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Pelvic and pulmonary benign metastasizing leiomyoma: A case report

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ABSTRACT

Seven years after she had a total abdominal hysterectomy for benign leiomyomas, a 46-year-old woman presented with a pelvic mass and multiple pulmonary nodules. She underwent resection of the mass and core needle biopsy of a pulmonary lesion. Histopathologic analysis revealed that both the pelvic and the pulmonary lesions were consistent with benign leiomyomas. Benign metastasizing leiomyoma should be considered if a woman of reproductive age and with a history of leiomyomas presents with extrauterine nodules without evidence of malignancy. The final diagnosis should be based on histopathological examination. Treatment depends on tumor size, location, receptor positivity, and disease progression.

1. Introduction

Benign metastasizing leiomyoma (BML) is a rare disorder in which histologically benign smooth muscle tumor is found in extrauterine sites. The condition was first described in 1939 by Steiner in a report of a patient who died from the effects of extensive pulmonary metastases of benign-appearing leiomyomas that were histologically identical to multiple leiomyomas concurrently present in the uterus [1].

The condition typically affects women of late reproductive age with a history of leiomyomas, the majority of whom have undergone surgical management with myomectomy and/or hysterectomy [2]. The mean age of women at the time of primary surgery and BML diagnosis is 38.5 and 47.3 years, respectively. The most frequent site of metastasis is the lungs (80%), with other organs including the heart, liver, esophagus, abdominal lymph nodes, skeletal muscle, skin, and central nervous system occasionally reported [2]. We present the case of a woman who had undergone a distant total hysterectomy for benign leiomyomas who subsequently developed a local pelvic recurrence and was simultaneously found to have numerous benign pulmonary metastases.

2. Case Report

A 46-year-old Hispanic woman presented to her gynecologist for her annual exam. She had no significant past medical history and took no medications. Her past surgical history was notable only for an uncomplicated total abdominal hysterectomy without salpingo-oophorectomy for benign leiomyomas at the age of 39.

Physical examination was concerning for a large central pelvic mass, which was subsequently confirmed on pelvic ultrasound (Fig. 1). Computed tomography (CT) of the abdomen and pelvis was ordered to better characterize the mass and revealed a large heterogeneous solid mass measuring 16.4 cm × 13.0 cm × 18.2 cm arising centrally from the mid and upper pelvis. The mass appeared separate from the ovaries and bowel with well-defined margins and no associated fat or calcification (Fig. 2A–C). The CT also identified multiple bibasilar non-calcified pulmonary nodules measuring up to 4.2 cm × 2.3 cm noted to be highly suspicious for metastatic disease (Fig. 2D).

In order to establish a diagnosis, CT-guided needle core biopsy of the pelvic mass was performed. On microscopy, the specimen was notable for the proliferation of bland spindle cells without evidence of necrosis, cytologic atypia, or increased mitotic activity.

Immunohistochemistry revealed diffuse positivity for desmin and smooth muscle actin, confirming smooth muscle differentiation. Negative staining for the cell surface marker CD117 and the chloride channel protein DOG-1 excluded the diagnosis of gastrointestinal stromal tumor (GIST), another spindle cell neoplasm in the histomorphological differential. Taken together, the findings were consistent with a smooth muscle neoplasm without definitive features of malignancy.

The patient underwent an exploratory laparotomy and was found to have a 15 cm × 11 cm × 11 cm right para-adnexal mass densely adherent to the right tube and ovary. She underwent bilateral salpingo-

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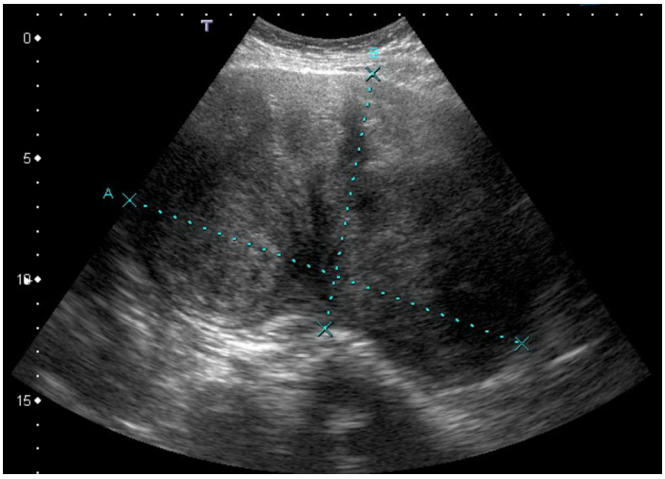

Fig. 1. Pelvic ultrasound with sagittal view of pelvic mass measuring 17.4 cm × 10.7 cm.

oophorectomy with resection of the mass as well as an opportunistic appendectomy. Final pathology confirmed the findings of the needle biopsy and supported the diagnosis of a benign leiomyoma. Given low concern for malignancy, the patient was started on transdermal estradiol 0.025 mg every 24 h as hormone replacement therapy (HRT) in the setting of surgical menopause following her bilateral oophorectomy.

Following her abdominal surgery, the patient was referred to a pulmonologist for evaluation of the concerning CT findings. On evaluation, the patient denied any symptoms of chest pain, hemoptysis, cough, or shortness of breath. She denied recent weight loss, and reported no history of smoking or recent travel. A positron emission tomography CT (PET-CT) was ordered to better characterize the pulmonary nodules which were found to have no increased 18F–fluorodeoxyglucose (18F–FDG) avidity or evidence of hypermetabolic activity (Fig. 3). The scan identified over 30 pulmonary nodules; however, given the low likelihood of primary pulmonary or metastatic cancer, the decision was made to observe the patient off HRT with repeat imaging in six months. The patient was unable to tolerate HRT cessation secondary to menopausal symptoms and was restarted on estrogen supplementation shortly thereafter. Repeat CT six months later revealed largely stable pulmonary nodules with no new lesions but with interval enlargement of the most prominent nodule at the right posterior medial lung base from 4.2 cm × 2.3 cm to 5.2 cm × 2.3 cm (Fig. 4) prompting a CT-guided percutaneous biopsy of the lesion. Final pathology confirmed the diagnosis of benign leiomyoma. Pathology was reviewed with that of the excised pelvic mass and felt to be cytologically similar (Figs. 5 and 6). Of note, the tumor was strongly estrogen and progesterone receptor positive (>95%). The risks and benefits of continuing HRT were discussed and the patient opted to discontinue estrogen supplementation.

The patient has continued to undergo CT surveillance every six months. Twelve months after her initial diagnosis, a scan again revealed a slight increase in the size of her pulmonary BML prompting referral to a gynecologic oncologist who started her on letrozole, an aromatase inhibitor, to further restrict her endogenous estrogen production. Two years later, surveillance imaging has confirmed stability of her pulmonary BML and the patient remains asymptomatic.

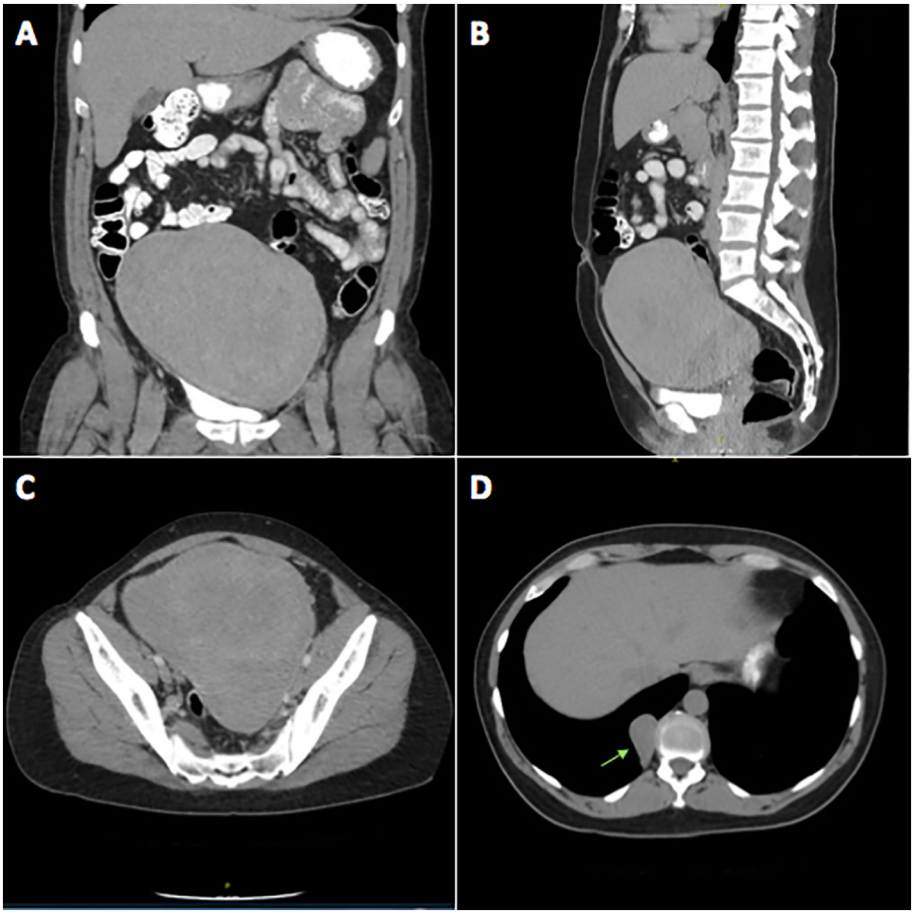

Fig. 2. CT abdomen and pelvis at the time of initial diagnosis. Large heterogeneous solid mass measuring 16.4 cm × 13 cm × 18.2 cm arising centrally from the mid and upper pelvis as seen in the coronal, sagittal, and axial planes, respectively (A–C). Largest of the incidentally noted bibasilar non-calcified pulmonary nodules at the right posterior medial lung base measuring 2.3 × 4.2 cm marked with the arrow (D).

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