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Case Report

Pulmonary capillary hemangioma diagnosed by needle core biopsy: Case report and review of the literature

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ABSTRACT

Pulmonary hemangiomas are extremely rare. Here we report a case of a 42 year old healthy woman with an incidental nodule identified by computed tomography (CT). In consideration of her smoking history and the appearance of irregular borders of the nodule on CT imaging, despite lack of change after a 9-month follow-up, a CT-guided percutaneous needle core biopsy was performed to rule out lung cancer. A diagnosis of solitary pulmonary capillary hemangioma was made histopathologically. The feasibility of using needle core biopsy to diagnose pulmonary hemangiomas is discussed with the clinical, radiologic and pathological features of this case and a review of literature.

1. Introduction

Hemangiomas are common benign vascular tumors of infancy and childhood that are frequently located in the skin, subcutaneous tissue, and liver [1]. Although they can present in any organ system, primary pulmonary hemangiomas, involving either the parenchyma or the airways, are extremely rare [2].

Extensive search of the English literature reveals only a few cases of pulmonary hemangiomas. The diagnoses were all made through wedge resection, segmentectomy, or lobectomy with or without intraoperative frozen section examinations. Here, we report one additional case of pulmonary capillary hemangioma which was diagnosed by needle core biopsy. The feasibility of using needle core biopsy to diagnose pulmonary hemangiomas will be discussed with a literature review.

2. Case report

2.1. Clinical presentation

A 42 year old woman with a 5-pack-year smoking history while in college and no other significant past medical histories had an incidental nodule in the left lower lobe on chest X-ray performed at another hospital. Clinical follow-up at our institution revealed a left lower lobe pulmonary nodule measuring 1 cm in the greatest dimension with irregular borders and central fluid attenuation suggestive of a cystic component on a computed tomography (CT) scan (Fig. 1). No air-fluid level was seen to indicate cavitation or necrosis. This lesion is closely associated with an interlobular septum. No definite feeding or draining vessels had been identified. This pulmonary nodule appeared to be stable in size and appearance through a 9 month interval. There was no evidence suggestive of pulmonary hypertension clinically or radiographically.

In consideration of the irregular borders of this nodule on CT scan and the smoking history, a CT-guided percutaneous lung biopsy was performed to rule out the possibility of a malignant process. Patient experienced significant but transient hemoptysis after the procedure.

2.2. Histopathologic findings

Microscopically, five pieces of lung tissue were present, one of which was completely normal lung parenchyma. The remaining 4 tissue core fragments showed partial or complete involvement by a discrete and marked proliferation of thin-walled capillary vessels filled with red blood cells (Fig. 2A). Lesional tissue measures 5 mm in the longest dimension. The cells lining the vascular channels were cytologically bland and showed no mitotic activity (Fig. 2B); and immunohistochemically were strongly positive for ERG (Fig. 2C; clone: EP111, EPITOMICS, Burlingame, CA) and negative for the lymphatic endothelial marker, podoplanin (clone: D2-40, Dako, Carpinteria, CA). Immunohistochemical staining of TTF-1 was only positive in rare entrapped pneumocytes but negative in the lesional nuclei (Fig. 2D; clone: 8G7G3/1, DAKO, Carpinteria, CA).

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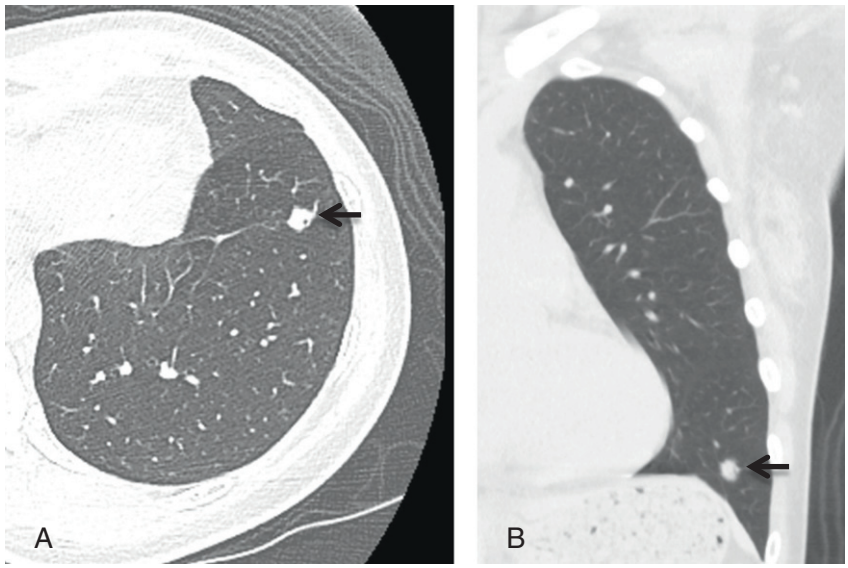

Fig. 1. CT images of the axial (1A) and coronal (1B) planes show a single, 1.0 cm left lower lobe pulmonary nodule with irregular borders and central fluid attenuation (arrow).

The capillary proliferations appeared to be adjacent to a bronchovascular bundle (Fig. 3A), however no obvious involvement or invasion of the bronchovascular wall was appreciated under high magnification (Fig. 3B and C). Iron stain did not reveal significant hemosiderosis in the form of either hemosiderin granules within alveolar macrophages or extracellular hemosiderin deposits in alveolar septa. In addition, there was no inflammation, fibrosis, or granulomas identified.

Based on the histomorphologic features, along with the clinical and radiologic findings, a diagnosis of solitary pulmonary capillary hemangioma was rendered.

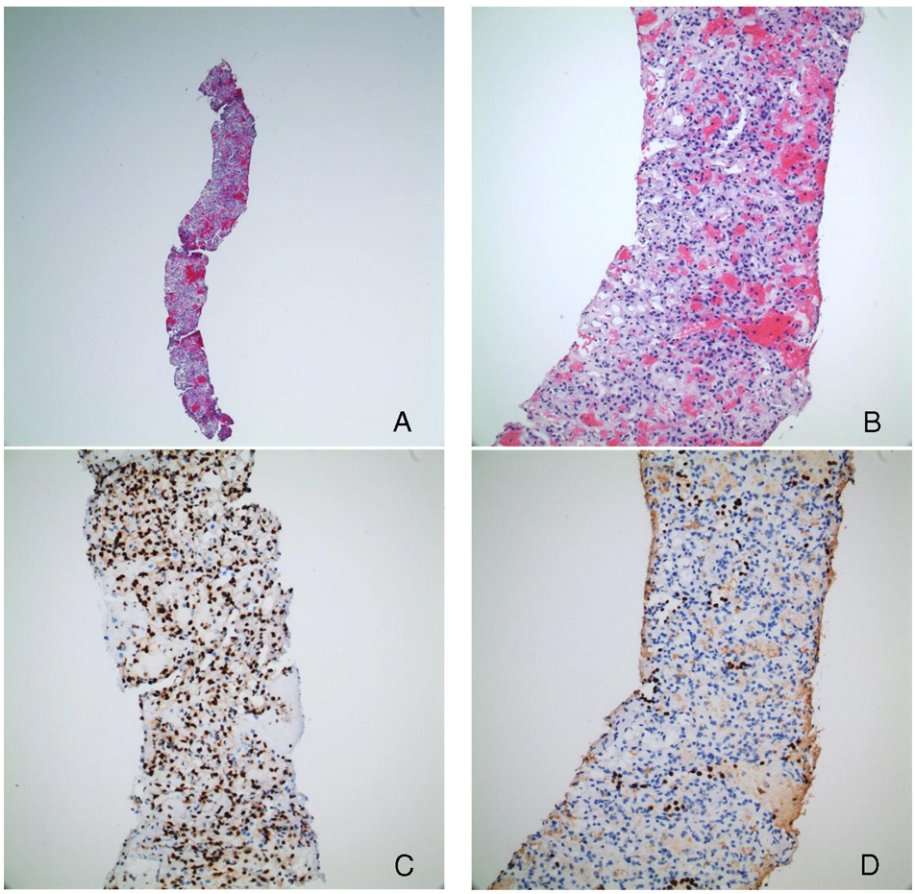

Fig. 2. Microscopic images of the lesion show marked proliferation of thin-walled capillary vessels (2A) with bland cytology and no mitosis (2B). The cells lining the vascular channels are positive for ERG (2C) and negative for TTF1 (2D).

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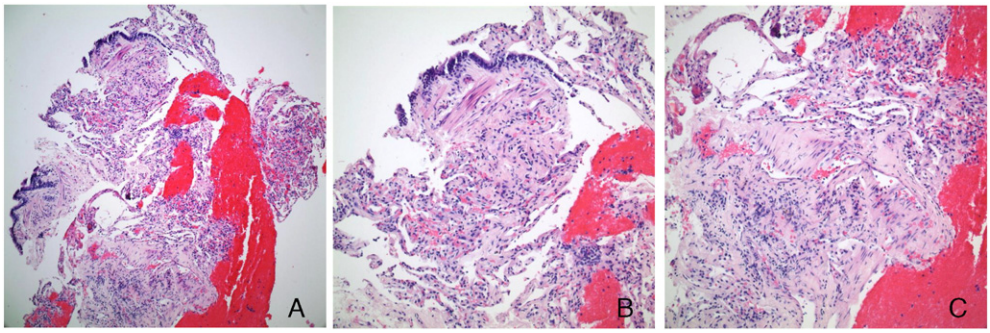

Fig. 3. The capillary proliferations are adjacent to a bronchovascular bundle (3A). No obvious involvement or invasion of the bronchial (3B) or vascular (3C) wall is appreciated under high power.

3. Discussion

Hemangiomas are benign vascular tumors commonly occurring in skin, subcutaneous tissue, and liver [1]. In contrast, hemangiomas of the lung are exceedingly rare. Reviewing the literature published in English, we identified 20 cases of hemangiomas involving either the lung parenchyma or the bronchus (Table 1) [3–20] The more common hemangiomas within the respiratory tract involving the subglottic region and the mediastinum have been reviewed by Shikhani [21] and Moran and Suster [22], which are not included in the current review.

Pulmonary hemangiomas have a broad range of clinical presentations depending on the location, size and number of lesions. Among the 20 published cases and one additional case from our current study, 7 cases were asymptomatic, whereas the rest were associated with symptoms ranging from mild cold-like symptoms, respiratory distress, cyanosis, and pleural effusion to hemoptysis, including one case of hemoptysis that was massive and life-threatening. Of these 21 patients,

Table 1
Summary of pulmonary hemangioma reported in the literature.

Reference	Age	Sex	Clinical symptoms	Site	Radiology	Pathology
Kayser et al. [3]	3 m	F	Dyspnea & tachypnea	Left main bronchus	Significant narrowing of the left main bronchus	Infantile hemangioma
Paul et al. [4]	5 m	F	Marked respiratory distress	Right main bronchus	Hyperinflation of the right lung	Capillary hemangioma
Cohen & Kaschula [5]	4 y	F	Cough, bronchitis, pneumonia	Right main bronchus	N/A	Capillary hemangioma
Harding et al. [6]	67 y	F	Severe chest pain, hemoptysis	Left main bronchus	Left hilar mass	Capillary hemangioma, pedunculated
Capizzani et al. [7]	New-born	M	Premature newborn w respiratory distress	Right hilum	Cystic and solid mass occupying most of the right hemithorax	Mixed cavernous and capillary hemangioma (Congenital hemangioma)
Maeda et al. [8]	54 y	M	Asymptomatic	Left hilum	Ill-defined mass adjacent to the bifurcation of the left main bronchus	Cavernous hemangioma
Abrahams et al. [9]	9 y	F	Cyanosis, clubbing, shortness of breath	Predominantly right lung	Bilateral interstitial and pleural nodules	Capillary hemangioma[a]
Kim et al. [10]	22 y	F	Asymptomatic	Initial: LUL & LLL Later: LLL & RLL	Initial: 2 round well-circumscribed nodules w enhancement Later: 2 round well-circumscribed nodules w enhancement	Capillary hemangioma
Yang et al. [11]	36 y	M	Pericardial & bilateral pleural effusion	Bilateral, multiple	Bilateral multiple nodules	Cavernous hemangioma
Miyamoto et al. [12]	61 y	M	Asymptomatic	Bilateral, multiple	Multiple well-defined nodules	Cavernous hemangioma
Fine & Whitney [13]	84 y	M	Pneumonia	Bilateral, multiple	N/A (incidental findings on autopsy)	Cavernous hemangioma
Siaghani et al. [14]	New-born	M	Respiratory distress, apneic episodes	LLL	2 homogenous, hypervascular masses at the periphery of LLL	Infantile hemangioma
Abrahams et al. [9]	8 wk	M	Respiratory distress	RLL	Cystic mass	Capillary hemangioma
Galliani et al. [15]	10 wk	M	Cold-like symptoms	RLL	An enhancement pattern suggestive hemangioma	Cavernous hemangioma
Bowyer & Sheppard [16]	9 m	F	Premature newborn w intubation	RLL	Smooth walled, air filled cyst	Capillary hemangioma
Huang et al., this study	42 y	F	Asymptomatic	LLL	Nodule with irregular borders and cystic components	Capillary hemangioma
Fugo et al. [17]	48 y	F	Asymptomatic	RML	Ground glass opacity	Capillary hemangioma[a]
Sakaguchi et al. [18]	53 y	F	Asymptomatic	LUL	Ground glass opacity	Capillary hemangioma[a]
Sirmali et al. [19]	54 y	M	Massive hemoptysis	LUL	Solid mass continuous w LUL vascular bundle	Cavernous hemangioma
Isaka et al. [20]	55 y	F	Asthma	LLL	Ground glass opacity	Capillary hemangioma[a]
Fugo et al. [17]	56 y	M	Asymptomatic	LLL	Ground glass opacity	Capillary hemangioma[a]

y: year; M: man; W: woman; w: with; wk.: week; m: month; LLL: left lower lobe; RML: right middle lobe; LUL: left upper lobe; RLL: right lower lobe.
[a] PCH-like histology.

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their ages ranged from newborn to 84 years old. 6 of the 21 cases were located in the main bronchus or hilum while the rest were in the periphery or subpleural region. Majority of the patients presented with solitary lung nodule, while 5 patients showed multifocal lesions (Table 1).

Radiographically, 2 cases of endobronchial hemangiomas showed either significant narrowing of the main bronchus or hyperinflation of the lung, respectively [3,4]. 4 cases showed mass lesions with cystic components, with 3 from newborns [7,9,16] and 1 from this current 42 year old patient. 4 cases showed characteristic enhancement pattern or hypervascular pattern, raising the suspicion of a pulmonary vascular lesion as one of the differential diagnoses preoperatively [7,10,14,15]. 4 cases showed ground glass opacity, making it difficult to differentiate radiographically from early lung cancer [17,18,20]. 1 case showed bilateral interstitial and pleural nodules suggestive of interstitial lung disease radiographically [9]. The rest of the cases showed either solitary or multifocal well-defined lung nodules, raising the concern for either early primary lung cancer or metastatic diseases. Based on literature review, there are no specific radiographic characteristics of pulmonary hemangiomas.

Pulmonary hemangioma is such a rare entity that inclusion of this lesion in the differential diagnosis rarely enters the threshold of suspicion, especially when preoperative radiology does not show a characteristic enhancement or hypervascular pattern. However, this should be considered as a differential diagnosis for both solid and cystic intrapulmonary mass lesions, especially when core needle biopsy is being planned for histopathological diagnosis. The patient that we are reporting here did experience significant, although transient, hemoptysis after CT-guided percutaneous needle core biopsy. The recent federal approval of annual screening for lung cancer with low dose CT has facilitated the early detection of small-sized lung cancers. However, it is necessary to be aware of lesions mimicking small-sized lung cancers, such as pulmonary hemangiomas, which could cause catastrophic bleeding complications through CT-guided lung biopsy. All the 20 cases reviewed here were diagnosed through lung resections (wedge resection, segmentectomy, or lobectomy) or incidental finding through autopsy. Ours is the 1st case that was diagnosed through CT-guided percutaneous needle core biopsy. If pulmonary hemangioma was already initially considered, longer follow-up or wedge resection may have been the favored options. Only 4 out of 21 reviewed cases showed characteristic enhancement or hypervascular pattern, indicating a vascular lesion on pre-operative radiography. More studies are needed to better characterize this rare pulmonary vascular lesion radiographically to facilitate pre-operative planning of what would be the best and safest procedure for a histopathologic diagnosis.

It can be challenging to make a diagnosis of pulmonary capillary hemangioma based on limited tissue. Entities that should especially be considered as differential diagnoses include pulmonary venous hypertension and pulmonary capillary hemangiomatosis (PCH). Pulmonary venous hypertension can be caused by obstruction of either intrapulmonary (so called pulmonary veno-occlusive disease) or extrapulmonary veins often due to chronic congestive heart failure or mitral stenosis. PCH is recognized in published literature as a progressive and fatal disease and the only definitive treatment for PCH is lung transplantation [24]. In both conditions, prominent capillary proliferations resulting in alveolar septal thickening, along with abundant hemosiderosis, are characteristic. The absence of hemosiderosis in our present case makes these differential diagnoses unlikely. The diagnosis of solitary pulmonary capillary hemangioma was not only based on the histomorphology and immunohistochemical reactions but also the clinical and radiologic findings, more specifically, the absence of symptoms, like dyspnea or hemoptysis, and not having a bilateral and diffuse infiltrate on CT scan.

Interestingly, Havlik et al. [23] reported 8 cases of incidental autopsy findings of PCH-like foci that were all localized and unrelated to the cause of death. And none of these 8 patients had any symptoms such as dyspnea or hemoptysis. Histologically, capillary proliferations within the alveolar walls were noted in all 8 cases. Additional studies are required to determine whether PCH-like lesions represent incipient stage of PCH or whether PCH-like lesions could progress into PCH with time. Among the 20 reviewed cases in here, 5 cases had PCH-like histology without associated symptoms of pulmonary hypertension [9,17,18,20]. A diagnosis of pulmonary capillary hemangioma was given for all 5 cases.

Solitary lesions were treated with surgical resection and often had a favorable outcome if totally resected. Despite the benign course of solitary pulmonary hemangioma, 2 of 5 patients with multifocal lesions died after initial wedge resection confirming a diagnosis of pulmonary capillary or cavernous hemangioma [9,11]. Interferon alfa-2a is reported to treat more diffuse angiomatous proliferation successfully with minimal side effects [24,25]. 1 of the 2 patients, mentioned earlier, who died from multifocal pulmonary hemangiomas was actually treated with interferon alfa-2a for 12 weeks but with no response [9]. For our present case, with the reassurance from the histopathologic diagnosis, no further resection is thought needed and the patient will be followed by imaging studies.

In summary, although pulmonary hemangiomas are rare, it is important to consider it as one of the differential diagnoses pre-operatively for lung nodules detected by CT, especially in the era of low dose CT screening of early lung cancers. Where appropriate, limited surgical lung resection instead of core biopsy to avoid complications of bleeding would be the choice for a histopathologic diagnosis.

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