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Case Report *Diagnostic Radiology*

Incidental detection of benign metastasizing leiomyoma in asymptomatic female

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ABSTRACT

Benign metastasizing leiomyoma (BML) is a rare finding of histologically benign smooth muscle tumors in extrauterine locations, most commonly the lungs. We report a case of BML found incidentally on pre-operative imaging in a 42-year-old patient. BML is found in premenopausal women with a history of leiomyoma and, often, hysterectomy. As in our case, the metastatic pulmonary nodules are not hypermetabolic on 18F-fluorodeoxyglucose Positron emission tomography/computed tomography. BML may be clinically malignant or asymptomatic. Since the imaging appearance of BML simulates metastatic disease of more malignant etiology, awareness of its multimodality imaging appearance and presentation can aid in diagnosis.

Keywords: Benign, Metastatic, Leiomyoma, Hysterectomy, Case report, Solid pulmonary nodule

INTRODUCTION

Benign metastasizing leiomyoma (BML) is a rare disease in which uterine leiomyomas are found in extrauterine sites, most commonly the lungs. The clinical presentation is often asymptomatic and discovered incidentally on chest imaging of premenopausal women. We report a case of a 42-year-old woman diagnosed with pulmonary BML 3 years after a hysterectomy.

CASE REPORT

A 42-year-old female presented for pre-operative clearance for a Roux-en-Y gastric bypass. Review of symptoms was negative. Her medical history was significant for morbid obesity, hyperlipidemia, and Type 2 diabetes mellitus. Her surgical history included a hysterectomy performed in another city (for unknown reason at the time of imaging). The patient denied personal or family history of cancer. Pre-operative workup including electrocardiogram and laboratory work (complete blood count, basic metabolic panel, and prothrombin time/international normalized ratio) were unremarkable.

Pre-operative chest radiographs revealed numerous small bilateral lung nodules [Figure 1]. Subsequent chest computed tomography (CT) confirmed numerous, well-circumscribed solid nodules throughout bilateral lungs measuring up to 1.2 cm [Figure 2]. At this point, the leading consideration was metastatic disease from an unknown primary site. Pulmonology was

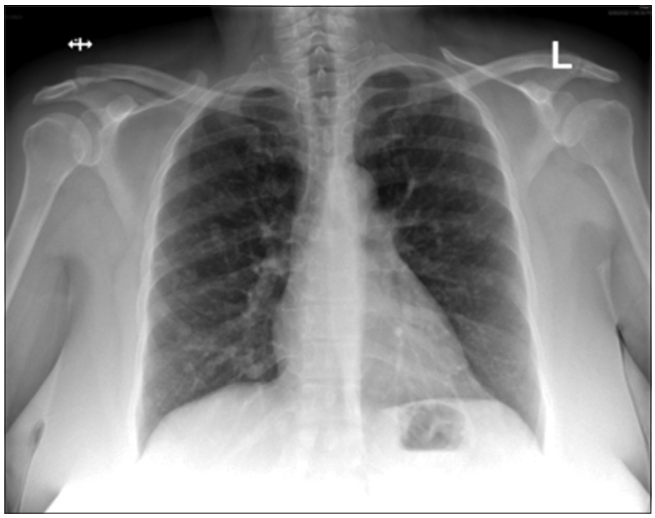

Figure 1: A 42-year-old asymptomatic female presenting for pre-operative clearance before a Roux-en-Y gastric bypass. The posteroanterior chest radiograph shows multiple bilateral pulmonary nodules.

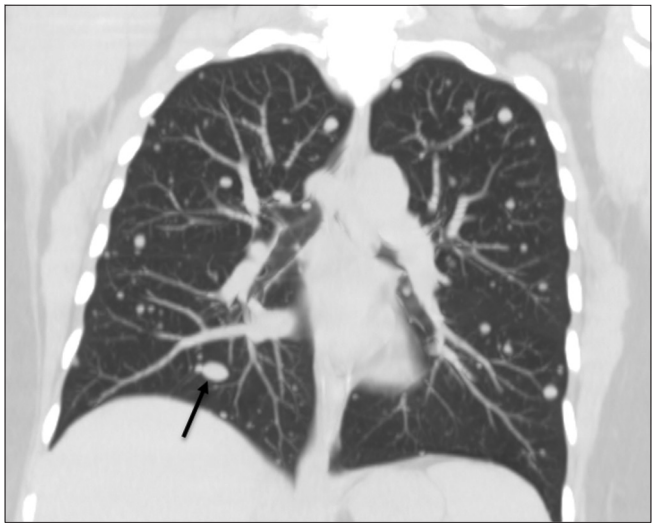

Figure 2: A 42-year-old asymptomatic female with numerous pulmonary nodules. Reformatted coronal computed tomography maximum intensity projection image confirms numerous well-circumscribed solid nodules throughout bilateral lungs measuring up to 1.2 cm (arrow).

consulted. Positron emission tomography (PET)/CT using 18-F fluorodeoxyglucose (FDG) was ordered to assess for metabolic activity in the nodules and to survey for a primary site of disease. No abnormal hypermetabolic foci were present on the PET/CT [Figure 3]. There was no evidence of a hypermetabolic primary neoplasm, and the solid pulmonary nodules demonstrated a maximum standardized uptake value of 1.3 (below mediastinal blood pool activity). Note was made of a prior hysterectomy on the PET/CT and the possibility of BML was suggested. Additional non-

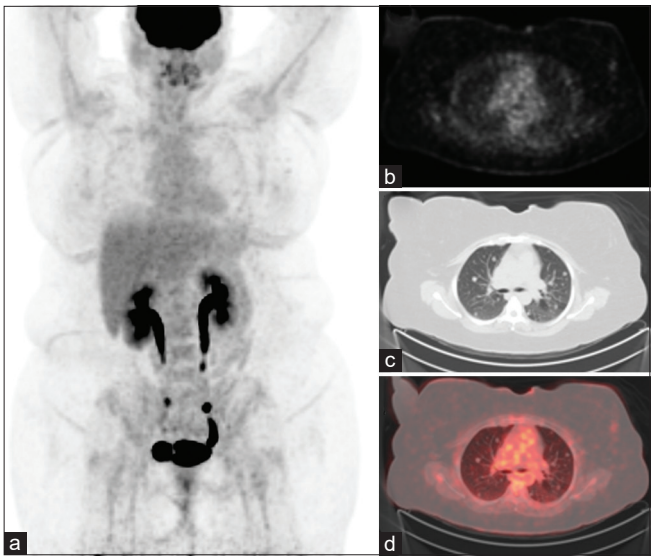

Figure 3: A 42-year-old asymptomatic female with numerous solid pulmonary nodules. (a) 18-fluorodeoxyglucose (FGD) Positron Emission Tomography (PET)/Computed Tomography (CT) maximum intensity projection does not reveal any abnormal hypermetabolic activity. Axial PET (b), CT (c), and fused PET/CT (d) images show the pulmonary nodules have minimal FDG uptake (less than mediastinal blood pool).

hypermetabolic metastatic etiologies were posed, included benign metastasizing meningioma, giant cell tumor of bone (GCTB), and thyroid adenoma.

Additional history from the patient revealed, she had obtained the hysterectomy for uterine fibroids. There was a multidisciplinary consensus to biopsy the nodules. The patient underwent video-assisted thoracoscopic surgery with wedge resection of all three lobes of the right lung. Histopathological examination revealed estrogen receptor positive bland spindle cell proliferation with entrapped Type II pneumocytes, consistent with BML [Figure 4]. The patient was referred to gynecologic oncology for further management. A 3-month follow-up CT demonstrated stability of existing nodules, and the patient was started on monthly leuprorelin injections. After an additional 3-months, the patient endorsed minimal side effects from treatment and CT redemonstrated nodule stability. She remains asymptomatic and continues to follow with gynecologic oncology for monthly injections and disease monitoring.

DISCUSSION

BML involves histologically benign smooth muscle tumors in extrauterine locations, with the lungs being the common site of metastasis. Since Steiner first proposed the diagnosis in 1939, at least 164 cases have been described in literature.[1,2] Most cases have been reported in women of late childbearing age with a history of uterine leiomyoma. The previous

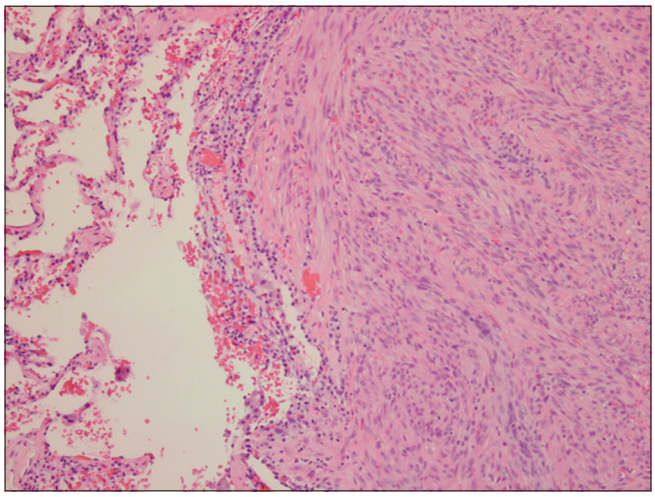

Figure 4: A 42-year-old asymptomatic female with numerous solid pulmonary nodules. Cytology from lung nodule shows bland spindle cell proliferation with entrapped Type II pneumocytes. The bland spindle cells are positive estrogen receptor. Taken together, the findings are consistent with metastasizing leiomyoma.

hysterectomy for fibroids is a typical finding.[3] BML is often diagnosed as a result of incidentally found lung nodules on chest imaging of asymptomatic patients.

The differential diagnosis for multiple pulmonary nodules is extensive and includes malignancy. As with our patient, 18F-FDG PET/CT may be used for further nodule characterization. BML lesions are not FDG avid. Literature assessing the prevalence and significance of non-FDG avid pulmonary lesions is lacking. One study of patients between 2003 and 2005 demonstrated that 19% of incidental non-FDG avid lung nodules turned out to be malignant.[4] However, subjects of this study were known to have extrathoracic malignancies. In addition, sensitivity has improved with technological advancement in PET/CT since this study was conducted. It is assumed that non-FDG avid solid pulmonary nodules measuring at least 0.8 cm are likely benign. The differential diagnosis for benign pulmonary nodules includes BML, benign metastasizing meningioma, benign adenoma of the thyroid, and GCTB. Pulmonary metastases are the most common extracranial manifestation of meningioma, although they are exceedingly rare as seen in a 2023 systematic review which counted 13 reported cases.[5] Only seven cases of intrapulmonary benign thyroid tissue have been reported as of 2015, and all of them involved a single lung lesion as opposed to the numerous nodules seen in our patient.[6] Lung metastases from GCTB are also rare and tend to occur very late in the disease course, typically in the setting of locally recurrent disease.[7] GCTB is aggressive and commonly presents symptomatically at its primary site.

Given their technically benign determination, there is the option to forego treatment of BML and manage conservatively with periodic imaging. Although often asymptomatic, BML may become clinically malignant. Steiner's earliest literature on BML reports a case of pulmonary metastases causing heart failure and, ultimately, death of a 36-year-old woman.[1] Since this 1939 report, numerous case reports have shown BML to cause pulmonary symptoms and complications including pneumothoraces.[3,8] Several theories have been proposed for the pathogenesis of BML, including hematogenous spread through peritoneal seeding during hysterectomy. While some evidence has been found in support of hematogenous spread, the validity of seeding during hysterectomy seems less certain.[9] BML is most commonly discovered in women status post-hysterectomy; however, there have been at least 10 reported cases in women with no surgical history.[10] Although the underlying etiology and pathogenesis of uterine leiomyomas and their metastases remain ambiguous, leiomyomas are regarded as estrogen dependent tumors. They may progress with a high estrogenic state, such as our patient's pre-menopausal status and adiposity. BML can be treated with estrogen reduction through hormone therapies or oophorectomy/hysterectomy, offering a favorable prognosis with stability or regression of nodules.[3] Both recent gastric bypass and leuprorelin therapy are contributing to reduction of estrogen in our patient.

CONCLUSION

We report a case of incidental lung nodules concerning for malignancy. 18-F FDG PET/CT revealed non-FDG avid lesions and histopathology confirmed uterine origin. This case suggests the inclusion of BML in the differential diagnosis of pulmonary nodules in an asymptomatic and premenopausal woman with a history of leiomyoma.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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