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Endobronchial Metastases of Gynecologic Leiomyosarcoma

A Case Report and Review of the Literature

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Abstract: Gynecologic leiomyosarcoma are uncommon malignancies. Although pulmonary metastases are frequent complications, endobronchial metastases are rare. We report 1 case of gynecologic leiomyosarcoma with endobronchial metastases, along with a review of the topic and bronchoscopic image of this uncommon phenomenon.

Key Words: leiomyosarcoma, pulmonary metastases, endobronchial

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CASE REPORT

A 46-year-old Philippino woman presented with 7 months of dyspnea, dry cough, night sweats, and 5-pound weight loss. She had 1 episode of hemoptysis 2 days before admission. In 2001, the patient had a hysterectomy and right oophorectomy for fibroids. In 2003, a left ovarian tumor was removed; the pathologic diagnosis was “benign leiomyoma.” She denied tobacco, alcohol, or drug use. She had never been sexually active. Her only out-patient medication was multivitamins.

Her initial vital signs were normal. Physical examination revealed diminished breath sounds in the right lower lung area and a palpable mass extending from the pelvis into the lower abdomen. Her chemistry and hematologic studies were normal.

Pelvic ultrasound showed a 0.5 × 13 × 12.3-cm left complex pelvic mass with both solid and liquid components. Chest x-ray revealed multiple nodules in both lungs, right middle and lower lung atelectasis, and a right pleural effusion; this was confirmed by computed tomography (Fig. 1). Interventional radiology-guided biopsy of the pelvic mass revealed malignant spindle cell neoplasm with myxoid stroma, atypical hyperchromatic nuclei, large atypical cells, and necrosis consistent with leiomyosarcoma (LMS). The specimen had the following immunohistochemical stain pattern: vimentin, myosin, desmin, CD117 were positive; keratin, S100, actin, and CD34 were negative.

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Bronchoscopy revealed a large, white, glistening endobronchial lesion obstructing the proximal bronchus intermedius (Fig. 2) with the same histology as the pelvic mass (Fig. 3). The patient’s hemoptysis and shortness of breath resolved spontaneously.

The patient underwent 3 cycles of gemcitabine and docetaxel for stage IV LMS; failing that, she received 7 cycles of ifosfamide. She died from postobstructive pneumonia 1 year after diagnosis.

DISCUSSION

LMSs are generally of uterine or retroperitoneal origin and commonly metastasize hematogenously to the lung.¹ However, the central bronchi are rarely affected. This case is particularly unusual in this respect.² Diagnosis of metastatic LMS may be unsuspected because there is often a long interval (up to 18y)³ between initial diagnosis and the clinical manifestation of metastases, and many of the primary tumors are initially misdiagnosed as simple or degenerating leiomyoma. This was most likely the situation in the case presented: the pathology of the 2003 left ovarian tumor was probably incorrectly interpreted as benign leiomyoma when it was malignant LMS. Until definitive pathology returned, the differential diagnosis included benign metastasizing



FIGURE 1. Enhanced computed tomography scan demonstrating multiple, diffuse metastatic pulmonary nodules, right middle and lower lobe atelectasis, and a right pleural effusion.



FIGURE 2. Bronchoscopic image showing large, white, glistening endobronchial lesion obstructing the proximal bronchus intermedius.

leiomyoma of the lung,⁴⁻⁶ an entity that is now considered low-grade LMS.⁷

Clinically important endobronchial metastases of solid tumors are rare. Although 30% of patients who die from malignancy have pulmonary metastases,⁸ one review of nearly 1400 consecutive autopsies revealed metastatic involvement of the major bronchi in only 2% of patients with solid tumors.⁹ However, microscopic, subclinical involvement occurs frequently (50% of patients with pulmonary metastases).¹⁰

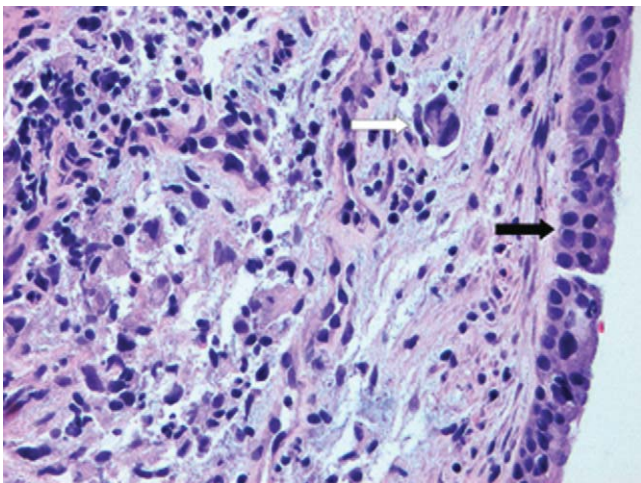


FIGURE 3. Biopsy of the right bronchus intermedius showing pleomorphic and atypical malignant spindle cells with bizarre, large atypical cells (white arrow) underneath benign bronchial epithelium (black arrow) (hematoxylin and eosin, 40 × 10, original).

The most common malignancies to metastasize to the bronchi are breast, colorectal, gastric, renal cell, and melanoma. Primary tumors almost always manifest locally before clinically apparent endobronchial metastases. However, renal cell cancer may be an exception. In the series of 1400 consecutive autopsies noted above, 7 of 15 cases of renal cell cancer presented with symptoms of endobronchial metastases between 3 weeks and 10 months before discovery of the renal tumor.⁹

The differential diagnosis of an endobronchial mass should include primary pulmonary LMS and fibrosarcoma. Although rare malignancies, these are often endobronchial when they do occur and are among the more common primary pulmonary sarcomas.^{11,12} Parenchymal metastases of any solid tumor, or primary pulmonary malignancies, may exert extrinsic pressure on the bronchi, mimicking a polypoid mass without involvement of the bronchial wall.¹

Not surprisingly, patients with endobronchial tumors frequently present with dyspnea, cough, and hemoptysis, as was the situation with the patient in the case presented. Other symptoms may include chest pain and arrhythmias from hypoxia. Radiographs will often reveal a luminal mass with postobstructive atelectasis or pneumonitis. The clinical and radiographic appearance of primary bronchogenic carcinoma and endobronchial metastases from distant organs may be indistinguishable.⁹ Therefore, diagnosis is confirmed with tissue sample. Rarely, patients may spontaneously expectorate tissue revealing spindle-shaped cells.^{13,14}

Long-term survival in patients with extraterine disease (stages III and IV) is rare. Initial management of uterine LMS with pulmonary metastases includes surgery, chemotherapy, and radiation therapy. Total hysterectomy/bilateral oophorectomy is followed by induction chemotherapy with a regimen that often includes doxorubicin alone or with agents such as methotrexate, cyclophosphamide, ifosfamide, or cisplatin. A case report of 1 patient who underwent an induction regimen of carboplatin and paclitaxel followed by maintenance oral etoposide demonstrated a stable pelvis and pulmonary tumor burden for at least 13 months.¹⁵

Surgical or endobronchial management of pulmonary metastases of LMS hinges on the distinction between true endobronchial metastases versus parenchymal metastases with gross endobronchial extension.¹ True endobronchial metastases are usually small and responsive to endobronchial laser or brachytherapy. In contrast, gross endobronchial extension of parenchymal metastases is usually bulky and the tumor frequently has invaded the distal parenchyma, rendering laser and brachytherapy ineffective.

A retrospective study of surgical resection of pulmonary metastases in 133 cases of uterine malignancies revealed a 38% 5-year survival rate (n = 11) for LMS with a median survival of approximately 20 months.¹⁶ In multivariate analysis, only disease-free interval (DFI) less than 12 months (defined as time from treatment of primary tumor to detection of metastases)

was predictive of poor survival postsurgery. Primary site (cervix vs. body of uterus), histology, age, number of metastases, and tumor size were not independently associated with risk of death. Among patients with a single metastasis and DFI < 12 months, 5-year survival rate approached 50%; therefore, short DFI should not preclude the option of surgery. Bilateral lung transplantation was successful in 1 case of benign metastasizing leiomyoma of the lung with no evidence of extrapulmonary or mediastinal metastases.⁷

Endobronchial lesions are often associated with mediastinal lymph node involvement. In the series of 133 patients noted above, 16 of 45 (36%) who underwent mediastinoscopy had positive lymph nodes. Patients taken for surgical resection should, therefore, have intraoperative mediastinal lymph node sampling, even in the absence of radiographic evidence of mediastinal lymphadenopathy.¹⁶

In summary, pulmonary metastasectomy may be appropriate for uterine malignancies in patients with DFI of 12 months or more who have good control of the primary tumor, no extrapulmonary metastases, and medical status and pulmonary function to withstand complete resection of metastases. Endobronchial laser or brachytherapy may be appropriate for smaller endobronchial metastases or in patients who are otherwise ineligible for metastasectomy.

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