



Pulmonary benign metastasizing leiomyoma presenting as small, diffuse nodules

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TO THE EDITOR:

We report the case of a 46-year-old nonsmoking female patient with controlled asthma, under regular treatment with a combination of inhaled fluticasone and salmeterol. She presented with a one-year history of cough, wheezing, and dyspnea on moderate exertion. There was no history of weight loss, hemoptysis, chest pain, environmental exposure, or drug exposure. On physical examination, the uterus was palpable and firm, the uterine fundus being midway between the symphysis pubis and the umbilicus. The remainder of the physical examination was normal. Sputum examination, blood biochemistry, and tumor markers (including CA 19-9 and CA 125) were all normal. Spirometry showed mild obstructive lung disease and positive bronchodilator test results. Chest X-rays (Figure 1A) and chest CT scans (Figures 1B and 1C) showed numerous ill-defined, confluent, rounded, nodular opacities of 2-10 mm in diameter in both lungs. Bronchoscopy was normal. Bronchial lavage fluid cytology was negative for infectious and malignant etiologies. Transbronchial lung biopsy showed relatively ill-circumscribed nodules consisting of interlacing bundles of uniform spindle cells with oval nuclei and inconspicuous nucleoli in a background of collagen, the adjacent lung parenchyma being compressed (Figure 1D). Immunohistochemistry was positive for vimentin, smooth

muscle actin, and desmin, as well as for estrogen and progesterone receptors, but was negative for cytokeratins and p63, the nodules therefore originating from uterine smooth muscle cells. The Ki-67 proliferation index was < 5%, which is typical of benign smooth muscle tumors. A diagnosis of benign metastasizing leiomyoma (BML) was made on the basis of histological and imaging findings. At this writing, the patient is receiving monthly treatment with goserelin acetate on an outpatient basis and undergoing preoperative evaluation for hysterectomy and oophorectomy.

Pulmonary BML is a rare condition characterized by benign uterine leiomyoma metastasizing to the lung. Although pulmonary BML is most commonly seen in women of reproductive age presenting with a history of uterine leiomyoma and undergoing hysterectomy, it can affect women who have not undergone hysterectomy, as was the case here. Metastatic spread is believed to occur through the blood. Although the lung is the most common metastatic site, BML can metastasize to the lymph nodes, central nervous system, mediastinum, bones, and heart.⁽¹⁻⁴⁾ Given that most patients are asymptomatic, pulmonary BML is usually an incidental finding on routine chest X-rays. However, patients with pulmonary BML can present with cough, hemoptysis, dyspnea, and reduced lung function. Typical imaging findings include multiple

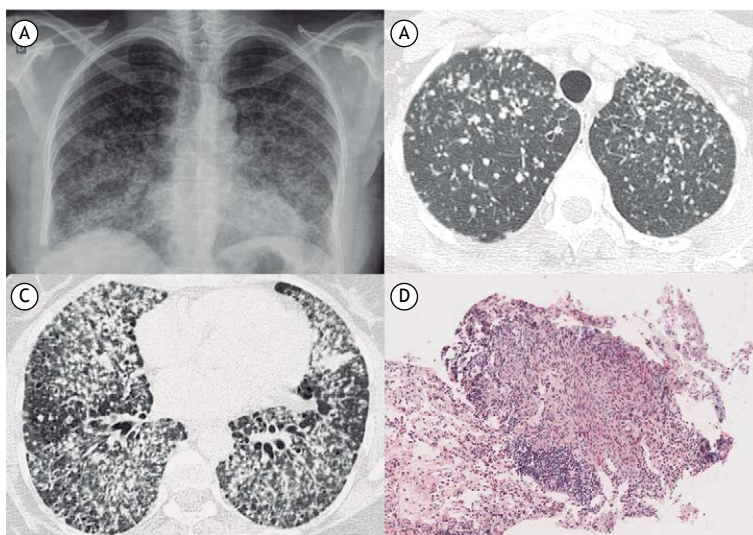


Figure 1. In A, posteroanterior chest X-ray showing diffuse infiltration of the lung parenchyma by small pulmonary nodules. In B and C, axial CT scans showing confluent nodules of varying sizes with irregular margins. In D, histological staining showing a nodule with ill-defined margins, consisting of interlacing bundles of spindle-shaped cells without atypia, with low mitotic activity and without necrosis (H&E; magnification, ×10).

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(but no more than ten) pulmonary nodules of varying sizes. However, pulmonary BML can present as cystic, cavitory, or miliary lesions.⁽²⁻⁴⁾ Metastatic uterine leiomyosarcoma is the main differential diagnosis.⁽²⁾ Surgical biopsy with immunohistochemical staining is the gold standard for the diagnosis of pulmonary BML.^(2,5) In the case reported here, the histopathological features were characteristic of pulmonary BML, including interlacing bundles of smooth muscle cells without vascular invasion or cellular atypia, as well as very low mitotic activity. Multiple treatment options have been described, including watchful waiting, surgical resection, and antiestrogen therapy.⁽²⁾ BML tends to have an indolent course and a favorable outcome, and

can regress spontaneously after menopause, although lung lesions can continue to progress, leading to pulmonary insufficiency and even death.^(2,4)

It is of note that, in our patient, lung lesions occurred concomitantly with uterine leiomyoma rather than having developed from it, as is commonly reported. The number of small, confluent nodules is also of note, being much higher than that reported in the literature.

In conclusion, BML should be included in the differential diagnosis of micronodules and diffuse pulmonary nodules in women. Attending physicians should collect information on symptoms and gynecological history in order to screen for uterine leiomyoma.

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