Single Pulmonary Langerhans Cell Histiocytosis Mimicking Metastasis in Lung Cancer: Imaging with ^{18}F -FDG PET/CT

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We report a rare case of single pulmonary Langerhans cell histiocytosis synchronous with lung cancer in the contralateral lobe. Chest CT as part of a general health checkup revealed two abnormal pulmonary nodules. Because ^{18}F -FDG PET/CT showed increased ^{18}F -FDG uptake in both nodules, each was resected for exact staging, revealing adenocarcinoma and Langerhans cell

histiocytosis, respectively. To the best of our knowledge, this may be the first report of pulmonary Langerhans cell histiocytosis presenting as a single nodule simultaneously with lung cancer in a contralateral lobe. However, due to its hypermetabolism, PET/CT failed to differentiate Langerhans cell histiocytosis from malignancy (Figs. 1 and 2).

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Fig. 1 Contrast-enhanced computed tomography (CT) reveals one solid nodule in the right upper lobe (**a–b**, empty arrows) and another 1.3-cm cavitary nodule in the left upper lobe (**c–d**, arrow heads). Multiple mediastinal lymph nodes are enlarged on the right side (not shown).

These findings are considered highly suggestive of lung cancer with metastatic lymphadenopathy in the right upper lobe, with possible lung-to-lung metastasis or double primary malignancy in the left upper lobe

Fig. 2 ^{18}F -FDG PET/CT demonstrates a hypermetabolic pulmonary nodule in the right upper lobe with a maximal standardized uptake value (SUV_{max}) of 5.3 (**a–b**, **e**, empty arrows) and another mild focal ^{18}F -FDG uptake in the left upper lobe with a SUV_{max} of 2.9 (**c–e**, arrow heads), also suggestive of metastasis or malignancy in the left upper lobe. Multiple hypermetabolic lymph nodes are noted in the right lower paratracheal and subcarinal space, likely indicating lymph nodes metastasis (**e**). Histopathological analysis of the wedge-resected biopsies of both upper lobes reveal moderately differentiated adenocarcinoma in the right upper lobe and Langerhans cell histiocytosis (LCH) positive for CD1a and S-100 protein in the left upper lobe (**f**; $\times 1.25$, **g**; $\times 400$, hematoxylin and eosin stain, **h**; $\times 400$, CD1a immunohistochemical stain). After the diagnosis, right upper lobectomy and mediastinal lymph node dissection were performed. The pathologic stage of lung cancer was T1aN2M0 (stage IIIA). LCH is rare and can be distinguished by unrestrained Langerhans cell proliferation and infiltration of various organs [1, 2]. Pulmonary Langerhans cell histiocytosis (PLCH) usually occurs in adult smokers but can be part of a multiorgan disease [1, 3]. Based on epidemiologic data in Japan, the estimated crude prevalences of PLCH in males and females are 0.27 and 0.07 per 100,000 [4]. The symptoms of PLCH are non-specific and include dyspnea, cough, and chest pains. Because it has characteristic findings on chest CT (nodules and cysts predominantly in the upper lung zones with relative sparing of the lung base), PLCH is usually diagnosed by imaging [5–7]. However, PLCH presenting as a single pulmonary nodule, as in the present case, is very rare. ^{18}F -FDG PET/CT detects PLCH with various ^{18}F -FDG uptake patterns according to disease stage; early stages are more likely to show intense ^{18}F -FDG uptake [1, 3, 8]. Krajicek et al. reported that the SUV_{max} in PLCH varies from 0.6 to 18.2 and attributed ^{18}F -FDG uptake in PLCH to granulomas [1]. Therefore, high ^{18}F -FDG uptake on PET may yield false-positive findings in patients with cancer. PLCH coexisting with lung cancer is quite rare, and the association between the two is still unclear [9–11]. Few cases of coexisting lung cancer and PLCH without extrapulmonary lesions on ^{18}F -FDG PET/CT have been reported. To the best of our knowledge, the present case may be the first report to observe PLCH presenting as a single nodule simultaneously with lung cancer in the contralateral lobe, mimicking metastasis or double primary malignancy in both chest CT and ^{18}F -FDG PET/CT

Disclosure

Conflict of Interest Heesung Song, Dakeun Lee, Seong Yong Park, Young-Sil An, Joon-Kee Yoon, and Su Jin Lee declare that they have no conflict of interest and no source of funding.

Informed Consent All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation and with the Helsinki Declaration of 1975, as revised in 2000. Informed consent was obtained from the patient for being included in the study.

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