



## Case report

# Pulmonary hamartoma mimicking a mediastinal cyst-like lesion in a heavy smoker



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## ARTICLE INFO

## Keywords:

Pulmonary hamartoma  
Computed tomography  
Differential diagnosis  
Histology

## ABSTRACT

Pulmonary hamartoma (PH) is the most common benign tumor of the lung, typically presenting as a peripheral solitary nodule with round shape and smooth margins. The main computed tomography (CT) features that allow a confident diagnosis of PH are intranodular fat and popcorn-like calcifications. However, the presence of these features within PHs is variable. Thus, a reliable diagnosis of PH cannot be formulated in approximately 30% of cases. Furthermore, PHs may occasionally show atypical CT features.

The present article reports the case of a centrally located PH with an extremely rare and previously unreported CT presentation consisting of fluid attenuation, rim enhancement and thick enhancing septa that mimicked a mediastinal cyst-like lesion.

## 1. Introduction

Pulmonary hamartomas (PHs) constitute 6–8% of solitary lung nodules and account for 75% of all benign lesions [1,2]. PHs occur mostly as peripheral solitary nodules and are incidentally detected on routine radiological examinations, such as radiography and computed tomography (CT) [2,3]. CT is the diagnostic tool of choice in the study of the internal characteristics of pulmonary nodules. The main CT features suggestive of PH are intranodular fat and popcorn-like calcifications [3].

However, the presence of these features within PHs is variable and their frequency increases with the tumor's size [4]. Thus, a reliable diagnosis of PH cannot be formulated in approximately 30% of cases [2,3]. Furthermore, PHs may occasionally show atypical CT features [5].

The present article reports the case of a centrally located PH with an extremely rare and previously unreported CT presentation consisting of fluid attenuation, rim enhancement and thick enhancing septa that mimicked a mediastinal cyst-like lesion. The radiologic-pathologic correlation of this unusual presentation was also described.

## 2. Case report

A 59-year-old Caucasian woman with a 40 pack/year smoking history was referred to our radiology department to undergo an ultra-low-dose chest CT scan as part of the protocol of an interventional clinical study started in December 2017. This study, titled “Ultra-low-dose chest CT for lung nodules detection in high risk subjects”, was approved by our local ethics committee as a prospective analysis (NP 2851). Therefore, informed consent for the use of personal data of the patient for scientific purpose was obtained.

The patient, with stage IA chronic obstructive pulmonary disease, reported occasional nonproductive cough and initial shortness of breath on exertion.

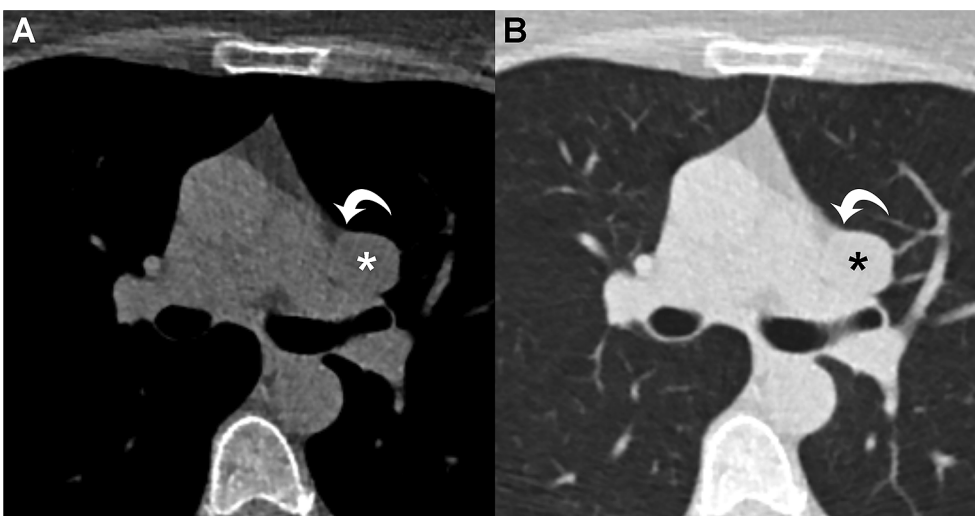
The ultra-low-dose CT scan revealed a round and smoothly marginated low-attenuation nodule (maximum diameter, 25 mm) located anterior to the left hilum in the phrenic nerve region (Fig. 1). This nodule created an obtuse angle with the lung; therefore, it was considered a mediastinal lesion. Based on the ultra-low-dose CT findings, our provisional diagnosis included bronchogenic cyst and tumors with cystic degeneration such as schwannoma and lymphadenopathy. Thus, a dynamic contrast-enhanced chest CT was performed one week later. The dynamic contrast-enhanced CT scan

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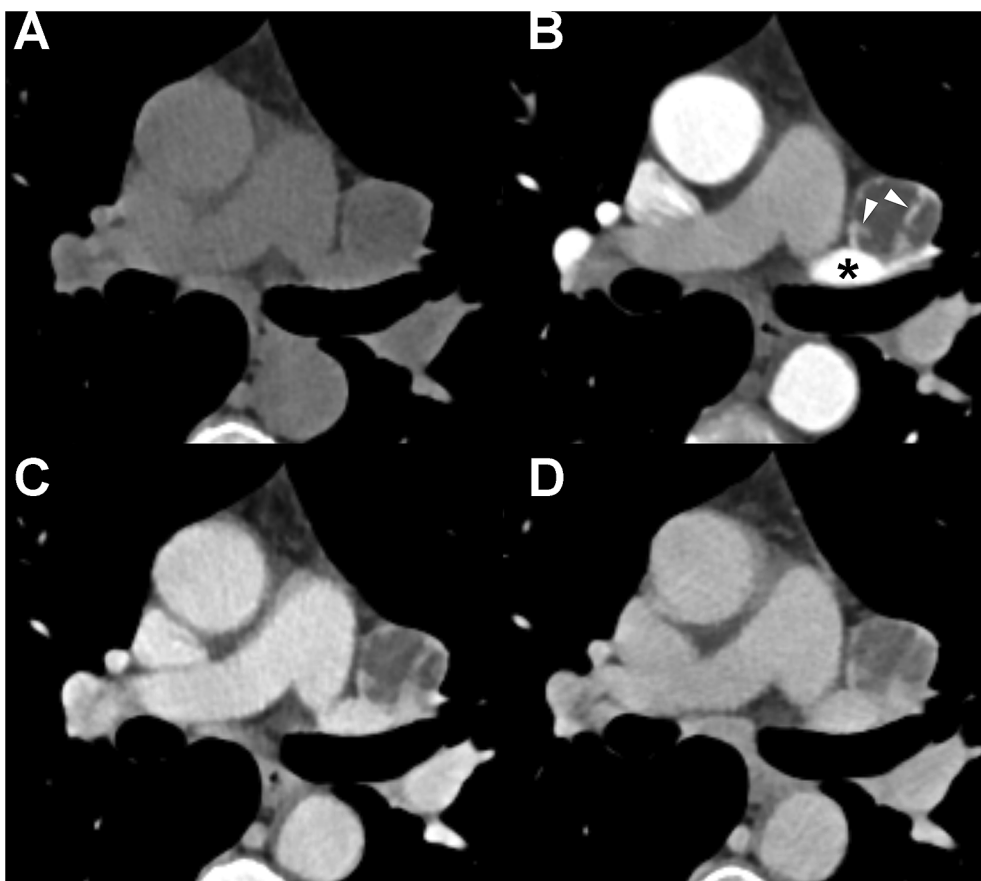
**Abbreviations list**

CT	computed tomography
FDG	fluorodeoxyglucose
PET	positron emission tomography
PH	pulmonary hamartoma

revealed that the nodule had predominant fluid attenuation (approximately 10 Hounsfield Units) with rim enhancement and thick hypervascularized septa and was closely abutted to the superior pulmonary vein (Fig. 2). Based on the site (phrenic region), shape (round), margins (smooth) and internal characteristics (fluid attenuation, rim enhancement and thick enhancing septa) of the nodule, our provisional diagnosis was schwannoma with cystic degeneration originating from the phrenic nerve. However, no sign of diaphragmatic eventration was observed.



**Fig. 1.** Axial ultra-low-dose CT images with mediastinal window (A) and lung window (B) reveal a round and smoothly margined low-attenuation nodule located anterior to the left hilum (asterisks). The obtuse angle formed between the nodule and the adjacent lung parenchyma (arrows) suggests a mediastinal lesion.



**Fig. 2.** Axial dynamic contrast-enhanced CT images obtained before contrast medium injection (A), at 30 seconds (B), 1 minute (C) and 3 minutes (D) after contrast medium injection show that the nodule has predominant fluid attenuation with rim enhancement and thick hypervascularized septa (arrowheads in B). Note that the lesion is closely abutted to the superior pulmonary vein (asterisk in B).

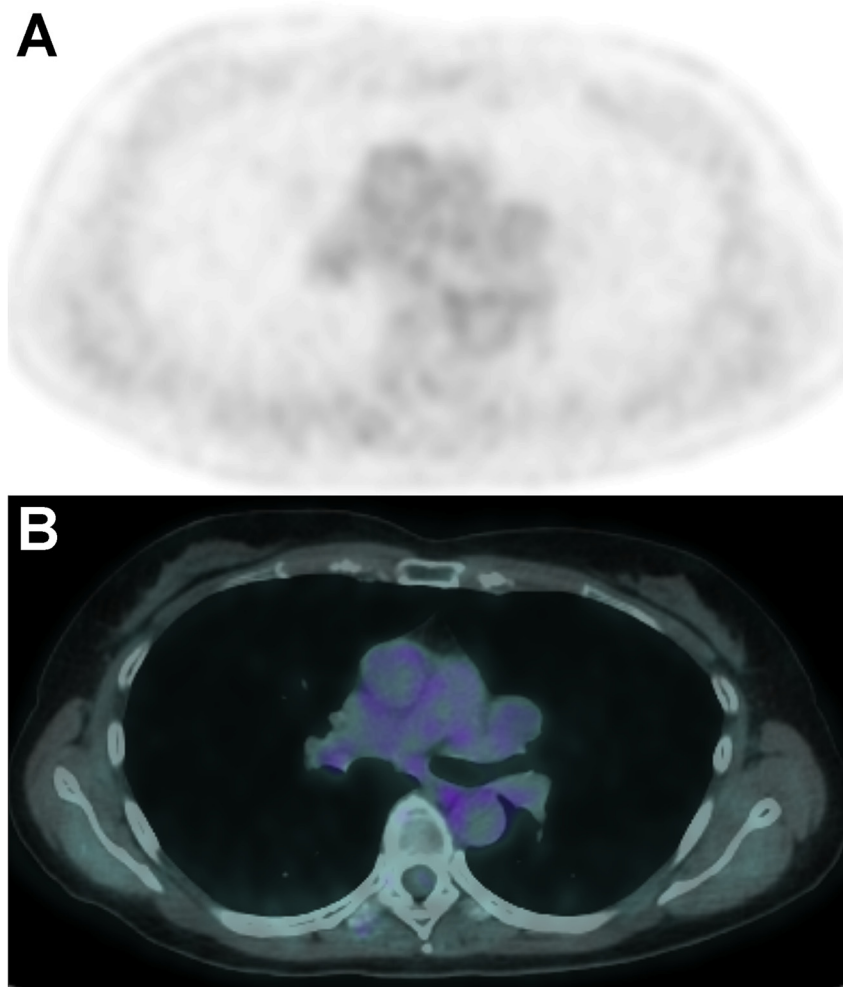


Fig. 3. Axial PET (A) and fused PET/CT (B) images show no pathological FDG uptake within the nodule.

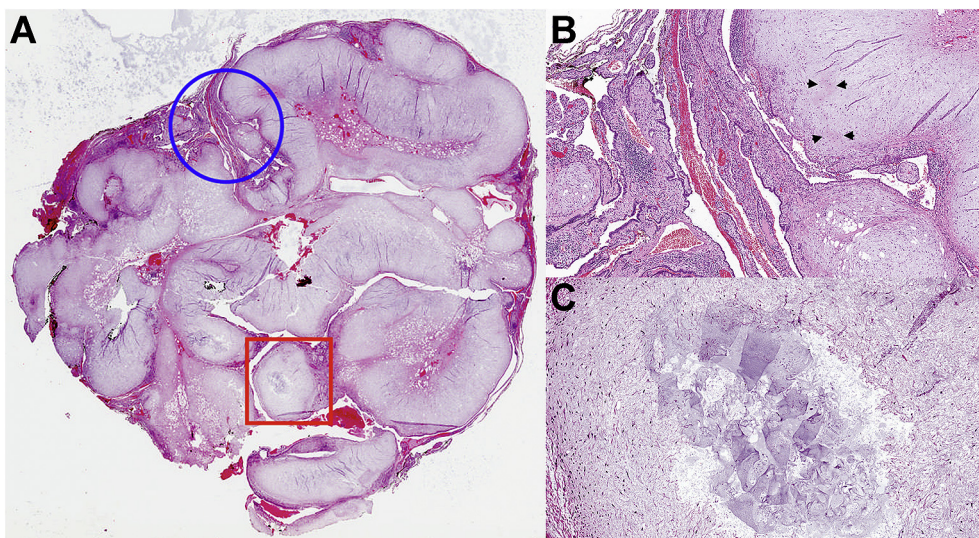


Fig. 4. Histological hematoxylin and eosin images of the pulmonary hamartoma at actual size (A), 20 × (B) and 40 × (C) showing the predominant myxomatous component and clefts lined by respiratory epithelium. The blue circle and red square in A indicate the area of magnification displayed in B and C, respectively. The magnification in B shows a cleft lined by compressed respiratory epithelium with increased vascularity. Note the minimal area of cartilage (arrows) within the nodule. The magnification in C depicts the predominant myxomatous component with a central pseudocystic appearance.

To evaluate the metabolic activity of the nodule, a positron emission tomography (PET)-CT scan was performed. No pathological fluorodeoxyglucose (FDG) uptake was observed within the lesion (Fig. 3) or in the rest of the body.

Considering our provisional diagnosis and the close relationship of the lesion with the superior pulmonary vein, surgical resection was

recommended. One month later, the patient underwent video-assisted thoracoscopic surgery, and a well-defined subpleural pulmonary nodule, with increased vascularity, was found between the mediastinum and the left pulmonary hilum. The visceral and mediastinal pleura were compressed, and the phrenic nerve was displaced by the nodule. The lesion was completely resected (enucleated) with phrenic nerve sparing.

**Table 1**  
Atypical CT presentations of pulmonary hamartomas reported in the literature.

Authors	Year	Atypical CT feature
Tsitouridis et al. [9]	2010	Endobronchial nodule with mediastinal extension
Huang CC et al. [10]	2012	Pure ground-glass nodule with air cyst formation
Ladeira I et al. [11]	2015	Mediastinal mass
Sakamoto et al. [12]	2015	Pulmonary nodule with pleural dissemination
Minegishi K et al. [13]	2016	Endobronchial nodule with air crescent sign
Li L et al. [14]	2016	Multiple pulmonary nodules
Mertoğlu A et al. [15]	2017	Multiple endobronchial nodules
Fasanya AA et al. [16]	2017	Multiple pulmonary nodules and cysts

Histological examination revealed a PH with predominant myxomatous component, minimal area of cartilage and clefts lined by respiratory epithelium that were compressed and hypervascularized (Fig. 4).

### 3. Discussion

PH is the most common benign tumor of the lung, typically presenting as a peripheral solitary nodule with round shape and smooth margins [4]. Endobronchial hamartomas occur much less frequently and represent 1.4–10% of PHs [6].

PH is more frequent in men than in women, with a male-to-female ratio of 2:1 or 3:1 and a peak incidence during the sixth decade of life. These benign lesions are very slow-growing tumors with a size less than 4 cm, although rapid growth [7] and giant mass have been reported in the literature [8]. The main CT features that allow a confident diagnosis of PH are intranodular fat and popcorn-like calcifications [2–4]. However, the literature reports that less than 50% of PHs show focal fat content [1,2], and only 20% of PHs present popcorn-like calcifications [3]. Furthermore, the literature reports that PHs may occasionally show atypical CT features [5]. In a literature search of the PubMed database for publications from the past 10 years using the key words pulmonary hamartoma and CT, we identified 8 English-language case reports in which atypical CT presentations of PHs were described (Table 1) [9–16].

The PET-CT imaging features of PHs are less known; however, the literature reports that these lesions generally do not show a pathological FDG uptake [2].

Histologically, PHs show a variable mixture of cartilage, myxomatous tissue, fat and smooth muscle [6]. A peculiar feature of PH is the presence of internal cleftlike spaces (entrapped airways) lined by respiratory epithelium, mainly located in the periphery [6].

In our case, PH was centrally located (phrenic region) and had an atypical CT presentation consisting of fluid consisting of fluid attenuation, rim enhancement and thick enhancing septa that seemed to be similar to a mediastinal tumor with cystic degeneration. A typical non-pathological FDG uptake was observed within the lesion.

Similarly to the typical form, histological examination revealed the presence of cleftlike spaces lined by respiratory epithelium. However, our case showed a predominant myxomatous component with only a minimal area of cartilage.

To the best of our knowledge, this article describes the first reported case of a centrally located PH with atypical CT features mimicking a mediastinal cyst-like lesion. In the present case, the fluid attenuation detected on CT images corresponded to the predominant myxomatous component with areas of pseudocystic appearance (Fig. 4C). The rim enhancement and peripheral enhancing septa corresponded to clefts lined by hypervascularized respiratory epithelium (Fig. 4B). These latter CT findings appeared to be similar to those observed on magnetic resonance imaging in previous reports [17,18]. In our case, the clefts lined by respiratory epithelium are so prominent on dynamic contrast-enhanced CT images due to their increased vascularity probably related to the central location of the PH.

### 4. Conclusion

The present case involves a 59-year-old, heavy-smoking, Caucasian woman who was diagnosed with centrally located pulmonary hamartoma mimicking a mediastinal lesion. This centrally located pulmonary hamartoma showed an extremely rare, and previously unreported, CT presentation that seemed to be similar to a tumor with cystic degeneration. The imaging findings observed in this case suggest that PH may be included in the differential diagnosis of a centrally located nodule with fluid attenuation, rim enhancement and thick enhancing septa.

### Conflict of interest

The authors have no financial or other conflict of interest.

### Appendix A. Supplementary data

Supplementary data related to this article can be found at <https://doi.org/10.1016/j.rmcr.2018.08.007>.

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