Benign metastasizing leiomyoma in the lung: a case report

R. Sabatini, *M.D.*; **R. Ferreri,** *M.D.*; **G. Distante,** *M.D.*; **V. Loizzi,** *M.D.*; **P. Loizzi,** *M.D.*

Division of Obstetrics and Gynecology "B", University of Bari (Italy)

Summary

The authors report a rare case of leiomyomatosis of the lung diagnosed in a women with uterine leiomyomatosis not previously treated.

The absence of mitosis with nuclear atypism in all histological samples opens the question of whether the origin of the tumor in the lung was from uterine leiomyomas metastasizing or multifocal amarthomas, synchronous or metachronous. Hamartomas or real metastasis of uterine leiomyomas? The response to GnRH analogue treatment was evaluated.

Key words: Uterine leiomyoma; GnRH agonist therapy; Lung metastasis.

Introduction

Leiomyoma of the lung is a very rare benign tumor with approximately 80 well documented cases in the literature [1, 2, 3].

A recent study carried out by Sekine reports that leiomyoma represents 0.085% of pulmonary benign tumors [4]. Multiple pulmonary leiomyomas represent an exceptional condition that occurs in sexually mature women.

It remains unclear whether this evolves from a low grade "malignant uterine leiomyoma" or from proliferation of multifocal, synchronous or metachronous, but autochronous cellular foci [5, 6, 7]. The occurrence of sex hormone receptors in myomas of the lung suggests the influence of the bilateral ovariectomy and/or sex hormones [8, 9, 10, 11]. The reponse of the disease to gonadotropin-releasing hormone (GnRH) treatment is reported.

Case Report

The authors report a rare case of uterine leiomyomatosis metastasizing in the lung.

A 45-years-old woman was admitted to the III Clinic of Obstetrics and Gynecology, University of Bari, for uterine leiomyomatosis and significant haemorrhage. The past medical history revealed that she had undergone right nefrectomy at the age of 35 for pyoneprhrosis.

Moreover, the patient had undergone "in vitro" fertilization three times in the previous ten years. The patient had presented a chronic obstructive airway disease for five years.

Routine chest X-rays revealed bilateral pulmonary multiple nodules and cystic lesions similar to bronchiectasis (Figure 1).

The HRTC detected multiple round lesions ranging from 5 to 10 mm and diffuse bronchiectasis (Figure 2). This clinical picture could be compatible with pulmonary metastasis. The pulmonary function tests revealed a serious obstructive ventilation deficit.

Due to her critical respiratory condition curettage was possible only under spinal anaesthesia. Histopathologic examination demonstrated a follicular endometrium and necrotic haemorrhagic aspects. The patient was transferred to the Pneumology Unit where she was submitted to right thoracoscopic biopsies. The histological finding revealed benign leiomyomas, without any evidence of malignancy (Figure 3).

Revised manuscript accepted for publication February 20, 2002

Leiomyomas showed a reactivity against actin and desmin. Estrogen and progesterone receptors were present.

The multifocal pulmonary localizations and severe condition of the patient prevented any surgical approach. Waiting for lung transplantation, the pneumologists consulted the gynaecologists for possible alternative therapy.

Because the histologic report showed a high concentration of estrogen receptors, an analogue gonadotropin-releasing hormone (GnRH) was given.

Six months later an evident decrease in size of the nodular lesions in both lung fields and uterus was observed. Therefore, the different clinical condition of the patient permitted a hysterectomy with bilateral salpingo-oophorectomy. No metastatic disease was identified at laparotomy. Both adnexa were normal.

Pathologically all samples revealed that the lesions consisted of interlacing bundles of smooth muscle cells and connective tissue without evidence of malignancy.

The histopathological characteristics were remarkably similar to those of the previously resectioned lung leiomyomas, without high cellularity and mitosis with nuclear atypism.

Today, five years after surgery, the patient is in good condition especially for respiratory compliance, and she does not show any evidence of tumor localization. Only a discrete dyspnoea is present, caused by chronic obstructive airway disease.

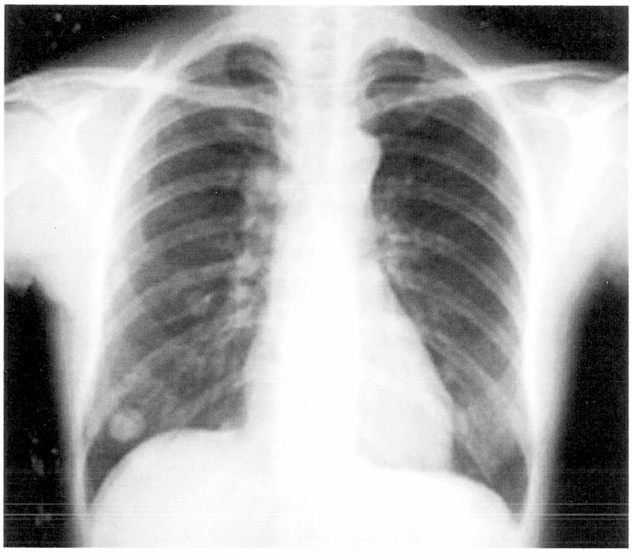
Discussion

There are rare cases of histologically benign-appearing uterine leiomyomas with subsequent development of multifocal smooth muscle tumors, very often located in the lung.

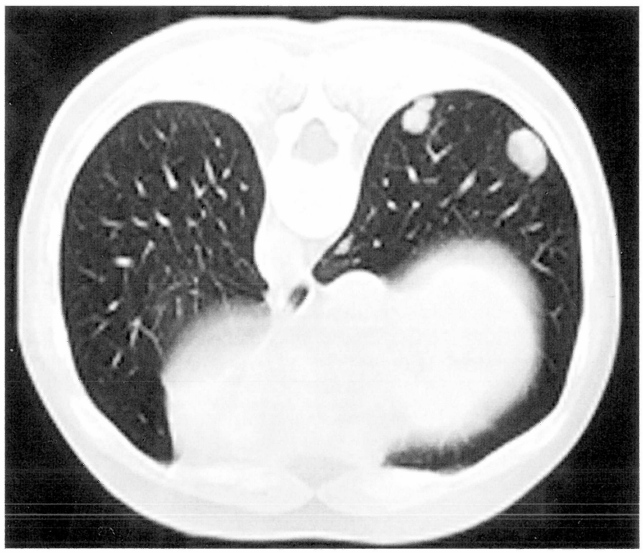
Frequently lung localization appears several years after myomectomy or hysterectomy with gonadal preservation for uterine leiomyomas.

Multifocal synchronous or metachronous origin is another possible explanation. In our case the histological diagnosis of the leiomyomas was primarily pulmonary and subsequently uterine. However, no evidence of atypical mitosis was detected in any histological samples of the lung or the uterus. The true nature of metastasizing leiomyomas is uncertain.

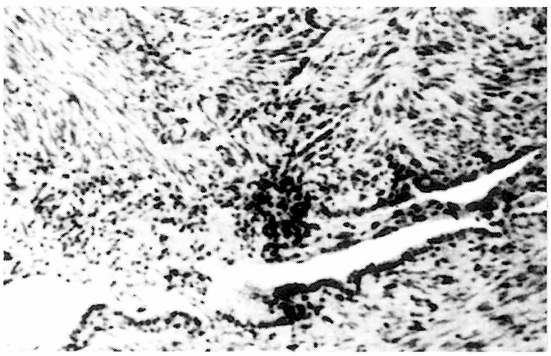
Our results open the question of whether the origin of the tumor in the lung was due to real uterine benign metastasizing leiomyoma or multifocal hamarthomas (synchronous or metachronous). Thus, the cases reported in the literature concerning the metastasis of leiomyosarcoma



1



2



3

Figure 1. — X-ray showing several subpleural and intraparenchymal nodules which varied in size from 5 to 10 mm in diameter.

Figure 2. — CT confirms the presence of multiple bilateral pulmonary nodules.

Figure 3. — Histological finding consists of recognizable smooth muscle cells arranged in intersecting bundles without any evidence of malignancy.

may not be considered in benign metastasizing leiomyomas [12, 13, 14, 15].

Measuring estrogen and progesterone receptors in lung biopsy material may help to determine the most appropriate therapy.

Infact, the GnRH analogue treatment permitted a decrease in leiomyomas of the lung before the ovariectomy.

References

[1] Nishida T., Inove K., Yamamoto R., Takehara S., Kaji M.: "Case of benign metastasizing leiomyoma of the lung". *Nippon - Kyobu - Geka - Gakkai - Zasshi*, 1996, *44 (2)*, 182.

[2] Jacobson T. Z., Rainey E. J., Turton C.W.: " Pulmonary benign metastasizing leiomyoma: response to treatment with goserelin". *Thorax*, 1995, *50 (11)*, 1225.

[3] Abramson S., Gilkeson R. C., Goldstein J. D.: "Benign metastasizing leiomyoma: clinical, imaging, and pathologic correlation". *Am. J. Roentgenol.*, 2001, *176*, 1409.

[4] Sekine I., Kodama T., Yokose T., Nishiwaki Y., Suzuki K., Goto K. *et al.*: "Rare pulmonary tumors: a review of 32 cases". *Oncology*, 1998, *55 (5)*, 431.

[5] Tietze L., Gunther K., Merkelbach-Bruse S., Handt S.: "Benign metastasizing leiomyoma? A case report without evidence for unibalanced cytogenetic aberrations". *Verh - Dtsch - Ges - Pathol.*, 1998, *82*, 332.

[6] Gabka C. J., Muller C., Baretton G., Di Enemann H., Schilderg F. W.: "Recurrent multiple leiomyomatosis hamartomas of the lung". *Chirurg.*, 1995, *66 (5)*, 530.

[7] Cho K. R., Woodruff D., Epstein J. I.: "Leiomyoma of the uterus with multiple extrauterine smooth-muscle tumors. A case report suggesting multifocal origin". *Hum. Pathol.*, 1989, *20 (1)*, 80.

[8] Nakanishi S., Nakano K., Hiramoto T.: "So-called benign metastasizing leiomyoma of the lung presenting with bone metastasis". *Nihon - Kokyuki - Gakkai - Zasshi*, 1999, *37 (2)*, 146.

[9] Montegi M., Takayanagi N., Sando Y., Ubukata M., Imai S., Suzuki T.: "A case of so-called benign metastasizing leiomyoma responsive to progesterone". *Nippon - Kyobu - Shikkau - Gakkai - Zasshi*, 1993, *31 (7)*, 890.

[10] Abu-Rustum N. R., Curtin J. P., Burt M.: "Regression of uterine low grade smooth-muscle tumors metastatic to the lung after oophorectomy". *Obstet. Gynecol.*, 1997, *89*, 850.

[11] Pawlik C., Wildberege J. E., Tietze L.: "Benign metastasizing leiomyoma of the lung. A rare differential diagnosis of pulmonary space-occupying lesions". *Dtsch. Med. Wochenschr.*, 2001, 11, *126 (19)*, 551.

[12] Shneider T., Kugler C., Kayser K., Dienemann H.: "Benign, pulmonary metastatic leiomyoma of the uterus". *Chirurg.*, 2001, *72 (3)*, 308.

[13] Ohno K., Kuwata K., Hashimoto J., Yamamoto S., Noshida Y., Sagaguchi T.: "A case of multiple pulmonary leiomyomatous hamartoma". *Nippon Kyobu Shikkan Geka Gakkai Zasshi*, 1996, *44 (5)*, 723.

[14] Yoshitomi A., Sato A., Imokawa S., Suganuma H., Tamura R.: "A case of so-called benign metastasizing leiomyoma". *Nihon Kyobu Shikkan Gakkai Zasshi*, 1994, *32 (4)*, 373.

[15] Koh D. M., Burn P. R., King D. M.: "Benign metastasizing leiomyoma with intracaval leiomyomatosis". *Brit. J. Radiol.*, 2000, *73*, 435

Address reprint requests to:
P. LOIZZI, M.D.
Direttore Dip. Scienze Chirurgiche Generali
Sezione di Ginecologia e Ostetricia "B"
Univ. Studi di Bari - Piazzo G. Cesare, 11
70124 Bari (Italy)