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CASE REPORT

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A case of resected pulmonary capillary hemangioma with a literature review

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Abstract

Pulmonary hemangiomas are benign, relatively rare tumours. Because computed tomography (CT) findings show a variety of images, it is often difficult to distinguish hemangiomas from lung cancer and other benign tumours. We report a 63-year-old man who was diagnosed with a pulmonary capillary hemangioma (PCH). A right lung basal segmentectomy was performed for diagnosis and treatment. On chest CT, the lesion was shown to be a solid nodule with contrast-enhanced margins. This finding was thought to reflect the dense vascular hyperplasia of the central part of the tumour based on the pathologic findings. Although few studies involving PCH have referred to contrast-enhanced CT, the findings of contrast-enhanced CT might be a valuable indicator for diagnosing PCH.

KEYWORDS

benign lung tumor, pulmonary capillary hemangioma, pulmonary hemangioma, solid nodule, surgery

INTRODUCTION

Pulmonary hemangiomas are rare, benign tumours that are often incidental findings in asymptomatic patients. On chest computed tomography (CT), pulmonary hemangiomas present as pure ground-glass (GGNs), partially solid (PSNs), or solid nodules, which makes pulmonary hemangiomas difficult to distinguish from other masses, such as early-stage lung cancer. We report a patient with a pulmonary capillary hemangioma (PCH) and review the relevant literature.

CASE REPORT

A 63-year-old man was referred to the hospital for evaluation of an abnormal shadow noted on a chest X-ray during a health check-up. The patient had hypertension and hyperuricemia. He smoked 20 cigarettes per day for 20 years but quit smoking 23 years ago. He was a factory worker with no history of dust exposure. Blood tests, including tumour markers (CEA, SLX, SCC, CYFRA, and ProGRP), were negative. A chest CT showed a lobular solid nodule measuring 1.9 × 1.6 × 1.6 cm in the lower lobe of the right lung (Figure 1). An air bronchogram was demonstrated on the lower edge of the nodule (Figure 1). The edges of the tumour had minimal contrast uptake. The Hounsfield units of the tumour edge were 34 at 60 s and 56 at 100 s (Figure 2). On positron emission tomography-CT (PET-CT), the standardized uptake value of the nodule was up to 1.6, which was suggestive of an early-stage adenocarcinoma (Figure 3). No hilar or mediastinal lymph node metastases and no distant metastases were detected. A bronchoscopy was performed but was non-diagnostic.

Based on the imaging findings, we suspected a low-grade mucinous adenocarcinoma and performed a right lower basal segmentectomy for diagnostic and therapeutic purposes. The lesion was present at the S10 segment and pleural changes were observed. The nodule was soft on palpation. The operative time was 4 h and 34 min, and the blood loss was 134 mL.

On the cut surface, a circumscribed dark red nodule was demonstrated, measuring 1.5 × 0.7 cm (Figure 4). Based on microscopic findings, the tumour consisted of a benign proliferation of capillary-sized vascular channels. The capillary

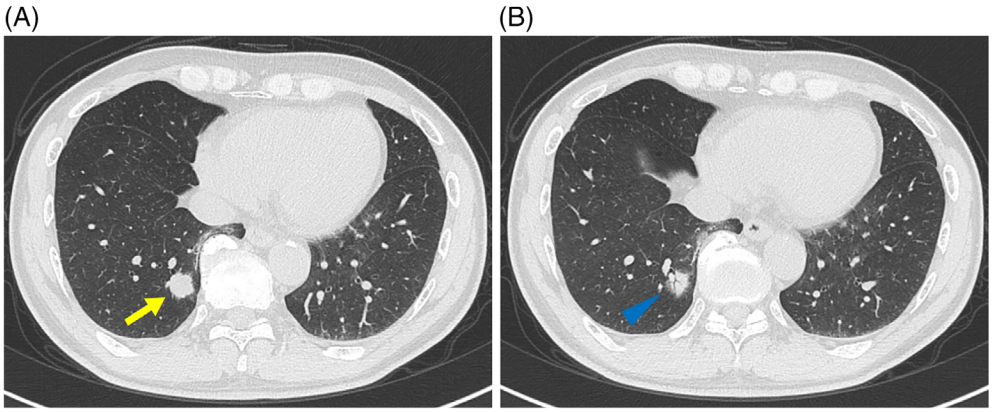

FIGURE 1 (A) Chest CT scan revealing a lobular solid nodule with a maximum diameter of 1.9 cm in the lower lobe of the right lung. (B) The arrowhead indicates an air bronchogram on the lower edge of the nodule

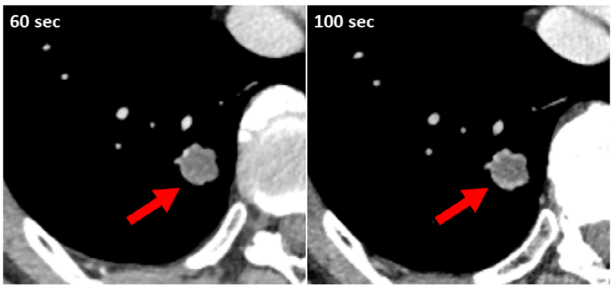

FIGURE 2 Contrast-enhanced CT showing the peripheral nodular enhancement in late phase (100 s)

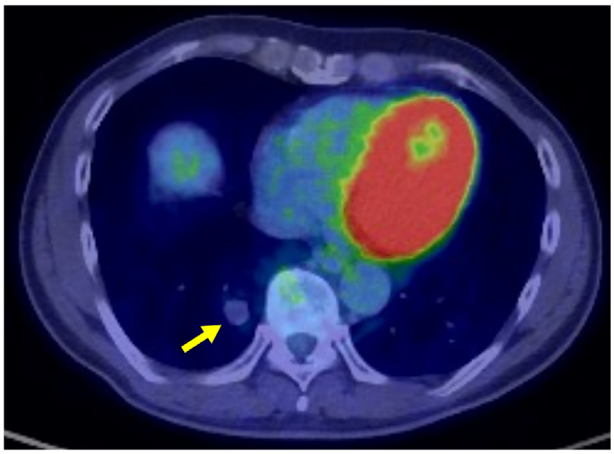

FIGURE 3 On PET-CT, the maximum standardized uptake value of the nodule is 1.6

vessels extended along the alveolar septa but did not destroy the alveolar structure (Figure 4). Numerous capillaries were immunohistochemically stained for vascular endothelial markers (CD31, CD34, Factor VIII-related antigen, claudin 5, and the erythroblast transformation specific-related gene [ERG]). Based on the above findings, we diagnosed the nodule as a PCH. The patient did well after surgery. A follow-up CT obtained 6 months after surgery confirmed no recurrence.

DISCUSSION

Pulmonary hemangiomas are rare, benign vascular tumours that typically consist of vascular spaces, ranging from capillary-size to larger vessels with a cavernous appearance. Pulmonary hemangiomas present as parenchymal or endobronchial lesions in the lung and often mimic early-stage lung cancer and other benign tumours, such as non-specific interstitial pneumonia, early-stage organizing pneumonia, and minute pulmonary meningothelial-like nodules.[1] Sometimes pulmonary hemangiomas present as multiple lesions in the lung concurrent with other organs, such as the brain, skin, liver, or heart. The age of onset ranges from children to the elderly, and there is a slight female predominance.[2] Most patients are asymptomatic and the diagnosis is an incidental finding; however, hemoptysis, respiratory distress, and pneumothorax have been reported. Based on pathologic examination, hemangiomas consist of a proliferation of vascular channels, and the alveolar structure is not destroyed, being preserved and excluded with vascular hyperplasia. There are no cell atypia and no malignant findings cytologically. Based on immunohistochemical staining, vascular endothelial cells are stained with vascular endothelial markers, such as CD31, CD34, Factor VIII-related antigen, claudin 5, erythroblasts, and ERG.

Indeed, we identified 48 cases of PCH in adults in the English literature since 1990.[1,3–18] The detailed characteristics of these cases are described in Table 1. A slight female predominance was noted (60.4% [29/48]). The age of onset

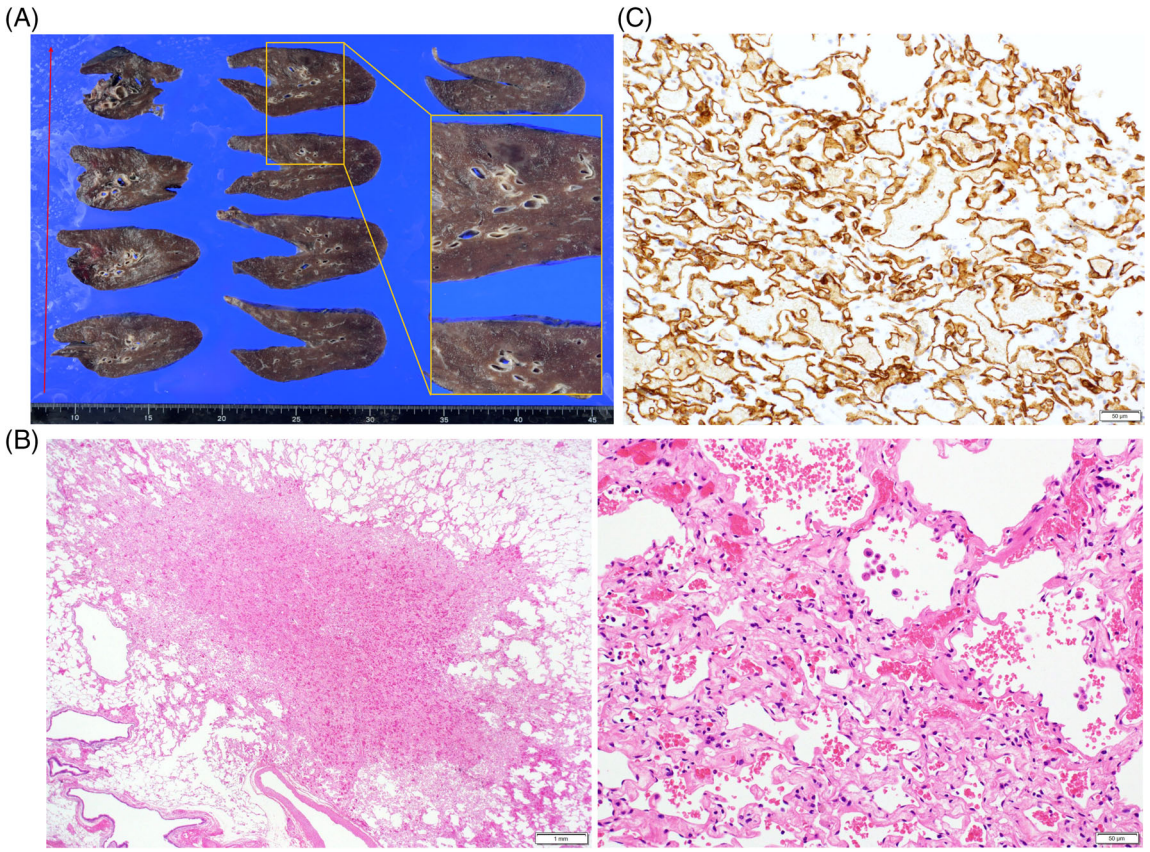

FIGURE 4 (A) On the cut surface of the lung specimen, a circumscribed dark red mass is seen macroscopically, measuring 1.5 × 0.7 cm. (B) Haematoxylin and eosin staining. Benign proliferation of vascular channels is observed, especially the high density in the center of the tumour. (C) Immunohistochemical staining. The endothelial cells stain with CD31

ranged from 19 to 68 years (median, 48.5 years). Most patients had no symptoms. The size ranged from 3 to 25 mm (median, 10 mm). The tumours exhibited various findings on CT in each case report. Based on our literature search, there were 17 pure GGNs, 22 PSNs, and 6 solid nodules. Two cases had cystic lesions and one case had a lumen dilatation suggestive of bronchiectasis. Zhu et al.[13] reported that pure GGNs may present with minimal vascular hyperplasia and a relatively preserved alveolar space containing more air, while solid nodules may have a dense proliferation of capillary vessels. It has been reported that pulmonary hemangiomas exhibit a pattern on contrast-enhanced CT similar to the characteristics of hepatic hemangiomas, in which the margins are slightly stained in the early phase and the entire lesion is deeply stained in the late phase.[8] In our case, the tumour exhibited a solid nodule and peripheral nodular enhancement on contrast-enhanced CT. The pathologic finding in our case showed dense capillary hyperplasia in the center of the tumour. Based on this finding, we concluded that a solid pattern was shown on the image and the contrast medium did not reach the dense central part, thus a gradual increase in the contrast effect of the margin was shown. Based on the above, we further reasoned that the solid pattern and peripheral nodular enhancement were evident only on CT, suggesting a hemangioma with dense vascular hyperplasia. To date, few reports have referred to contrast-enhanced CT findings of PCH. Although it is still difficult to distinguish PCH from other diseases, such as early lung cancer, based on imaging findings alone, we suggest contrast-enhanced CT might help diagnosing PCH.

In 2012 Kim et al.[8] reported a patient with a pulmonary hemangioma recurrence. A 22-year-old woman underwent partial lung resection for pulmonary lesions in the left upper and lower lobes and was diagnosed with a PCH. New nodules were found in the left and right lower lobes, and a left lower lobectomy and partial lung resection for the right lesion were performed. The pathologic findings were very similar to the previous lesions. The final diagnosis was a pulmonary hemangioma recurrence. New lesions were not demonstrated on CT in our patient at the 6-month follow-up evaluation. Pathologically, a pulmonary hemangioma is classified as a benign tumour, but given the report of a recurrence, it is necessary to consider whether follow-up, including imaging tests, should be performed in patients with a pulmonary hemangioma to rule out malignancy.

In summary, we managed a patient with a PCH that was difficult to distinguish from lung cancer. Although

TABLE 1 Clinical features of 48 resected cases of PCH

Total number of patients[1,3–18]		**48**
Gender	Male	19
	Female	29
Age (years)	median	48.5
	range	19–68
Symptoms	No symptoms	20
	Pneumothorax and hemoptysis	1
	Cough	1
	Hemoptysis	1
	ND	25
Size (mm)	median	10
	range	3–25
CT findings	pure GGN	17
	PSN	22
	Solid	6
	Cyst	2
	Others[a]	1
Location	RUL	4
	RML	6
	RLL	13
	LUL	7
	LLL	17
	Multiple	1
PET findings	Normal uptake	8
	Not done	5
	ND	35
Surgery	Wedge resection	28
	Segmentectomy	8
	Lobectomy	12
Prognosis	Alive, more than 1 month	9
	Alive, more than 1 year	14
	Alive, more than 5 years	3
	Recurrence	1
	ND	21

Abbreviations: CT, computed tomography; GGN, ground-glass nodule; LLL, left lower lobe; LUL, left upper lobe; ND, not described; PET, positron emission tomography; PSN, part-solid nodule; RLL, right lower lobe; RML, right middle lobe; RUL, right upper lobe.
[a]Lumen dilatation suggestive of bronchiectasis.

the characteristic imaging findings of PCH have not been established and additional case reports are needed, contrast-enhanced CT may facilitate the diagnosis, and if diagnostic imaging becomes more useful, surgery may be avoided. In contrast, the possibility of a recurrence has been noted. Although no malignant findings have been observed pathologically, it is necessary to consider the treatment policy and follow-up period. We hope this case report adds further information to diagnose and manage pulmonary hemangiomas.

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CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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