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Case Report

Pulmonary lymphatic involvement in metastatic uterine sarcomas: imaging and pathological appearance ☆

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ABSTRACT

Pulmonary lymphatic involvement of sarcomas is an extremely rare form of metastases. We report the computed tomography (CT) features of pathologically confirmed pulmonary lymphatic involvement from metastatic uterine sarcomas. The CT illustrated smooth or nodular thickenings of the interlobular septa and bronchovascular bundle. Moreover, ground-glass opacity along the interlobular septa was also detected. These findings suggest that lymphatic involvement has diagnostic value for detecting this rare form of metastatic sarcomas. We also discuss possible differential diagnoses in this case and review previous cases reporting pulmonary lymphatic involvement in metastatic sarcomas. To the best of our knowledge, this is the first report describing pulmonary lymphatic involvement in metastatic uterine sarcomas. Pulmonary lymphatic spread of sarcomas is a rare form of metastatic sarcomas, but it should be considered when these findings suggesting lymphatic involvement are detected on CT.

Introduction

Pulmonary lymphangitic spread of carcinomas can be seen in metastatic lung disease and is shown as diffusely spread through the pulmonary lymphatic vessels in advanced carcinomas. On the other hand, the common finding in pulmonary metastatic sarcomas is multiple tumor nodules, and pulmonary lymphangitic spread of sarcomas is extremely rare. As far as we know, there are only a few case reports describing a correlation between the imaging and pathological characteristics of pulmonary lymphangitic spread of sarcomas, and this is the first case reporting pulmonary lymphatic involvement from metastatic uterine sarcomas. In this report, we

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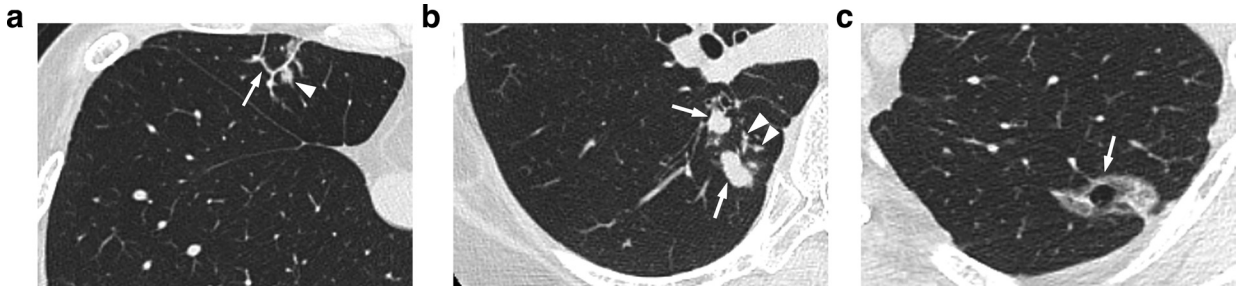

Fig. 1 – The unenhanced computed tomography scan in the axial plane. (A) a smooth thickening (arrow) and a nodular thickening (arrowhead) of the interlobular septa surrounding a lobule in the right middle lobe; (B) irregular nodular thickenings (arrows) along the bronchovascular bundle, and smooth thickenings (arrowheads) of the interlobular septa in the right lower lobe; (C) well-circumscribed ground-glass opacity (arrow) along the interlobular septa surrounding a lobule in the left upper lobe.

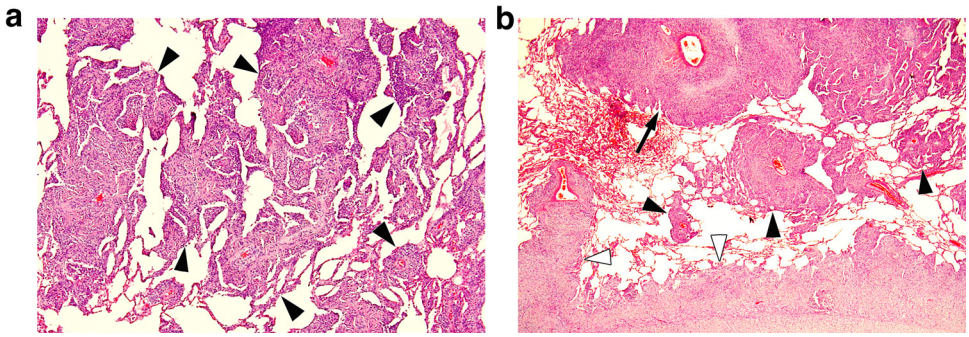

Fig. 2 – Pathological findings of the right middle lobe (hematoxylin and eosin stain). (A) thickenings of the perivascular interstitium (black arrowheads) by spindle-shaped tumor cells, which are consistent with metastases of known uterine sarcoma (original magnification: 40x); (B) thickenings of the bronchovascular bundle (black arrow), perivascular interstitium (black arrowheads), interlobular septa, and subpleural area (white arrowheads) (original magnification: 100x).

present a 37-year-old-woman with a history of postoperative uterine sarcoma and radiological-pathological correlation of pulmonary lymphatic involvement of sarcomas.

Case report

Our patient was a 37-year-old-woman with a history of surgical resection of a uterine sarcoma and pelvic lymph node metastases 2 years ago, radiotherapy for a cervical spine metastasis, and resection of a solitary pulmonary metastasis in the right lower lobe last year. The patient had continued chemotherapy after the surgery, but a follow-up ^{18}F-fluorodeoxyglucose positron emission tomography/ computed tomography showed bilateral lung metastases. The patient was referred to our hospital for the purpose of surgical resections of the bilateral lung metastases. The patient had no respiratory symptoms and the laboratory test results were normal. The patient underwent a chest radiograph and a computed tomography (CT) scan as preoperative assessments for multiple pulmonary metastases. No obvious abnormalities were noted on the chest radiograph. The chest CT revealed several known localized thickenings of the interlobular septa or bronchovascular bundle in the bilateral lungs (Fig. 1). All lesions were approximately 3 cm or less in size. The lesion in the right middle lobe showed smooth and nodular thickenings of the interlobular septa surrounding a lobule (Fig. 1A). The lesion in the right lower lobe showed irregular nodular thickenings along the bronchovascular bundle, and smooth thickenings of the interlobular septa (Fig. 1B). Additionally, the lesion in the left upper lobe showed well-circumscribed ground-glass opacity along the interlobular septa surrounding a lobule (Fig. 1C). The lung architecture at the lesions was preserved. No tumor invasion into the chest wall or pleural effusion was observed. No hilar or mediastinal lymphadenopathy was detected. These CT findings suggested pulmonary metastases of the uterine sarcoma with lymphatic involvement. Subsequently, partial resections of the lung lesions were performed. The pathological examination demonstrated spindle-shaped tumor cells in the perivascular interstitium, interlobular septa, and subpleural area along to the lymphatic vessels (Fig. 2), which was suggestive of lymphatic involvement from uterine sarcoma metastases. No malignant cells were found in cytology of pleural effusion or pathology of regional lymph nodes resected at the same time. The postoperative course was uneventful and the patient returned to the referral hospital. After that, the patient underwent resection of a disseminated lesion in the pelvis at another hospital. However, the patient died 1 year after the lung surgery due to exacerbation of the disease.

Discussion

The predominant pathway for pulmonary metastatic sarcomas is through hematogenous dissemination and tumor embolization, hence metastatic sarcomas of the lung are observed primarily as randomly distributed nodular lesions [1]. Thus, pulmonary metastatic sarcomas are generally not recognized as showing lymphangitic spread and Liau et al. named this rare form of metastases "pulmonary lymphangitic sarcomatosis" (PLS) [1].

According to a previous report, pulmonary lymphangitic carcinomatosis (PLC) most often coexists with adenocarcinoma (80%), while PLC induced by sarcomas are rare (3.6%) [2]. To the best of our knowledge, three previous reports have described the CT findings of the patients with PLS in detail; two patients with angiosarcoma, one with neuroectodermal/Ewing's sarcoma [1,3,4]. These findings included uneven thickenings of the interlobular septa and bronchovascular bundle, which were similar to the CT findings of PLC. Similar to the previous articles, the chest CT images of our patient showed several localized smooth or nodular thickenings of the interlobular septa, part of which had nodular thickenings of the bronchovascular bundle. However, ground-glass opacity along the interlobular septa due to tumor extension has not been reported previously for PLS. Although there have been a few case reports describing relative diffuse ground-glass opacity on CT in the patients with PLC, the well-circumscribed and localized ground-glass opacity along the interlobular septa like this case has not been reported previously [5–7].

Differential diagnoses in this case included pulmonary edema, infectious lesions, and sarcoidosis. These nodular patterns are atypical in cases with only pulmonary congestion. The nodular lesions in sarcoidosis are usually symmetrical with the predominance in the upper and middle lung zone. Furthermore, the distortion of lung architecture is a crucial finding of sarcoidosis [8]. However, these findings were not seen in this case; bronchial pneumonia and post-inflammatory changes were also considered, but the course was atypical. Interstitial pneumonia and infections, including Covid-19 pneumonia, could be the differential diagnoses of diffuse ground-glass opacity; however, these diagnoses were not positively suggested in this case because of the well-circumscribed and localized nature of the ground-glass opacity surrounding the lobule [5–7,9].

Regarding the mechanism of PLS, two metastatic pathways can be considered in the same way reported in PLC: one is hematogenous metastases to the lung followed by interstitial and lymphatic invasion, the other is direct extension of tumor cells from hilar or mediastinal lymph nodes [10]. As opposed to carcinomas, sarcomas that shows metastases to lymph nodes account for only 2% to 5% [1]. Meanwhile, two of the three patients with PLS mentioned above showed hilar or mediastinal lymphadenopathy [1,3]. This difference may be explained by that PLS is more likely to be found in a more advanced state of the disease. In this case, findings suggesting these metastatic pathways were not observed.

As for uterine sarcomas, there has been a single case presenting a "tree-in-bud" pattern for pulmonary metastases, but PLS of uterine sarcoma has not been reported previously [11]. Because of these rare pathways of metastatic sarcoma, we cannot rule out the metastases from these findings. In our case, surgical resections were performed and the pathological examination revealed the extension of tumor cells along the lymphatic spaces.

In summary, pulmonary lymphatic involvement is a rare form of metastatic sarcomas, but it should be considered when these CT findings suggesting lymphatic involvement are detected.

Patient Consent

No written consent has been obtained from the patient as there is no patient identifiable data included in this case report and the patient is deceased.

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