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CLINICAL VIGNETTE

Pulmonary Benign Metastasizing Leiomyoma Presenting with Chronic Cough

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Case Presentation

The patient is a 57-year-old nonsmoking female who presented with nonproductive cough for two months. It was of insidious onset with no exacerbating or alleviating factors. She denied any systemic symptoms of fever, chills, or night sweats. There were no complaints of sinus or gastrointestinal symptoms. She reported a previous episode with similar symptoms 2 years prior that gradually abated after several courses of empiric antibiotics and inhaled bronchodilator therapy. Past medical history included hypertension treated previously with lisinopril that did not improve cough after discontinuation. She had undergone hysterectomy for leiomyoma and adenomyosis 10 years previously. The patient is a lifelong nonsmoker.

Physical examination revealed normal vital signs and pulse oximetry. Auscultation of the chest demonstrated normal breath sounds. The remainder of the examination was unremarkable.

CXR revealed a right lower lobe 18 mm smooth bordered pulmonary nodule. CT scan showed a 18X12mm well circumscribed RLL nodule, a 14X18mm subpleural nodule in the RML, a 7mm nodule in the anterior segment RLL, and a 6mm nodule in the lingula. PET scan showed no evidence of hypermetabolic activity. Review of a prior CXR 11 years ago retrospectively showed the presence of the i RLL nodule which had increased in size. Routine CBC and chemistry panel were unrevealing. Tuberculin skin testing was negative, as well as coccidioidal antibody serologies. Bronchoscopy revealed no evidence of endobronchial abnormalities.

CT guided needle biopsy of the RML subpleural nodule was nondiagnostic but was complicated by hemothorax and anemia due to acute blood loss. The patient required transfusion of 2 units PRBC, as well as chest tube thoracostomy. The patient underwent subsequent video assisted thoracoscopy performed with pleural decortication and removal of a large loculated hemothorax. Wedge resection of the right middle lobe and right lower lobe was performed. Pathological examination revealed well demarcated fibrotic nodules, which contained bland stromal and epithelial proliferation. Immunochemical stains were positive for estrogen receptor (ER) progesterone receptor (PR) CD 10, TTF-1, WT-1, CD 10, as well as desmin in stromal and glandular elements. CD 117 and CD34 were negative.

Findings were compatible with the diagnosis of pulmonary benign metastasizing leiomyoma (PBML).

Discussion

Overall, uterine leiomyomas are the most common neoplasm in women over 30 years of age with a prevalence of nearly 50 percent.¹ The vast majority of these are benign, and the frequency of malignant leiomyosarcomas are uncommon with relative frequency ranging from 0.13 to 6 percent.² Since the initial case reported by Steiner in 1939,³ more than 100 cases of PBML have been reported in the literature. Pulmonary benign metastatic leiomyoma is hormone dependent. Estrogen causes tumor proliferation while progesterone causes growth to subside.⁴

The pathogenesis of PBML is unknown.⁴ The most common theory is that the tumor is hematogenously spread. Surgical procedures including hysterectomy, myomectomy, and uterine curettage have been associated with development of this condition, presumably through the venous circulation.⁵ The mean time from hysterectomy to the findings of lung lesions is 15 years.⁶ This does not explain the rare discovery of simultaneous uterine leiomyoma and multiple pulmonary lesions.⁷

Due to the indolent nature of PBML, patients are frequently asymptomatic, and the majority of cases are discovered by routine chest radiographs. Patients may have complaints of dry cough, chest congestion, and, rarely, fever. Hemoptysis and chest pain are rare presentations and usually are an indication of multiple or large lesions.⁸

Plain radiographs or CT imaging most frequently demonstrates multiple well circumscribed without evidence of mediastinal or hilar adenopathy. Rarely a diffuse miliary pattern found. Metabolic imaging by FDG PET-CT scans are most often non avid, or with minimal uptake.⁹

Video assisted thoracoscopy is the preferred method of diagnosis. Fine needle aspiration biopsy is limited due to inadequate tissue sampling for histopathology and immunochemical examination.^{8,9}

Treatment options for PBML include surgical excision with close surveillance to monitor newly evolving lesions, although impractical for multiple bilateral lesions.⁹ Bilateral oophorectomy has been effective in halting progression of tumor size.¹⁰ Due to PBML having both estrogen and progesterone receptors, drug therapy can provide advantages over surgery, including lack of surgical morbidity, reversibility, and the possibility of promoting lesion regression. Progesterone treatment has been shown to cause regression in leiomyomatous tumors.¹¹ Newer agents such as raloxifene, a selective estrogen receptor modulator that exhibits estrogenic antagonistic effects on breast and uterine tissue, as well as anastrozole an aromatase inhibitor have demonstrated efficacy in lesion regression size. Luteinizing hormone releasing hormone agonists such as leuprolide and goserelin have also demonstrated activity against PBML.¹²

In summary, pulmonary benign metastasizing leiomyoma is a rare entity that should be considered in the differential diagnosis of multiple pulmonary nodules, particularly in women of late childbearing age or premenopausal with a history of prior uterine leiomyomas and hysterectomy. Diagnosis by video assisted thoracoscopy is recommended. Noninvasive and reversible hormonal receptor modulation is increasingly favored over surgical castration. Our patient is currently being followed by serial CT imaging with consideration for medical therapy if the lesions progress. Her cough continues to be managed with supportive measures including antitussives.

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