Case Report

Endoscopic Treatment of Metastasis in the Main Bronchi From Sarcoma: A Report of 2 Cases

Carmen Montero,* Paz Valiño, Ana Souto, María del Mar Fernández, Juan Suárez, and Héctor Verea

Servicio de Neumología, Hospital Universitario, A Coruña, Spain

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ABSTRACT

Leiomyosarcoma is a cancer that can affect the soft tissues or organs. The standard treatment is complete tumor resection. Prognosis is difficult to predict and distant metastases can occur after a long disease-free period. Lung metastases are common but metastasis to the main bronchi with pulmonary atelectasis is very rare.

We describe 2 cases of pulmonary atelectasis and obstructive pneumonitis due to metastasis to the main bronchi from leiomyosarcoma of the uterus in one of the patients and leiomyosarcoma of the thigh in the other. Both patients were treated with endoscopic resection. We discuss the role of endoscopic laser treatment in the palliation of symptoms and as an initial procedure before other cancer treatments are started.

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Tratamiento endoscópico de Metástasis en Bronquios Principales de Sarcoma: Aportación de 2 Casos

RESUMEN

El leiomiomasarcoma es un tumor que puede localizarse en tejidos blandos o en órganos. El tratamiento consiste en la resección quirúrgica completa del tumor. El pronóstico es difícil de predecir y después de un largo periodo sin enfermedad pueden aparecer metástasis a distancia. Las metástasis pulmonares son frecuentes, pero su localización en bronquios principales con atelectasia pulmonar es muy rara.

Presentamos 2 casos de atelectasia pulmonar y neumonitis obstructiva secundaria a metástasis en bronquios principales de un leiomiomasarcoma de útero y de un leiomiomasarcoma de muslo, respectivamente, que tratamos con resección endoscópica. Analizamos el papel del tratamiento endoscópico con láser para paliar los síntomas y como ayuda previa a la realización de otro tipo de tratamiento oncológico.

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Introduction

Soft tissue sarcomas are very rare. The estimated annual incidence is 38 cases per million inhabitants1 and the tumors most often affect the extremities, the trunk, and the viscera. In women, leiomyosarcoma occurs in the uterus in 40% of cases2 and accounts for approximately 4% to 9% of all invasive uterine tumors.3 Lung metastasis is common in sarcoma4 but metastases to the main bronchi causing obstructive pneumonitis and pulmonary atelectasis are very rare.5,6 Furthermore, there is little experience with interventional bronchoscopy in the treatment of these lesions. Endoscopic treatment has proven to be effective in improving quality of life and survival in patients with endobronchial metastases from other types of solid tumors.7

We describe the case of 2 patients who underwent surgery for leiomyosarcoma, of the uterus in one case and of the thigh in the other. Both patients had a single metastatic lung lesion with extensive endobronchial growth that had appeared following a long disease-
free period, and in both cases, endoscopic treatment was successfully used to resolve atelectasis and respiratory failure. We also analyze the role of endoscopy in treating metastases of this type.

**Case Descriptions**

**Patient 1**

The first patient was a 58-year-old woman with no history of smoking or substance abuse. In 1999, she had undergone a total hysterectomy and bilateral adnexectomy and was diagnosed with a submucosal leiomyosarcoma following evaluation of the resulting surgical specimen. The rest of the uterus and the adnexa were found to be normal and no metastases were detected. The patient completed 6 cycles of adjuvant treatment with cisplatin and doxorubicin.

In March 2005, the patient consulted for cough and dyspnea, and a chest radiograph showed a mass measuring 6 cm in diameter in the left hemithorax. The laboratory workup was normal. A computed tomography (CT) scan of the chest, abdomen, and pelvis showed a single 6-cm mass in the left hemithorax that was in contact with the mediastinum. Bronchoscopic findings were unremarkable and there were no signs of tumor recurrence in the gynecologic examination.

The mass was resected by thoracotomy and following confirmation of invasion into the mediastinal pleura by a leiomyosarcoma, the patient was initiated on ifosfamide. In March 2007, the patient presented dyspnea, cough, and fever, as well as a loss of volume in the left lung evidenced by a chest radiograph. A CT scan of the chest and abdomen revealed pneumonia and a mass in the left hilum occluding the left main bronchus, and a bronchoscopy revealed a polypoid tumor that was almost completely occluding the left main bronchus (Figure 1). A diagnosis of leiomyosarcoma was confirmed by bronchial biopsy. Antibiotic treatment with ceftazidime and amikacin was initiated and, following photocoagulation with a diode laser (Diomed), the tumor was resected during rigid bronchoscopy. On completion of this treatment, the tumor was seen to be located in the posterior wall of the left upper lobe bronchus, but the left upper lobe segments and the left lower lobe bronchi were macroscopically unremarkable (Figure 2). The chest radiograph showed great improvement, and the CT scan showed just a small residual lesion, measuring 7 mm in diameter, in the left upper lobe. The patient was completely asymptomatic when she was discharged. Although she remained free of symptoms at 2 years follow-up, a new tumor was detected in the area from which the mediastinal mass had been removed. The tumor was diagnosed as a local recurrence. There were, however, no respiratory symptoms or CT abnormalities suggestive of bronchial growth. The treatment prescribed was radiotherapy.

**Patient 2**

The second case we describe concerns a 71-year-old woman who presented with a 1-week history of cough, pain in the left hemithorax, and dyspnea that had appeared 48 hours earlier. The patient had never smoked. Ten years earlier, she had undergone surgery for a leiomyosarcoma of the thigh. The tumor, which measured 8 cm in diameter, had been resected with negative margins, and no evidence of disease had been detected in any of the follow-up visits. The patient presented a good general state of health during the physical examination, and had a temperature of 37.5 °C and a blood pressure of 140/90 mm Hg. The only notable finding in the physical examination was the total absence of vesicular murmur in the left hemithorax. The laboratory workup was normal. Arterial blood gas measurements taken in ambient air revealed a PaO$_2$ of 57 mmHg, a PaCO$_2$ of 35 mmHg, and a pH of 7.44. The chest radiograph showed complete left lung atelectasis (Figure 3) and the CT scan revealed a 5-cm mass in the left upper lobe extending into the left main bronchus and causing pulmonary collapse. There was no evidence of mediastinal lymph node involvement or metastases in the abdomen or pelvis. Bronchoscopy revealed a polypoid tumor in the distal portion of the left main bronchus, which was totally occluded. Finally, leiomyosarcoma was confirmed by bronchial biopsy. The fever subsided on initiation of antibiotic treatment with intravenous...
amoxicillin-clavulanic acid, but the atelectasis and respiratory failure persisted. Photocoagulation with a diode laser was performed and the mass was removed during bronchoscopy. This procedure confirmed that the tumor had originated from the left upper lobe and the left main bronchus and that the left lower lobe was free of disease. Following the operation, the patient developed a left pneumothorax, which was resolved with the placement of a chest drain. The follow-up chest radiograph (Figure 4) showed a mass in the left upper lobe.

Discussion

The treatment of choice for leiomyosarcoma is complete tumor resection. Even when this is achieved, however, distant metastases can occur after a long disease-free period. The lung is a common site for metastasis. In a review of 73 autopsies of patients who had died of leiomyosarcoma of the uterus, Rose et al. found that the tumors underwent hematogenous spread and that the lung was a common site for metastasis. In view of the limited success of chemotherapy in the treatment of metastases from uterine tumors, surgical resection has emerged as an alternative. One study of 133 patients with metastases from malignant uterine tumors, including 11 cases of leiomyosarcoma, showed a 5-year-survival rate of 37%. In the same study, the only predictor of good prognosis identified by multivariate analysis was a disease-free interval of over 12 months. Other studies that have analyzed the resection of metastases from solid tumors have also concluded that disease-free progression and complete surgical resection are the most important prognostic factors.

Metastases to the main bronchi are very uncommon. In a review of 1400 autopsies, pulmonary metastases were observed in 30% of cases, but the main bronchi were involved in just 2%,1,2 Some metastases to the main bronchi display a polypoid growth pattern, with extensive occlusion of the bronchial lumen but no infiltration of the bronchial wall.3-5 Because the surgical resection of these lesions requires the removal of extensive amounts of lung tissue, some authors have proposed the use of rigid bronchoscopy to accurately evaluate the extent of bronchial tumor invasion and thus avoid the removal of excessive lung tissue. The use of bronchotomy in such cases has also been reported. Another option is endoscopic resection.6,7 While there are no specific data on the benefits of this treatment in sarcoma metastases, its use in other types of tumors to clear the bronchi has been seen to improve quality of life and survival.8

The experience with our 2 patients shows that interventional bronchoscopy plays an important role in the evaluation of the extension of metastatic endobronchial tumors prior to surgical resection and that it can also help to resolve local complications and alleviate symptoms when resection is not indicated. This treatment should be considered in the management of these type of tumors.

References


