

Recurrent Pulmonary Capillary Hemangioma: Dynamic Contrast-Enhanced CT and Histopathologic Findings

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We report the dynamic contrast-enhanced CT and histopathologic findings of a rare case of recurrent pulmonary capillary hemangiomas. The findings consisted of peripheral nodular enhancement at the early arterial phase and a subsequent "central filling-in" enhancement pattern on the delayed scans, which was identical to the well-known enhancement pattern of hemangiomas of the liver. Although there was no evidence of histological malignancy, pulmonary capillary hemangiomas manifested as multiple nodular lesions and showed postoperative recurrence.

Index terms: Hemangioma; Lung neoplasms; Tomography; X-ray computed

INTRODUCTION

A pulmonary hemangioma is an extremely rare disease with only a few cases reported to date (1-5). To the best of our knowledge, this is the first report of a recurrent pulmonary hemangioma with dynamic contrast-enhanced CT findings for this type of pulmonary neoplasm. We report here the dynamic contrast-enhanced CT and histopathologic findings of a pulmonary capillary hemangioma.

CASE REPORT

A 22-year-old woman was referred to our institution with a past medical history of wedge resection of the left

lung for two incidentally detected pulmonary nodules at another hospital fifty months earlier. The initial outside multi-phase dynamic CT scan obtained at that time using a multidetector CT scanner (Sensation 16, Siemens, Erlangen, Germany) after intravenous injection of contrast media was available for review, although the information on the amount and the brand name of the contrast media was not available. The CT images revealed two nodules each in the left upper and lower lobes. These nodules were round in shape with a well-circumscribed margin, and maximum diameters of 22 and 20 mm, respectively. On non-contrast CT scan, the nodules revealed no calcification. On multi-phase dynamic CT images, the nodules showed peripheral nodular enhancement at the early arterial phase obtained 1 minute after intravenous administration of contrast media and a subsequent "central filling-in" enhancement pattern on the delayed scans (obtained 2 and 4 minutes after contrast enhancement) (Fig. 1), which was identical to the well-known enhancement pattern of hemangiomas of the liver. Wedge resection of those left pulmonary nodules was performed at the other hospital, and the review of the pathologic specimen revealed pulmonary capillary hemangiomas.

At 50 months follow-up, new pulmonary nodules were detected on a chest radiograph, and the patient visited our

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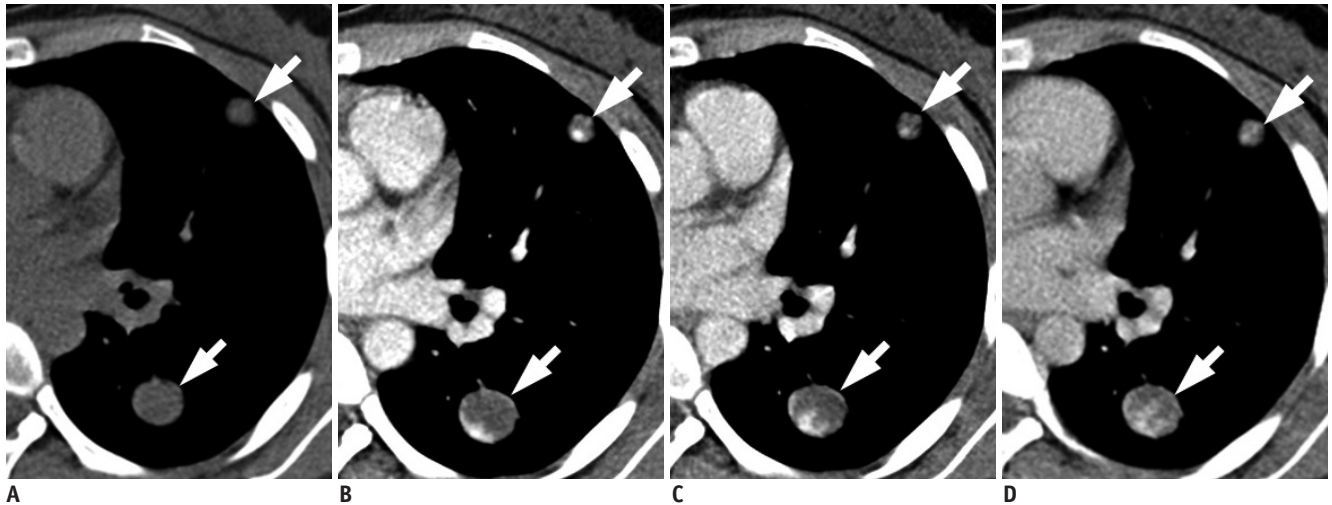


Fig. 1. Pulmonary capillary hemangiomas in 22-year-old female.

Initial multi-phase dynamic CT scan obtained at outside hospital shows two pulmonary nodules in left lung. Dynamic postcontrast CT scans (B-D) show peripheral nodular enhancement in early phase CT (B) and "central filling-in" enhancement pattern of these nodules in subsequent delayed images (C, D), which are identical to well-known enhancement pattern of hepatic hemangiomas (A: precontrast image, B: obtained 1 minute after contrast enhancement, C: 2 minutes, D: 4 minutes). Wedge resection of these left pulmonary nodules was performed at outside hospital, and review of pathologic specimen revealed pulmonary capillary hemangiomas. Initial multi-phase dynamic CT scan obtained at outside hospital shows two pulmonary nodules in left lung.

institution for further evaluation. The CT scan obtained at our institution using a multidetector CT scanner (LightSpeed VCT, GE Healthcare) after intravenous injection of 100 mL (3 mL/s) of nonionic contrast media (Iopamiron 300; Bracco, Milan, Italy) revealed two recurrent pulmonary nodules each in the left lower lobe and the right lower lobe with maximum diameters of 30 and 12 mm, respectively. These nodules also showed a round shape with a well-circumscribed margin. The larger one detected in the left lower lobe especially showed the prominent "peripheral nodular enhancement" pattern at post-contrast CT images obtained 40 seconds after contrast administration (Fig. 1E), which was the same as the previous CT finding.

A left lower lobectomy and wedge resection of the right lower lobe was performed, and the gross specimens of the resected nodules showed well-demarcated hemorrhagic or vascular nodules (Fig. 1F). Histopathologic examination revealed vascular tumors of a capillary growth pattern. These nodules were composed of anastomosing capillary vascular channels filled with red blood cells. These capillary lumina were divided by narrow trabeculae of hyaline stroma (Fig. 1G), which were identical to those of the previously resected pulmonary nodules at the other hospital. The final diagnosis was recurrent pulmonary capillary hemangiomas, and the tumors showed no evidence of histologically malignant features such as cellular atypia or mitosis.

DISCUSSION

A hemangioma is a neoplasm of benign vascular proliferation which can be divided into cavernous and capillary types, depending on the diameter of the vascular channels that comprise the tumor, while a capillary hemangioma can be further subdivided into localized and multifocal types. Although they typically present in the skin and the liver, hemangiomas can be found within the thorax, most often in the subglottic area of the respiratory tract (3, 6). Being an extremely rare disease, only a few pulmonary hemangioma cases have been reported (Table 1) (1-5). Most patients were very young, except for two adult cases. The patients had diverse clinical presentations ranging from asymptomatic to catastrophic (1-5). On CT, they showed various imaging findings according to the hemangioma components of the cavernous or capillary growth pattern (Table 1). Microscopically, a cavernous hemangioma is a well-demarcated lesion composed of large spaces filled with red blood cells and thrombotic materials (2, 3), while capillary hemangiomas do not have such large vascular spaces (3). Instead, capillary hemangiomas have small and regular vascular spaces that are lined by flattened endothelial cells where the vascular spaces can appear engorged with red blood cells (3). On CT, cavernous hemangiomas can present as a discrete mass (2, 5), while localized capillary hemangioma manifests appear as a

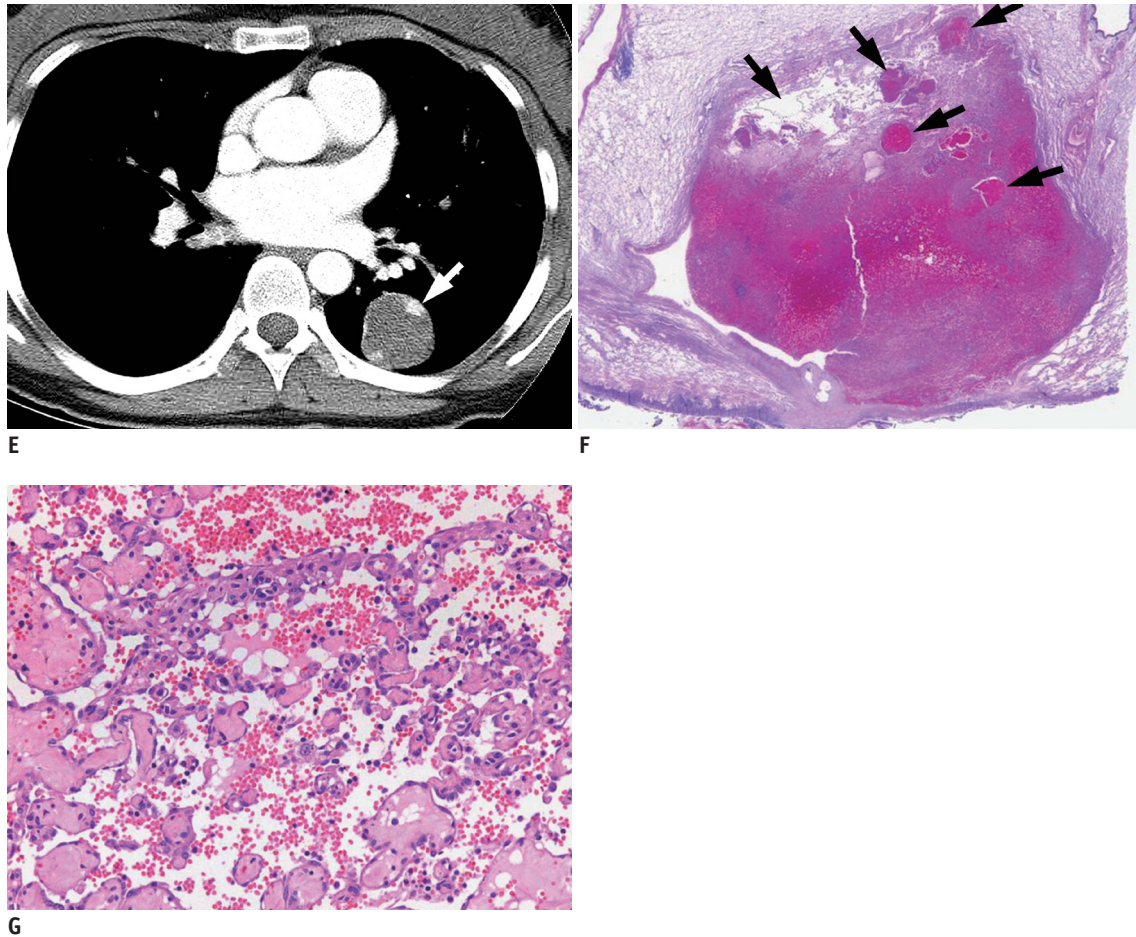


Fig. 1. Pulmonary capillary hemangiomas in 22-year-old female.

E. Fifty months after tumor removal, chest CT scan obtained from our institution shows 30-mm, recurrent pulmonary nodule in left lower lobe. Another smaller nodule is also detected in right lower lobe (not shown here). Nodule in left lower lobe shows prominent “peripheral nodular enhancement” pattern at this post-contrast CT image obtained 40 seconds after contrast administration, which is identical to previous CT finding. **F.** Scanning photograph of histopathologic specimen obtained from left lower lobectomy shows well-demarcated hemorrhagic nodule containing multiple dilated vascular spaces (arrows), many of which are compactly packed with red blood cells (H and E, x1). **G.** Low-power photomicrograph shows that nodule is composed of anastomosing capillary vascular channels filled with red blood cells. These capillary lumina are divided by narrow trabeculae of hyaline stroma (H and E, x40). These findings are compatible with capillary hemangiomas.

localized cystic lesion (3) or focal ground-glass opacity nodule (4). In multifocal capillary hemangiomas, a CT scan shows numerous nodules on the pleural surface with septal thickening, which can simulate an interstitial lung disease (3).

There were no recurrence cases of pulmonary hemangioma in the literature, although the postoperative follow-up periods were short. Despite no histological evidence of malignancy including cellular atypia and increased mitosis, our case of pulmonary hemangioma manifested as two nodules at the initial manifestation and showed recurrence presenting as two new nodules 50 months after the initial operation. This indicates that pulmonary hemangiomas might be included in the category of neoplasms of “uncertain

malignant potential” in which behavior cannot be predicted from their histopathologic characteristics alone (7). Periodic follow-up evaluation by way of a CT scan would be helpful for early detection of the recurrence of pulmonary hemangioma.

Sclerosing hemangiomas of the lung can be confused with pulmonary capillary or cavernous hemangiomas due to their similar name. However, it was named as such because of the frequent hemorrhage and hemosiderin within the tumor, even though it is not considered to be a vascular tumor like a pulmonary hemangioma (8). The alternative designation is sclerosing pneumocytoma since current evidence suggests that sclerosing hemangiomas originate from primitive undifferentiated respiratory epithelial cells (8, 9). Dynamic

Table 1. Summary of Pulmonary Hemangiomas Reported in Literature

Reference	Cases (n)	Age at Diagnosis	Sex	Clinical Symptoms	Radiologic Findings (location)	Follow-Up	Pathology
Bowyer and Sheppard (1)	1	Neonate	F	Respiratory distress	A 6 cm sized air-filled cyst (RLL)	No recurrence after lobectomy, (follow up period is NA)	A cyst entirely surrounded by capillary hemangioma
Galliani et al.(2)	1	10 wks	M	Rhinorrhea, cough	A 7 cm sized mass (RLL)	No recurrence after lobectomy (follow up period : 12 months)	Cavernous hemangioma
Abrahams et al. (3)	2	8 wks	M	Respiratory distress	A cystic mass (RLL)	No recurrence after wedge resection (follow up period is NA)	Localized capillary hemangioma
		9 yrs	F	Cyanosis, clubbing,	Multiple nodules simulating ILD (predominantly right lung)	Died of massive hemoptysis	Multifocal capillary hemangioma
Fugo et al. (4)	2	56 yrs	M	No specific Symptom	A small semisolid nodule (LLL)	NA after segmentectomy,	Localized capillary hemangioma
		48 yrs	F	No specific Symptom	A small semisolid nodule (RML)	NA after wedge resection	Localized capillary hemangioma
Capizzani et al. (5)	1	Neonate	M	Respiratory distress	Huge mass (right lung)	No recurrence after resection (follow up period : 6 months)	Both of capillary and cavernous component

Note.— F = female, M = male, RLL = right lower lobe, LLL = left lower lobe, RML = right middle lobe, ILD = interstitial lung disease, NA = not available

enhancement characteristics of the tumors depend on the levels of histologic components (papillary, sclerotic, solid, and hemangiomatous); a hemangiomatous or papillary component reveals early and strong enhancement, while a solid or sclerotic component shows slow and persistent enhancement (10). Although these enhancement patterns of sclerosing hemangioma can sometimes look quite similar to that of capillary hemangioma, the former usually do not show a striking peripheral nodular enhancement of the latter on the early arterial phase.

In conclusion, a pulmonary hemangioma is a rare vascular tumor predominantly occurring in younger individuals, but can sometimes be seen in adults without specific symptoms. Without any evidence of histological malignancy, the tumors can show multiplicity and have a postoperative recurrence. On dynamic contrast-enhanced CT, pulmonary capillary hemangiomas in our case showed a pattern of early peripheral nodular enhancement and a delayed “central filling-in” appearance, which is identical to the well-known, characteristic enhancement pattern of hepatic hemangiomas.

REFERENCES

1. Bowyer JJ, Sheppard M. Capillary haemangioma presenting as a lung pseudocyst. *Arch Dis Child* 1990;65:1162-1164
2. Galliani CA, Beatty JF, Grosfeld JL. Cavernous hemangioma of the lung in an infant. *Pediatr Pathol* 1992;12:105-111
3. Abrahams NA, Colby TV, Pearl RH, Chipps BE, Juris AL, Leslie KO. Pulmonary hemangiomas of infancy and childhood: report of two cases and review of the literature. *Pediatr Dev Pathol* 2002;5:283-292
4. Fugo K, Matsuno Y, Okamoto K, Kusumoto M, Maeshima A, Kaji M, et al. Solitary capillary hemangioma of the lung: report of 2 resected cases detected by high-resolution CT. *Am J Surg Pathol* 2006;30:750-753
5. Capizzani TR, Patel H, Hines MH, Mott RT, Petty JK. A unique case of a giant congenital pulmonary hemangioma in a newborn. *J Pediatr Surg* 2008;43:574-578
6. Dinehart SM, Kincannon J, Geronemus R. Hemangiomas: evaluation and treatment. *Dermatol Surg* 2001;27:475-485
7. Sienko A. *Pulmonary processes of indeterminate malignant potential*. In: Danis S, Zander, Farver CF, eds. *Pulmonary pathology*. Philadelphia, USA: Churchill Livingstone, 2008:636-648
8. Flieder DB. *Benign Neoplasms of the Lungs*. In: Danis S.

- Zander, Farver CF, eds. *Pulmonary pathology*. Philadelphia, USA: Churchill Livingstone, 2008:669-672
9. Katzenstein AL, Gmelich JT, Carrington CB. Sclerosing hemangioma of the lung: a clinicopathologic study of 51 cases. *Am J Surg Pathol* 1980;4:343-356
10. Chung MJ, Lee KS, Han J, Sung YM, Chong S, Kwon OJ. Pulmonary sclerosing hemangioma presenting as solitary pulmonary nodule: dynamic CT findings and histopathologic comparisons. *AJR Am J Roentgenol* 2006;187:430-437