CASE REPORT

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Pulmonary cavernous hemangioma: a case report



Wei Li¹, Xingxing Zheng¹, Hongzhe Tian¹, Yu Xi¹ and Yuhao Liu^{1*}

Abstract

Background Cavernous hemangiomas can occur in various internal organs like the liver, kidney, bladder, and skin, or even in subcutaneous tissues. However, they rarely occur in the lungs, making pulmonary cavernous hemangiomas (PCH) an uncommon finding. Herein, we report a rare case of pulmonary cavernous hemangioma that was surgically resected.

Case presentation A 16-year-old adolescent patient was diagnosed with a pulmonary cavernous hemangioma during a physical examination. During the entry physical examination, a chest X-ray showed shadows in the upper lobe of the right lung. The patient was admitted to the Thoracic Surgery Department for further assessment. The patient had a one-year history of smoking; however, he did not experience any clinical symptoms related to respiratory diseases. Among tumor markers assessed, only the cytokeratin-19 fragment (CYFRA21-1) was elevated. Chest computed tomography (CT) showed irregular soft tissue lesions in the upper lobe of the right lung. The lesion presented as a shallow lobe with clear boundaries, accompanied by the presence of spicules around it. The lesion showed mild to moderate uniform enhancement in the arterial phase and slightly reduced in the venous phase on imaging. The lesion was anatomically close to adjacent bronchus and blood vessels. In addition, the patient had multiple enlarged lymph nodes in the mediastinum. In summary, the thoracic surgeon could not rule out the possibility of lung malignant tumors and chose to proceed with surgical excision to secure a conclusive diagnosis. Post-operative histological examination showed that the lesion consisted of dilated nodular hyperplasia rich in red blood cells, leading to a diagnosis of PCH. No recurrence has been observed since postoperative follow-up.

Case presentation We reported a rare PCH case, reviewed the clinical features, imaging findings, histopathological features, and treatment options of PCH based on relevant literature, and gained a further understanding of PCH.

Keywords Pulmonary cavernous hemangioma (PCH), Solitary nodules in the lung, Chest computed tomography (CT), Video-assisted thoracoscopic surgery (VATS), Case report

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Background

Pulmonary cavernous hemangioma is a very rare benign lesion caused by congenital pulmonary vascular malformation that can sometimes create a direct arteriovenous communication causing a pulmonary circuit shunt. It is composed of large, dilated vascular spaces, essentially "caverns", lined by a single layer of endothelial cells and filled with blood, with only a small amount of supporting connective tissue between them [1]. Cavernous hemangiomas can occur in various internal organs such as the liver, kidney, bladder, and skin or subcutaneous tissues, however, their primary occurrence in the lungs is considered very rare [2, 3]. PCH do not present with specific clinical symptoms [4] and are easily misdiagnosed as lung conditions, including lung cancer, tuberculosis, or pulmonary cysts, during routine X-ray and CT examinations. This work retrospectively analyzes the clinical data and CT imaging findings of patients diagnosed with PCH through surgical and pathological confirmation at our hospital. The aim is to improve radiologists' understanding of PCH, thereby minimizing the occurrences of misdiagnosis and missed diagnosis.

Case presentation

A 16-year-old male was admitted to the thoracic surgery department for further diagnosis and treatment after a chest X-ray examination revealed a lesion in the upper lobe of the right lung. The patient presented with no signs of cough or sputum production, he did not experience chest tightness or dyspnea and reported no fatigue or night sweats. Additionally, there were no indications of chills, fever, hemoptysis, or chest discomfort. Overall, the patient did not present with any clinical symptoms. During the specialized examination, the respiratory sounds of both lungs were coarse, with no audible dry or wet rales. The patient had a 1-year smoking history, smoking an average of 3–4 cigarettes per day.

The laboratory test results for liver function, kidney function, electrolytes, blood glucose, and coagulation showed no abnormalities. Among tumor markers analyzed, only the CYFRA21-1 was elevated at 2.60 ng/ml (normal range :0-2.08 ng/ml).

Chest CT examination (Fig. 1A-I) showed irregular soft tissue lesions in the upper lobe of the right lung, approximately 2.5 cm×1.7 cm×1.5 cm in size. The lesions presented as shallow lobules with clear boundaries, accompanied by fine spicules surrounding them. On contrast-enhanced images, the lesions were mildly to moderately uniformly enhanced in the arterial phase, with a measured CT value of approximately 60HU. The degree of enhancement in the venous phase was slightly reduced, with a CT value of about 53HU. The lesion was anatomically close to the adjacent bronchus and blood vessels. There were multiple nodules in the mediastinum, with the largest measuring approximately 1.5 cm in short diameter, with slight enhancement in the arterial phase, with a CT value of about 49HU, and reduced enhancement in the venous phase, with a CT value of about 39HU. Bronchoscopy showed inflammatory changes in the trachea and bronchial mucosa, with no evidence of a current active bacterial, viral, or fungal infection found in the trachea lumen. A needle aspiration biopsy of the mediastinal lymph node showed no presence of tumor cells.

The patient in this case presented with a history of smoking, and the.

chest CT showed a solid hypervascular mass accompanied by elevated levels of CYFRA21-1. From these findings, lung cancer cannot be ruled out. Therefore, the patient underwent video-assisted thoracoscopic surgery (VATS) to remove the right upper lobe of the lung and also remove nearby lymph nodes. During the operation, it was found that the development of the lung fissure was poor, and the mass located in the upper lobe of the right lung was hard. Intraoperative freezing suggested benign lesions and no malignant features were observed. Therefore, the thoracic surgeon switched to wedge resection of the upper lobe of the right lung using VAT, which minimized patient trauma by only removing a small, wedgeshaped piece of lung tissue, thus avoiding the need for a more extensive lymph node dissection that would be required in a larger surgical procedure like a lobectomy.

Postoperative histological examination of the specimen (Fig. 2A-C) showed clear boundaries between the lesion area and its surroundings, which were characterized by dilated luminal nodular hyperplasia rich in red cells.Surrounding the nodules were damaged alveolar walls, bronchi, and proliferating collagen fiber tissue, as well as some lymphoid tissue hyperplasia. Immunohistochemical examination showed that the dilated lumen rich in red blood cells was Vimentin (+++), CD31 (+++), CK/ EMA/CK7/TTF-1/CD68/CD34/Syn/CgA/PR (-), Ki-67 (extremely low).

The patient was ultimately diagnosed with pulmonary cavernous hemangioma. The patient has made a satisfactory recovery following the surgical procedure and has not encountered any recurrence during subsequent follow-up assessments.

Discussion

Hemangioma is a benign vascular tumor [5], usually classified into capillary hemangioma, cavernous hemangioma, large-vessel hemangioma, skeletal muscle hemangioma, intravascular papillary endothelial hyperplasia, spindle cell hemangioma, and hobnail hemangioma based on their clinical presentation and the size of the blood vessels involved in the tumor. Among them, cavernous hemangioma is histologically characterized



Fig. 1 Chest CT examination findings: irregular soft tissue lesions in the upper lobe of the right lung (Red arrow, Fig. 1**A**), approximately 2.5 cm×1.7 cm×1.5 cm in size (Fig. 1**B** and **C**). The lesions presented as shallow lobules with clear boundaries, accompanied by fine spicules surrounding them. On contrast-enhanced images, the lesions were mildly to moderately uniformly enhanced in the arterial phase (Fig. 1**E**), with a measured CT value of approximately 60HU. The degree of enhancement in the venous phase (Fig. 1**F**) was slightly reduced, with a CT value of about 53HU. There were multiple nodules in the mediastinum, with the largest measuring approximately 1.5 cm in short diameter (Fig. 1**D**), with slight enhancement in the arterial phase (Fig. 1**E**), with a CT value of about 39HU.MinIP (Fig. 1**G**), MIP (Fig. 1**H**), VR (Fig. 11) showed: The lesion was anatomically close to the adjacent bronchus and blood vessels



Fig. 2 Postoperative histological examination of the specimen findings: Macroscopic findings of the resected specimen (Fig. 2A): the lesions were nonencapsulated nodules with dark red surface.HE staining microscope 200 times (medium magnification) (Fig. 2B), HE staining under microscope at 400 times (high magnification) (Fig. 2C) showed: Dilated lumens rich in red cells were seen within the lesion ((Black arrow, Fig. 2B and C)

by the presence of large, dilated vascular channels that essentially manifest as "capillary tangles" or markedly dilated vascular Spaces.Cavernous hemangiomas can occur in various parts of the body but are rarely found in the lungs. The exact etiology of PCH is still unknown, and it has been suggested that they are congenital vascular anomalies that may be caused by gene loss-of-function mutations [6]. PCH can present as single or multiple lesions. When multiple lesions are present, PCH is often associated with hereditary hemorrhagic telangiectasia (HHT) disease.

PCH can often arise at various points within the lower respiratory tract, including the lung parenchyma, airways, and even the bronchioles but does not extend into the bronchial lumen. According to available data [2, 7], pulmonary cavernous hemangiomas can occur at any age, and while some sources suggest a slight female predominance, there is no definitive data on a significant gender difference in their occurrence; additionally, while a right lung predilection may exist, the reported ratio of 2:1 between right and left lungs is not widely established, and the incidence across different lung lobes is generally considered equal. In the research conducted by Ishikawa [8], Zhuang [9], Miyamoto [10], and Takashi Ibe [1], it was found that most PCH patients exhibit no symptoms or may present with non-specific respiratory symptoms. The lung lesions are usually incidentally discovered during routine physical examinations. However, over time, PCH patients may develop associated symptoms, mainly manifested as cough, sputum, bloody sputum, hemoptysis, chest pain, sudden breathing difficulties, or intrathoracic bleeding. The symptoms are related to its location, its size, the number of lesions present, and whether the blood vessels within the hemangioma are degenerating. The site of occurrence of the case in this article is consistent with the literature, but the gender of incidence is inconsistent with the literature, which may be attributed to the uniqueness of this individual case. Additionally, the lesions identified in this case were discovered during a physical examination, and the patient exhibited no clinical symptoms, consistent with previous reports.

Preoperative diagnosis of PCH is difficult because most auxiliary examinations lack specificity and misdiagnosis is common. PCH is often confused with other lung diseases such as lung cancer, tuberculosis, inflammatory pseudotumor, and hamartoma [11]. On chest X-ray, PCH appears as a circular or quasi-circular mass shadow in the lungs, with clear contours, smooth edges, and uniform density. On chest CT scans, PCH presents as isolated or multiple nodular lesions, with dotted, small nodular, and needle-like calcifications visible inside. Calcifications within a vascular tumors is usually due to the formation of a primary venous thrombus in the blood vessel of the tumor with subsequent calcium deposition on the thrombus, however, the occurrence rate remains below 10%. In the case presented here, no calcified thrombus was found within the lesion. Research [12] has found that in organs with abundant blood supply such as the lungs, spleen, and liver, the incidence of calcified thrombus is relatively low, while calcified thrombus is more common in vascular malformations that occur in subcutaneous and muscular areas. From our findings, contrast-enhanced CT revealed mild to moderate enhancement in the arterial phase, with a CT value of approximately 30-60 HU, which is lower than the CT value of arteriovenous fistula. The degree of venous phase enhancement was slightly reduced, showing equal or slightly higher density. However, if the tumor lacks large supply vessels, exhibits no clear arteriovenous shunting, and demonstrates a slow blood flow velocity, the tumor enhancement will not be significant [13]. In this case, the arterial phase of the lesion showed mild to moderate enhancement, while the degree of enhancement in the venous phase was slightly reduced, which is consistent with the literature. We found that compared to cavernous hemangiomas involving other solid organs such as the liver, pulmonary cavernous hemangiomas do not show the same characteristic centripetal enhancement pattern, making it harder to diagnose them before surgery. Bae et al. [2] proposed the usefulness of dual-layer spectral CT in the diagnosis of PCH, especially for lesions with small volume, high density, and unclear enhancement patterns on conventional CT images. In this examination, iodine density images obtained from spectral-based images clearly showed both the presence and distribution of iodine absorption within the lesion, indicating blood supply. The research results indicate that [2] the iodine distribution pattern in PCH closely resembles the contrast enhancement pattern observed in hepatic cavernous hemangiomas, characterized by either peripheral nodular enhancement or complete enhancement with a gradual filling process. PET/CT may also assist in diagnosing PCHs, which typically do not demonstrate elevated uptake of 18 F-FDG and 11 C-choline, two widely used oncologic radiotracers. This capability allows for differentiation from malignant tumors and specific pulmonary inflammatory lesions that display increased uptake [14]. The presence of numerous blood vessels in PCH makes needle biopsy an unsuitable preoperative diagnostic technique, as it may lead to severe bleeding that could be life-threatening.

In summary, PCH presents as single or multiple round or ovoid masses in the lung with well-defined boundaries and mild to moderate enhancement. However, accurate diagnosis of PCH is challenging when the lesion appears atypical or invades adjacent tissues, resulting in blurred borders and irregular morphology, and thus may be confused with malignant lesions. The presence of calcified thrombus, pampiniform growth pattern and aberrant draining veins are relatively specific in diagnosing PCH [15]. Magnetic resonance imaging can be helpful in CT suspected cases of PCH. Markedly high intensity on fat suppression T2-weighted image might be a characteristic feature of PCH [16]. PCH needs to be distinguished from the following diseases:

Hamartoma

Pulmonary hamartoma is the most common benign tumor of the lung, which mostly occurs in middle-aged and elderly men. The typical CT findings were round or round-like masses, mostly less than 2.5 cm in diameter, with clear boundaries [17] and no spicules. Fat and popcorn calcification were characteristic features of the lesions.

Tuberculosis ball

Most patients with pulmonary tuberculosis had clinical symptoms such as low fever, night sweats and weight loss. The tuberculoma was mainly located in the posterior segment of the upper lobe and the dorsal segment of the lower lobe. The tuberculoma was round or round-like, with uneven density, calcification, satellite lesions around, and no enhancement or only marginal enhancement.

Inflammatory pseudotumor

It is common in young and middle-aged women with symptoms of pulmonary infection and a history of recurrent infection. It was often located under the pleura, round, oval or mass shaped, with clear boundary, thick and long burr, peach tip like protrusion, and some straight edge like knife cutting, uneven density, and could be significantly enhanced.

Lung cancer

Lung cancer is the most common primary malignant tumor of the lung, originating from the bronchial epithelium, glandular epithelium, or alveolar epithelium. PCH is often distinguished from peripheral lung cancer.Peripheral lung cancer presents as a single nodule or mass within the lung, appearing as a round or irregular shape, often accompanied by 'lobulation' and 'spiculation'.When close to the pleura, pleural indentation can be observed.The density is uneven, and cancerous cavities and calcification can be seen within the lesion.Enhancement scans show uniform or non-uniform enhancement.

The definitive diagnosis of PCHs depends on postoperative histopathological findings. PCH is generally characterized by unencapsulated nodules that exhibit a dark red exterior and a cross-section resembling a honeycomb. Microscopic examination reveals that it is composed of dilated blood sinuses or anastomosed capillary lumens. Endothelial cells cover the blood sinuses, while lymphocytes and a small amount of fibrous tissue septa surround the sinuses [18]. There are red blood cells and macrophages that engulf hemosiderin particles in the lumen. Immunohistochemical endothelial cell markers CD31, CD34, and FV VIII were positively expressed, while TTF-1 was negative.

For early-stage, small, asymptomatic, and diagnosed PCH, diligent monitoring may be appropriate. However, in cases where symptoms or diagnosis are unclear and the possibility of malignant tumors cannot be ruled out (as in this case here), surgical intervention should be considered [19]. PCH with larger lesions carries a risk of spontaneous rupture, leading to severe bleeding or hemoptysis. Therefore, timely surgical treatment is crucial once diagnosed [20]. Given the extensive connection between cavernous hemangioma and the surrounding vascular system, this may lead to uncontrollable bleeding. In view of this, it is not advisable to remove the lesion alone. Lung lobectomy or segmental resection should be considered and the lesion should be enlarged to avoid severe bleeding. However, for small vascular tumors located in the pleura or peripheral lungs, wedge resection can be considered. Intraoperative frozen section can help avoid unnecessary traumatic lymph node dissection. So far, there have been no reports of recurrence after complete resection. Although the disease is generally benign, it can rarely become malignant.Postoperative follow-up is essential to monitor recurrence. We recommend that a follow-up examination be conducted once a year after surgery and continue for at least 10 years.

Limitations

This study has the following limitations: Firstly, the scope of diagnostic investigations available onsite at our healthcare facility was not exhaustive. In future research, hospitals will have more experience in conducting more comprehensive examinations for similar patients. Whats more, given the rarity of PCH, our study is based on a single case, which limits the generalizability of our findings. Future studies with a larger sample size and access to a broader range of diagnostic tools may provide a more comprehensive understanding of PCH and its management.

Conclusions

In short, PCH is a rare benign disease that occurs in the lungs. Preoperative diagnosis is difficult due to the lack of specific clinical symptoms and imaging findings. In future work, patients aged ≤ 20 years old who present with isolated or multiple blood-supply nodules in the lungs will be selected for study. During examination, after excluding metastatic lesions, more consideration should be given to the potential presence of congenital pulmonary vascular lesions, such as PCH, and pulmonary arteriovenous malformations. Combining double-layer spectral CT with PET-CT can offer significant diagnostic value by allowing for more precise tissue characterization, which can lead to better treatment planning and potentially avoid excessive treatment. Close monitoring may be conducted for small, asymptomatic, and clearly diagnosed PCH. In cases where the diagnosis remains uncertain, surgical resection should be the preferred treatment option. Should the tumor be identified as a PCH during the

surgical procedure, the extent of resection may be minimized, and lymph node dissection omitted to reduce potential harm to the patient.For future studies, the genetic basis of PCH and advances in non-invasive diagnostic imaging will be further investigated.

Abbreviations

PCH	Pulmonary cavernous hemangioma
CYFRA21-1	Cytokeratin-19-fragment
CT	Computed tomography
VATS	Video-assisted thoracoscopic surgery
HHT	Hereditary hemorrhagic telangiectasia
MinIP	Minimum Intensity Projection
MIP	maximal intensity projection
VR	Volume rendering
HE	Hematoxylin - eosin staining

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Author contributions

WL: conceived the study, analyzed the data, and drafted/revised the manuscript. YHL: analyzed the data and edited the manuscript. XXZ: collected data and reviewed the manuscript.HZT and YX: reviewed the manuscript. All authors read and approved the final manuscript.

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Data availability

Data is provided within the manuscript.

Declarations

Ethics approval and consent to participate

Our study followed the Declaration of Helsinki and it was approved by the Ethics Committee of the Baoji Central Hospital (No. BZYL2022-14) and the requirement for informed consent from the patients was waived.

Consent for publication

Consent to publish the data has been obtained from the parent.

Competing interests

The authors declare no competing interests.

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