Multiple Benign Metastasizing Leiomyoma of the uterus in lung and retroperitoneum

Sir,

Benign metastasizing leiomyoma (BML) arises from a histologically benign uterine leiomyoma. It is characterized by multiple smooth muscle nodules, most often located in the lung, abdominal cavity, retroperitoneum, muscular tissue, lymph nodes, blood vessels, or heart. The coexistence of BMLs in the lung and retroperitoneum is extremely rare. Herein, we report a rare case of simultaneous occurrence of multiple BMLs in the lung and retroperitoneum of a 42-year-old woman who had previously undergone hysterectomy to treat benign uterine leiomyoma.

A 42-years-old female patient was referred for the evaluation of cough, abdominal pain, and abnormal chest X-ray findings. She had a history of multiple uterine leiomyomas and had undergone myomectomy at 30 years of age, subsequently undergoing total abdominal hysterectomy, because of recurrent multiple uterine leiomyomas, at 33 years of age. When she was 39 years of age, an abdominal computed tomography (CT) scan revealed a 5 cm × 4 cm nodule in the distal portion of the left para-aortic region and a 46 cm × 26 cm nodule just to the left of and posterolateral to the bladder. At that time, laparotomy, with excisional biopsy, was performed. Examination of the biopsy sample revealed a leiomyoma that tested positive for estrogen and progesterone.

In the initial physical examination at our facility, palpable masses were identified in the left upper and left lower quadrants of the abdomen. Routine laboratory tests showed that the values for biochemical variables, including those for the tumor markers such as alpha-fetoprotein, cancer antigen 19-9, and cancer antigen 125, were within the normal ranges. A chest X-ray revealed well-defined masses, with several nodules in both lung fields. The patient was submitted to CT scans of the chest and abdomen. In the lungs, the CT scans revealed multiple nodules with regular borders, distributed randomly and bilaterally, the largest with a diameter of 4.6 cm [Figure 1]. The abdominal scans showed a solid mass of approximately 18 cm × 13 cm, with regular borders and lobulated contours, that heterogeneously enhanced with contrast and filled the paravertebral area. To the right of and adjacent to the first abdominal mass were two additional masses, measuring 24 mm × 17 mm and 8 cm × 5 cm, respectively. In the pelvic adnexa, bilaterally, there were multiple solid lesions with lobulated contours, the largest being on the left and measuring approximately 4.6 cm × 3.3 cm. To identify the primary malignancy and metastatic lesions, we employed ^{18}F-fluorodeoxyglucose-positron emission tomography and CT (FDG-PET/CT). There was no pathologic FDG uptake to suggest the primary malignancy or metastatic lesions in any other part of the body. The retroperitoneal masses were excised. The patient subsequently underwent video-assisted thoracoscopic surgery with wedge resection of two of the nodules. Analysis of intraoperative frozen sections from the retroperitoneal tumors suggested leiomyoma. Microscopic examinations showed well-differentiated straight muscle cell bundles, although neither nuclear atypia nor mitotic activity was identified in the lung nodules or retroperitoneal masses. The proliferation index, as defined by the cell division marker Ki-67, was very low (<1%), and there was positive immunohistochemical staining for estrogen and progesterone receptors [Figure 2]. The pathological findings of both the lung nodules and retroperitoneal masses confirmed the diagnosis of BML. The patient was started on gonadotropin-releasing hormone (GnRH) agonist goserelin. At this writing, she is undergoing regular follow-up evaluations.

BML is a rare disease that originates from a histologically benign uterine leiomyoma, and is usually found 3 months

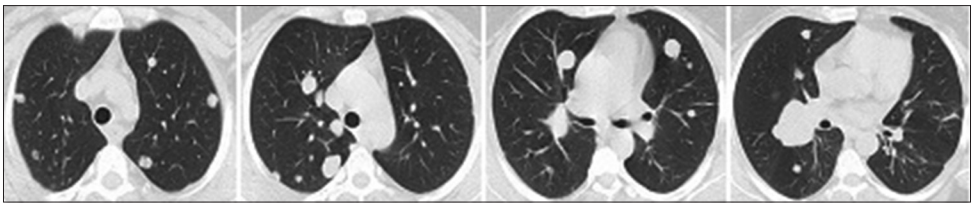

Figure 1: Chest computed tomography showing multiple pulmonary nodules

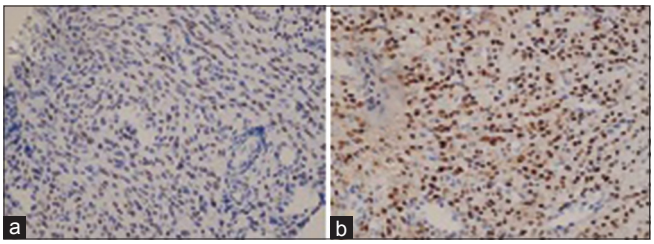

Figure 2: Immunohistochemical staining sections of the pulmonary nodule. Positive immunoreactivity for estrogen receptor (a) and progesterone receptor (b) was observed (×400)

to 20 years after hysterectomy or myomectomy.[1] Several theories related to the pathogenesis of BML have been proposed, one of which speculates that BML is a benign lesion that has the potential to metastasize to the lungs or other organs via a hematogenous route.[2]

BMLs are slow growing and are usually found incidentally. They have only a slight effect on lung function. Radiographically, BML appears as well-circumscribed nodules, varying sizes and can be found as solitary or multiple soft-tissue masses scattered throughout both lungs.[3] Pulmonary nodules can remain stable, decrease in size, or increase in size. The FDG-PET/CT scan revealed no evident concentration of radioactivity.

Histological examination shows that BML is characterized by the absence of malignant features such as hypercellularity, cytologic atypia, high proliferative status, and necrosis.[4] Various immunohistochemical markers, such as desmin, muscle-specific actin, and vimentin, confirm mesenchymal derivation with smooth muscle differentiation of these tumors.[5] The presence of estrogen and progesterone receptors strongly suggests BML of uterine origin, which supports the rationale of treatment with hormonal agents. Reversible medical castration with GnRH agonists, which suppress the endogenous gonadotropin secretions required for gonadal steroid production, has been reported to provide satisfactory therapeutic outcomes.[6]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

Aysel Sunnetcioglu[1], Erbil Karaman[2], Mesut Ozgokce[3], Remzi Erten[4], Remzi Kızıltan[5], Ufuk Cobanoglu[6]

[1]Department of Chest Diseases, Yuzuncu Yil University Medical Faculty, Van, Turkey, [2]Department of Obstetrics and Gynecology, Yuzuncu Yil University Medical Faculty, Van, Turkey, [3]Department of Radiology, Yuzuncu Yil University Medical Faculty, Van, Turkey, [4]Department of Pathology, Yuzuncu Yil University Medical Faculty, Van, Turkey, [5]Department of General Surgery, Yuzuncu Yil University Medical Faculty, Van, Turkey, [6]Department of Thoracic Surgery, Yuzuncu Yil University Medical Faculty, Van, Turkey.
E-mail: izciaysel@mynet.com

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