.

The follow up chest CT revealed decreased size with disap-

pearance of the inner cystic lesion (Fig. 1C). The mass exhibited a smooth margin, homogeneous density without significant enhancement. A fluorine-18 fluorodeoxyglucose (^{18}F-FDG) PET-CT scan revealed mild marginal hypermetabolism and inner metabolic defect, which was interpreted to be likely benign nature (Fig. 1D).

However, surgical removal of the mass was performed to exclude the possibility of malignancy during his second hospitalization. The operators performed a lateral thoracostomy through the left 8th intercostal space. Grossly, the mass was located in the posterolateral side of left hemidiaphragm (Fig. 1E). The mass was soft and exhibited inner sticky material greenish in color. There was no invasion of the thoracic cavity. Final pathological diagnosis of the mass was intradiaphragmatic foregut cyst, consistent with bronchial cyst. Hematoxylin & eosin stain revealed pseudostratified ciliated respiratory epithelium which is lined mainly in normal respiratory tract (Fig. 1E).

The second follow up postoperative chest CT scan demonstrated no evidence of a remnant mass.

DISCUSSION

Bronchogenic cysts are congenital lesions that are believed to arise from an abnormally budding ventral foregut, which then develops into a blind ended fluid filled pouch (1). They occur between the 26th and 40th days of gestation. They are usually found in the mediastinum, near the tracheal carina, in 85% of patients, and 79% occur in middle mediastinum. They also may be found in the lung parenchyma, pleura, retroperitoneum, and neck (2). However, intradiaphragmatic bronchogenic cysts are exceedingly rare and only a few cases have been reported.

Symptoms are frequently nonspecific; however, when present, pain is the most common clinical symptom (1, 3). Other presenting symptoms may include fever due to infection and symptoms ascribable to pressure on adjacent structures (4).

The pathological hallmark of bronchogenic cysts is the presence of ciliated pseudostratified columnar epithelium, cartilage, and smooth muscle within the cyst wall. Grossly, there is a variable presentation, which likely contributes to their variable radiologic appearance (1).

CT findings of bronchogenic cyst have been well described in the literature. They are usually sharply margined with soft tissue or water attenuation, with cystic characteristics. Some bronchogenic cysts may have soft tissue attenuation, and contrast enhanced CT may help in distinguishing malignancy by the lack of enhancement. Ten percent of bronchogenic cysts can have calcification (4). In our patient, the mass was a well margined, cystic lesion in the left diaphragmatic crus; these characteristics could be regarded as typical findings of bronchogenic cyst retrogradely. On the other hand, the mass showed peripheral and septal enhancement, which should be regarded as a possibility of malignancy with cystic change. After ^{18}F-FDG PET/CT evaluation, and changes in size and attenuation in follow up chest CT, peripheral and septal enhancement of the mass were thought to be inflammatory changes rather than malignant enhancement. Therefore, we considered benign lesions in the differential diagnosis including hemangioma, rhabdomyoma, teratoma and hematoma. After confirming gross pathology, increased density of the mass on chest CT was believed to be due to the inner sticky materials.

The evaluation of bronchogenic cysts depends on the size of the cyst and patient symptoms. Small, asymptomatic cysts can be followed conservatively. However, enlargement of bronchogenic cysts over a span of years is typical, and rapid enlargement associated with pain indicates hemorrhage or infection. Because of their tendency to grow, bronchogenic cysts are traditionally treated using complete surgical resection (5). On occasion, bronchogenic cysts may harbor malignancy and, therefore, necessitate surgical resection for clear diagnosis (1).

There are few reported cases of intradiaphragmatic bronchogenic cysts, with most removed by surgical procedure after detection for accurate diagnosis. However, our case shows alterations of the lesion, from a soft tissue mass to a cystic lesion with enhancement. These changes suggest complications such as inflammation or malignant changes. In conclusion, awareness of an unusual location of bronchogenic cysts with variable CT findings, as in our case, would help in differential diagnosis and management of these thoracic lesions.

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횡경막 내에 위치한 기관지원성 낭종: 증례 보고

윤재홍[1] · 이기남[1]* · 강은주[1] · 김문성[1] · 최필조[2] · 노미숙[3]

기관지원성 낭종은 주로 종격동, 특히 기관 분기부나 중간 종격동에서 볼 수 있는 선천성 병변이다. 여기에 우리는 52세 남자에서 발생한 횡경막 내의 기관지원성 낭종과, 함께 동반된 감염성 합병증에 관한 매우 드문 증례에 대해 보고하고자 한다. 흉부 전산화단층촬영술과 3차원 볼륨 렌더링 영상에서 횡경막좌각을 중심으로 한 타원형의 낭종성 종괴와 내부의 다수의 결절성 석회화들이 관찰되었다. 전산화단층촬영술에서 크기감소와 내부 음영의 증가와 같은 이 드문 질환의 회복 과정을 확인할 수 있었다.

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