Ann Thorac Cardiovasc Surg 2014; Supplement: 578–581
Online March 22, 2013
doi: 10.5761/atcs.cr.12.02222

Case Report

A Resected Case of Solitary Pulmonary Capillary Hemangioma Showing Pure Ground Glass Opacity

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A 53-year-old Japanese woman was referred to our hospital for pure ground glass opacity (GGO) in the left upper lung on computed tomography. 18F-fluorodeoxyglucose positron emission tomography revealed no abnormal uptake in the lesion. After 4 months of follow-up, we strongly suspected the lesion to be a bronchioloalveolar carcinoma and decided to perform a surgery for diagnosis. Planning to perform lingual segmentectomy by video-assisted thoracic surgery (VATS) with mini thoracotomy, we finally performed the left upper lobectomy to guarantee the optimal margin by VATS, because the lesion was not identified by palpation. Final pathological diagnosis was a solitary pulmonary capillary hemangioma (SPCH). We herein report a resected case of SPCH showing pure GGO and review all reported SPCH cases.

Keywords: solitary pulmonary capillary hemangioma, pulmonary capillary hemangiomatosis, ground-glass opacity, bronchioloalveolar carcinoma

Introduction

Solitary pulmonary capillary hemangioma (SPCH) is a rare benign lung tumor of good prognosis. To our knowledge, only 7 adult SPCH cases have been reported. We herein report a resected case of SPCH showing pure ground-glass opacity (GGO) and review all reported SPCH cases.

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Received: December 6, 2012; Accepted: January 9, 2013
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Case Report

A 53-year-old Japanese woman was referred to our hospital for further evaluation and treatment of an abnormal shadow on medical checkup computed tomography (CT). The chest CT showed a pure GGO lesion measuring 20 mm in diameter in the left upper lung (**Fig. 1a**). ^{18}F-fluorodeoxyglucose positron emission tomography (FDG-PET) revealed no abnormal uptake in the lesion. She had received medication for hypothyroidism for 8 years. Laboratory findings, including tumor markers such as CEA and CYFRA, were normal. We considered the lesion to be a probable focal inflammation and kept it under observation. A follow-up CT scan 4 months later, however, demonstrated no change in the appearance of the lesion. We strongly suspected the lesion to be a BAC and decided to perform surgery for this diagnosis, with the informed consent from the patient and her family. Although we planned to perform lingual segmentectomy by VATS with mini thoracotomy, the lesion was not identified by palpation. We finally decided to perform a left upper lobectomy to guarantee an optimal margin by

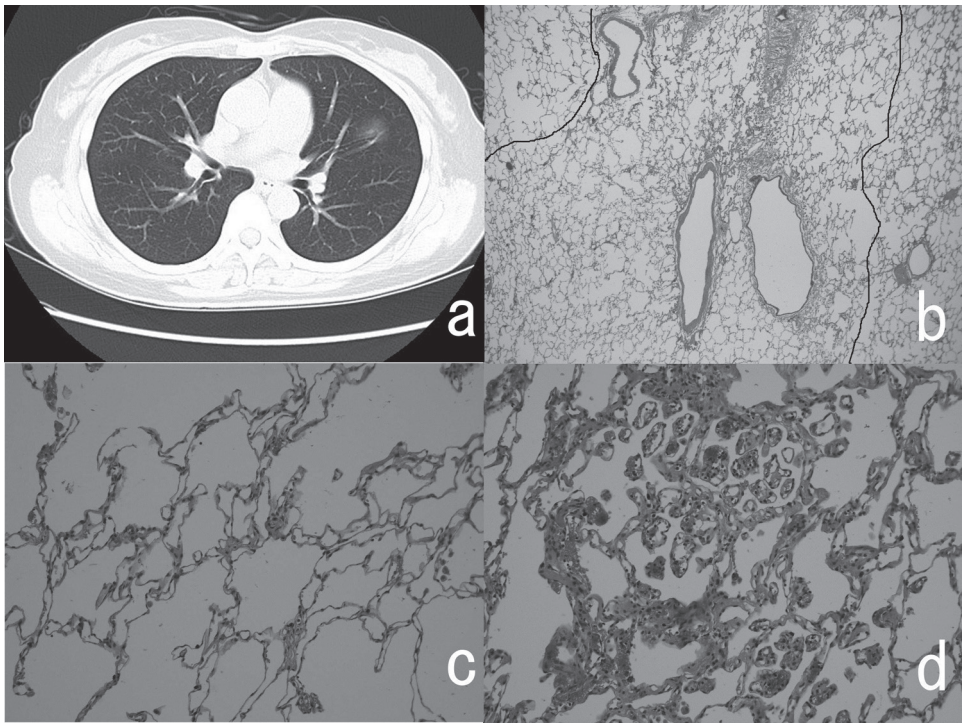

Fig. 1 (**a**) Chest computed tomography (CT) showing a pure ground-glass opacity (GGO) lesion measuring 20 mm in diameter in the left S4 near the boundary between S^4 and S^{1+2}. (**b, c**) The lesion shows thickening of alveolar septa caused by the proliferation of capillary vessels without cytological atypia. (**d**) A thickened alveolar wall and partial glomerular-like change are shown.

VATS. A pathologic examination showed thickening of alveolar septa caused by the proliferation of the capillary vessels without cytological atypia and immunohistochemical staining for CD34 in the lesion. Final diagnosis was a solitary pulmonary capillary hemangioma (**Fig. 1b, 1c, and 1d**). The postoperative course was uneventful, and she was followed up for 15 months after surgery with no recurrence.

Discussion

Pulmonary capillary hemangiomatosis (PCH) has been described as a disease that presents as multiple nodules in the lung. Prognosis of PCH is poor because it is characteristically associated with pulmonary hypertension or veno-occlusive disease.[1)] On the other hand, solitary pulmonary capillary hemangioma (SPCH), named by Fugo, is a rare disease that is characterized by a good prognosis because it is not associated with a specific clinical disorder.[2)] To our knowledge, only a few cases of SPCH in infants and 7 cases in adults have been reported.[2–7)]

Before Fugo's first SPCH report, Havlik, et al. had reported 8 autopsy cases with solitary or paired PCH-like foci that were found incidentally in the lung. These 8 cases did not present with pulmonary hypertension, and PCH was unrelated to the cause of death.[8)] Fugo pointed out that a few of Havlik's cases were applicable to PCH in a clinical setting, such as ageand the absence of specific symptoms.[2)]

On reviewing 8 SPCH cases, there were 4 males and 4 females, and the age range of SPCH cases was 45–59 years (mean age 53.1 years ± SD 4.2) (**Table 1**). Havlik's cases were 8 males, and the age range was 56–77 years (mean age 67.9 years ± SD 6.3).[8)] There were 17 males and 18 females in the previously reported PCH cases, and the age range was 6–71 years (mean age 34.5 years ± SD 19.1).[9)] The prognosis of PCH cases was 28 deaths for several days to years after the diagnosis, 3 cases cured by surgery, 2 cases improved by IFN –α-2a and 2 who were still living.[9)] In contrast to PCH cases, the mean age of SPCH cases was higher than PCH cases, and all SPCH cases were alive in good health. These results suggest that SPCH is differentiated clinically

Table 1 Patient profile of SPCH

Case	Sex/Age	Detected	CT finding	Chest Xp	Location/Surgery	Pathological finding
Case 1/Fugo	Male/56	medical checkup	GGO with partial solid lesion 13 × 12 mm	ground-glass opacity	Left lower lobe/ partial resection of Left lower lobe; S9+10	The lesion showed thickening of alveolar septa caused by the proliferation of the capillary vessels without cytological atypia or an inflammatory background. Staining for CD34
Case 2/Fugo	Female/48	medical checkup	GGO with partial solid lesion 13 × 12 × 8 mm	not described	Right middle lobe/ partial resection of Right middle lobe	Same as Case 1W
Case 3/Yanagawa	Male/58	medical checkup	Thick GGO 8 mm	not described	Right lower lobe/ partial resection of Right middle lobe	The lesion showed thickening of alveolar septa caused by the proliferation of the capillary vessels without cytological atypia. Staining for CD31 and CD34
Case 4/Uekami	Female/54	medical checkup	Solid nodule 12 × 11 × 8 mm	No finding	Right middle lobe/ peripheral: partial resection of Right middle lobe; s4	The lesion showed thickening of alveolar septa caused by the proliferation of the capillary vessels with slightly cytological atypia. Staining for CD34
Case 5/Kato	Male/55	medical checkup	GGO with partial solid lesion 11 mm	No finding	Right lower lobe/ Segmentectomy of Right lower lobe; S8	The lesion showed thickening of alveolar septa caused by the proliferation of the capillary vessels without cytological atypia. Staining for CD31 and CD34
Case 6/Hakiri	Male/45	medical checkup	GGO with partial solid lesion 12 × 11 mm	small nodule	Left lower lobe/ partial resection of Left lower lobe with marking; s9	The lesion showed thickening of alveolar septa caused by the proliferation of the capillary vessels without cytological atypia. Staining for CD31 and CD34
Case 7/Taniguchi	Female/59	medical checkup	Thick GGO 11 × 5 mm	Thin small nodule	Right lower lobe; peripheral/partial resection of Right lower lobe; S9	The lesion showed thickening of alveolar septa caused by the proliferation of the capillary vessels without cytological atypia. Staining for CD34
Case 8/Our Case	Female/53	medical checkup	Thick GGO 20 × 20 × 18 mm	No finding	Left upper lobe/left upper lobectomy;	The lesion showed thickening of alveolar septa caused by the proliferation of the capillary vessels and partial glomerular-like change. Staining for CD31 and CD34

from PCH. SPCH was reported as a good prognosis, but the long-term follow up data had never been reported. Havlik's cases, in which PCH was unrelated to the cause of death, were probably the natural course of SPCH. The difference between the mean age of Havlik's cases and SPCH cases might be one of evidences for a good prognosis in SPCH.

All SPCH cases were without symptoms and detected by a medical checkup. 7 SPCH cases showed a GGO with a high intensity or partial solid legion, and only one case showed a solid legion on CT (**Table 1**). These differences in image findings reflected the density of the capillary vessels proliferation because all SPCH cases had the same pathological findings, which showed thickening of alveolar septa caused by the proliferation of capillary vessels without cytological atypia (**Table 1**). SPCH and Havlik's cases have pathological findings in common, same as the SPCH cases, and all Havlik's cases showed a thickening of alveolar septa caused by the proliferation of capillary vessels without cytological atypia.[8] Additionally, there were no Havlik's cases that showed substantial luminal obstruction or secondary thromboembolism by proliferating capillary vessels.[8]

The cause of SPCH and PCH remain unknown, but evidence of increased expression of vascular endothelial growth factor and platelet-derived growth factor activity in patients with PCH have been reported.[10] In PCH case, these factors might be locally related to the cause of lesions.

It was reported to be easy to identify the SPCH which was located near pleura and to be excised by sublobar resection or segmentectomy for diagnosis and treatment (**Table 1**). In our case, the lesion was only detected as a pure GGO on CT, and was difficult to be examined by palpation during the operation. The recent spread of CT leads to the facile detection of a small-sized GGO lesion, which is probably a bronchioloalveolar carcinoma, an atypical adenomatous hyperplasia, or a focal infective lesion. It is difficult to distinguish a malignant lesion from a small-sized GGO on CT or FDG-PET. Because the diagnostic methods such as a bronchoscopic examination or a CT-guided needle biopsy are not helpful, the operation for diagnosis is often required. Although standard surgical treatment for non-small cell lung cancer is a lobectomy, sublobar resection or segmentectomy within the optimal margin is acceptable for treatment for BAC presenting pure GGO less than 20 mm in diameter. In our case, however, the optimal margin could not be decided; thus, lobectomy was selected.

8 SPCH cases have been reported in the past, and Havlik's cases might be applicable to SPCH.

Conclusion

SPCH should be considered in the differential diagnosis of a GGO lesion.

Disclosure Statement

None declared.

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