Arch Bronconeumol. 2010;**46(1)**:40-43

ARCHIVOS DE BRONCONEUMOLOGIA

www.archbronconeumol.org

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

ARTICLE INFO

Article history:
Received March 25, 2009
Accepted April 2, 2009
Available online June 21, 2009

Keywords:
Leiomyosarcoma
Sarcoma
Bronchial metastases
Endoscopic treatment

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.

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

Palabras clave:
Leiomiosarcoma
Sarcoma
Metástasis bronquiales
Tratamiento endoscópico

RESUMEN

El leiomiosarcoma 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 período 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 leiomiosarcoma de útero y de un leiomiosarcoma 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.

Introduction

Soft tissue sarcomas are very rare. The estimated annual incidence is 38 cases per million inhabitants[1] and the tumors most often affect the extremities, the trunk, and the viscera. In women, leiomyosarcoma occurs in the uterus in 40% of cases[2] and accounts for approximately 4% to 9% of all invasive uterine tumors.[3] Lung metastasis is common in sarcoma[4] 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-

*Corresponding author.
E-mail address: carmen.montero.martinez@sergas.es (C. Montero).

0300-2896/$ - see front matter

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 bronchus 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.

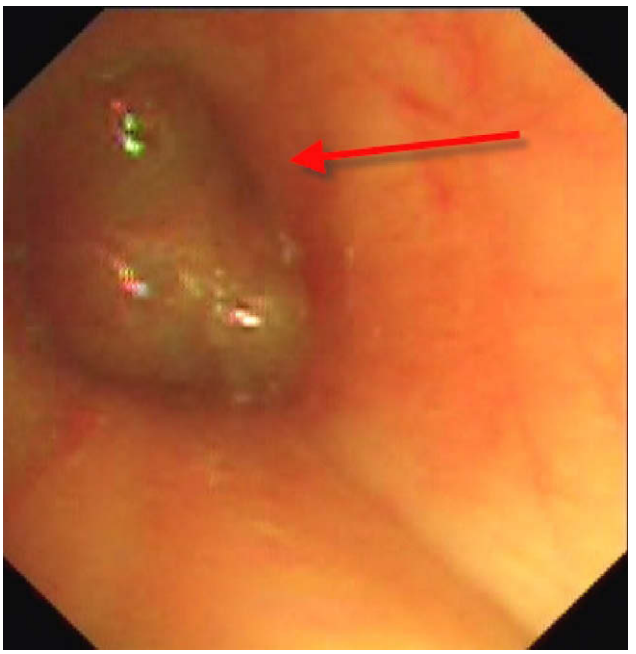

Figure 1. Polypoid tumor occluding the left main bronchus.

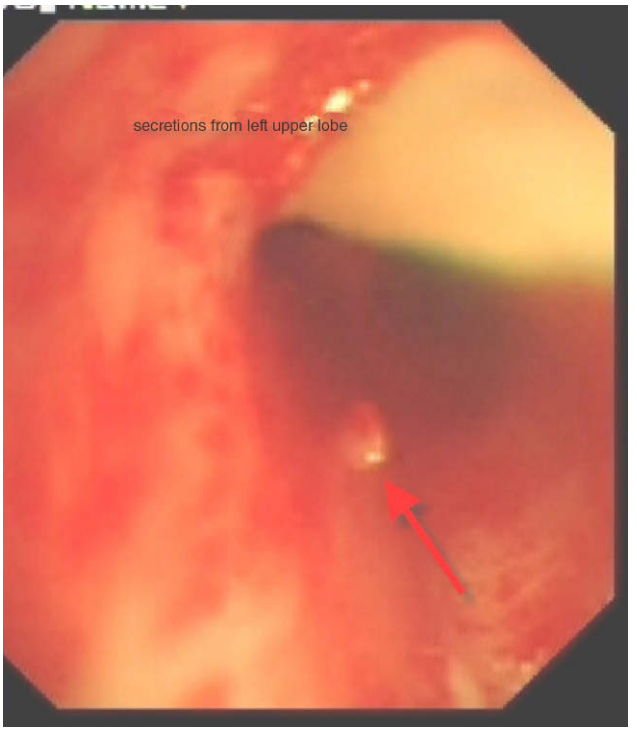

Figure 2. Residual tumor lesion on the wall of the left upper lobe bronchus following resection. Note the permeable segments of the left upper lobe involved in draining retained secretions.

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 mm Hg, a $PaCO_2$ of 35 mm Hg, 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

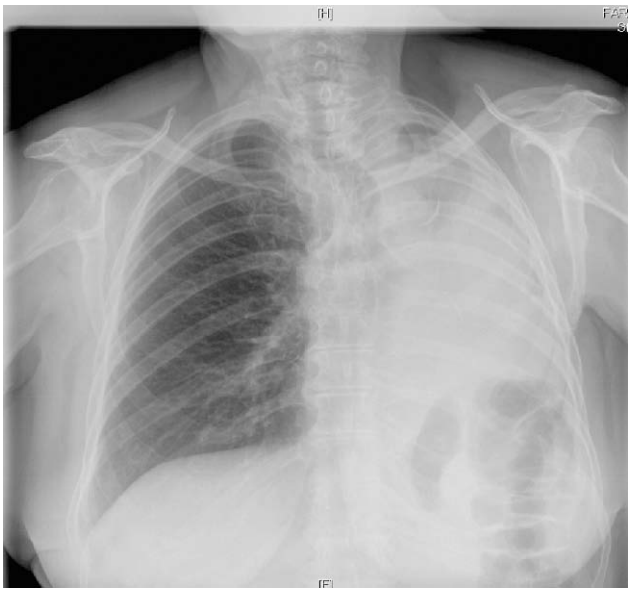

Figure 3. Complete pulmonary atelectasis secondary to endobronchial metastasis.

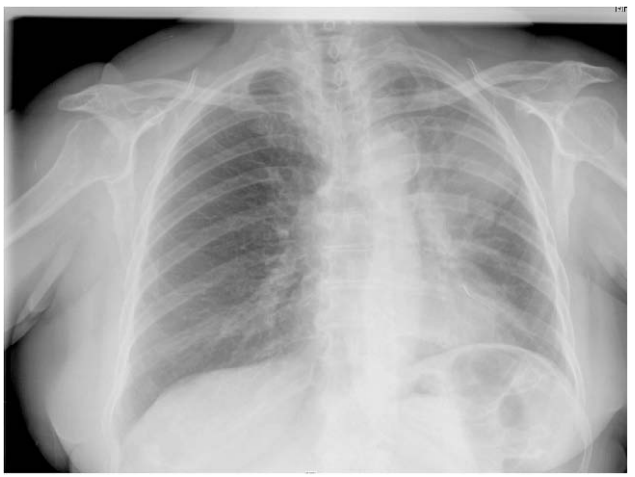

Figure 4. Resolution of atelectasis following endoscopic treatment. Note the mass in the left upper lobe.

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 and resolution of the left pulmonary atelectasis. Postoperative arterial blood gas values included a PaO_2 of 76 mm Hg, a $PaCO_2$ of 39 mm Hg, and a pH of 7.41. The patient was asymptomatic on discharge and completion of staging evaluation using positron emission tomography and brain magnetic resonance imaging revealed no other metastases. A left pneumonectomy was performed subsequently, and it was confirmed that the mediastinal lymph nodes and the bronchial resection margins were free of tumor. The patient is being monitored by our hospital's oncology department and has been found to be free of disease after 6 months of follow-up.

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[4] 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,[8] 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%.[9] 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.[10-12]

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%.[13,14] 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.[13-15] 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.[16] 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.[7]

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

1. Gutiérrez JC, Pérez EA, Franceschi D, Moffat FL Jr, Livingstone AS, Koniaris LG. Outcomes for soft-tissue sarcoma in 8249 cases from a large state cancer registry. J Surg Res. 2007;141:105-14.
2. Toro JR, Travis LB, Wu HJ, Zhu K, Fletcher CD, Devesa SS. Incidence patterns of soft tissue sarcomas, regardless of primary site in the surveillance, epidemiology and end results program 1978-2001: an analysis of 26,758 cases. Int J Cancer. 2006;119:2922-30.
3. Brooks SE, Zhan M, Cote T, Baquet CR. Surveillance, epidemiology, and end results analysis of 2677 cases of uterine sarcoma 1989-1999. Gynecol Oncol. 2004;93:204.
4. Rose PG, Piver MS, Tsukada Y, Lau T. Patterns of metastasis in uterine sarcoma. An autopsy study. Cancer. 1989;63:935-8.
5. Braman SS, Whitcomb ME. Endobronchial metastases. Arch Intern Med. 1975;135:543-7.
6. Gerst PH, Levy J, Swaminathan K, Kshettry V, Albu E. Metastatic leiomyosarcoma of the uterus: unusual presentation of a case with late endobronchial and small bowel metastases. Gynecol Oncol. 1993;49:271-5.
7. Fournel C, Bertletti L, Nguyen B, Vergnon JM. Endobronchial metastases from colorectal cancer: natural history and role of interventional bronchoscopy. Respiration. 2009;77:63-9.
8. Leitao MM, Brennan MF, Hensley M, Sonoda Y, Hummer A, Bhaskaran D, et al. Surgical resection of pulmonary and extrapulmonary recurrences of uterine leiomyosarcoma. Gynecol Oncol. 2002;87:287-94.
9. Anraku M, Yokoi K, Nakagawa K, Fujisawa T, Nakajima J, Akiyama H, et al; Metastatic Lung Tumor Study Group of Japan. Pulmonary metastases from uterine malignancies: results of surgical resection in 133 patients. J Thorac Cardiovasc Surg. 2004;127:1107-12.
10. Anderson TM, McMahon JJ, Nwogu CE, Pombo MW, Urschel JD, Driscoll DL, et al. Pulmonary resection in metastatic uterine and cervical malignancies. Gynecol Oncol. 2001;83:472-6.

11. Pagés Navarrete C, Ruiz Zafra J, Simón Adiego C, Díez Piña JM, Cueto Ladrón de Guevara A, Sánchez-Palencia Ramos A. Surgical treatment of pulmonary metastasis: survival study. Arch Bronconeumol. 2000;36:569-73.
12. Ley Tao MM, Bregan MF, Hensley M, Sonoda Y, Hummer A, Bhaskaran D, et al. Surgical resection of pulmonary and extrapulmonary recurrences of uterine leiomyosarcoma. Gynecol Oncol. 2002;87:287-94.
13. Giudice JC, Komansky H, Gordon R. Endobronchial metastasis of uterine leiomyosarcoma. JAMA. 1979;41:1684.
14. Flynn KJ, Kim HS. Endobronchial metastasis of uterine leiomyosarcoma. JAMA. 1978;240:2080.
15. Warren WH, Bleck P, Kittle CF, Faber LP. Surgical management of pulmonary metastatic leiomyosarcoma with gross endobronchial extension. Ann Thorac Surg. 1990;50:739-42.
16. Richman M, Au J, Aoyama CH, Kamangar N. Endobronchial metastases of gynecologic leiomyosarcoma. A case report and review of the literatura. J Bronchol. 2007;14:131-3.