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Case Report

Pulmonary benign metastasizing leiomyoma: a case report

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Abstract: Pulmonary benign metastasizing leiomyoma (PBML) is a rare entity usually occurring in females with history of uterine leiomyoma, and it is preferential to metastasize to the lung and appears as a histopathologic benign tumor of smooth muscle origin. In this article, the clinical and pathological data from 1 patient with PBML were analyzed. Chest CT scan showed that multiple well-defined nodules in the both lobes of the lungs. The tumor cells in the lung were well differentiated, and the pattern of tumor was similar to the original tumor. IHC identified it originated from smooth muscle cells, consistent with the diagnosis of PBML. Positive staining of estrogen and progestogen receptors was detected in both the leiomyoma and the metastasizing lesions. During two years of observation, pulmonary function parameters were within normal limits and there was no evidence of tumor recurrence.

Keywords: Benign metastasizing leiomyoma, leiomyoma of uterus, lung, metastasis

Introduction

Uterine leiomyoma is the most common benign gynecological tumor in women of reproductive age. Very rarely, it shows unusual growth patterns with extrauterine smooth muscle nodules named benign metastasizing leiomyoma (BML). It has been described as originating from the metastasis of the histologically benign uterine leiomyoma [1, 2]. No more than 100 cases were reported in the literature, and the first one was reported as fibroleiomyomatous hamartoma by Steiner in 1939.

Lung is known to be the most common metastatic site [3, 4]. In addition to lung, the extrauterine sites that these tumors can localize to include skin, pelvis, abdomen, muscle, greater omentum, inferior vena cava, right atrium, brain and bones [5, 6]. Pulmonary metastases are usually asymptomatic and detected by routine chest radiological examinations, years after hysterectomy. Some patients have symptoms such as cough, pain, or dyspnoea but they are frequently connected with underlying conditions such as pneumonia or bronchitis [7, 8]. However, the risk factors, related to the etiology and the clinical behaviors of BML have not been fully identified.

Case report

A 48-year-old woman, who was a nonmoker, was found to have an abnormal shadow on a chest X-ray during a regular health checkup. The patient had no history of neoplasm except for myomectomy for uterine leiomyoma 8 years ago. This study was performed in accordance with the ethical guidelines of the Tongji University School of Medicine.

The clinical examination and laboratory findings were normal. CT scan showed multiple small, well-circumscribed, noncalcified nodules in both lungs (**Figure 1**).

Postoperative histopathological examination that revealed a solid nodule was sharply demarcated from the adjacent lung parenchyma and no pre-existing pulmonary architecture was recognizable in the nodule. Plump spindle-shaped smooth muscle cells with elongated nuclei and eosinophilic cytoplasm formed anastomosing fascicles (**Figure 2A**). A comparison of the pulmonary findings with the pathology of previous leiomyoma was performed. The uterine leiomyoma, well circumscribed, with high cellularity, without nuclear polymorphism, without necrosis (**Figure 2B**).

Figure 1. Chest CT findings. Chest CT showed multiple, round, and well-circumscribed nodules in both lung.

For immunohistochemical staining, tumors were fixed in 4% paraformaldehyde. Tumor was determined by ER and PR staining. Based on immunohistochemical analysis, tumor cells were positive for estrogen and progesterone receptors, compatible with a tumor arising from the uterine smooth muscle (**Figure 3**).

During two years of follow-up, pulmonary function parameters were within normal limits and there was no evidence of tumor recurrence.

Discussion

BML is the term used to describe multiple benign leiomyomatous lesions occurring in patients with a previous history of uterine leiomyoma. The main metastatic site of benign metastasizing leiomyoma is the lung. Most patients with benign metastasizing leiomyoma are asymptomatic and are usually found during routine physical examinations. It has been reported that the average age of patients with benign metastasizing leiomyoma is 48 years old and pulmonary changes can be seen from 3 months to 26 years after hysterectomy [9].

Radiographically, the lung lesions tend to be well-circumscribed, multiple bilateral nodules that range from several millimeters to centimeters and are not typically calcified. The radiological presentation of pulmonary benign metastasizing leiomyoma is as multiple nodules in 87% of cases (70% bilateral nodules and 17% unilateral nodules) or as a solitary nodule in 13% of cases [10]. The patient in our study demonstrated the most common pattern of BML, disseminated spread in the lungs.

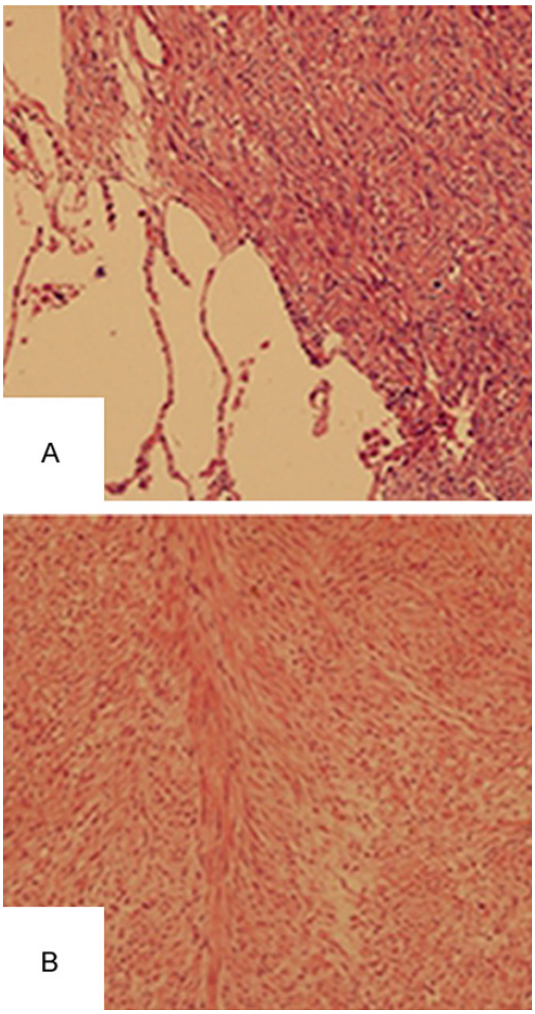

Figure 2. Microscopic changes of pulmonary benign metastasizing leiomyoma: A. Histologically, spindle-shaped smooth muscle cells without nuclear pleomorphism were arranged in anastomosing fascicles. B. The uterine leiomyoma, well circumscribed, with high cellularity, without nuclear polymorphism, without necrosis. HE, ×200.

Several hypotheses have been proposed regarding the etiology of BML: (1) hematogenous spread of a benign uterine tumor, (2) a low-grade leiomyosarcoma metastasizing to the lung; or (3) multifocal proliferations of smooth muscle cells occurring throughout the body in response to hormonal stimulation. The findings of recent cytogenetic studies are consistent with a monoclonal origin of both uterine and pulmonary tumors [11, 12]. Hormone receptor positivity in extrauterine lesions, the response to hormone treatment, and the exclusive occurrence in women also support this hypothesis [13]. Our study also tested that pul-

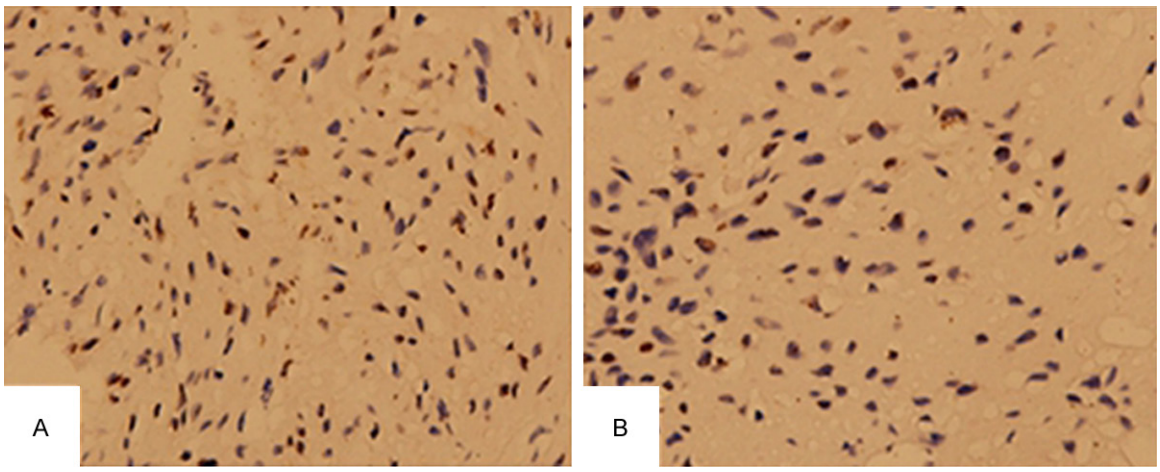

Figure 3. Immunohistochemistry of the lung nodule. The nuclei stained positive for estrogen and progesterone receptors: A. Immunostaining for estrogen receptor; B. Immunostaining for progesterone receptor. IHC, ×400.

monary tumors may origin from uterine leiomyomas by immunohistochemistry of estrogen and progesterone receptors.

At present, surgical resection is a primary treatment. If lesions are not resectable due to some reasons, hormone treatment should be considered instead. BML mainly affects sexually mature women when the hormonal effects are at a maximal level, and usually regresses after menopause or parturition. Tumors were also shown to be sensitive to treatment with progestin, goserelin, ovarian ablation, and oophorectomy [14, 15].

In conclusion, BML is a borderline tumor with benign histological features, in spite of its biological behavior suggesting malignancy. Although it is a rare condition, it should be considered in asymptomatic women of reproductive age with a history of uterine leiomyoma, who present with solitary or multiple pulmonary nodules.

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Disclosure of conflict of interest

None.

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References

[1] Awonuga AO, Rotas M, Imudia AN, Choi C, Khulpateea N. Recurrent benign metastasizing leiomyoma after hysterectomy and bilateral salpingo-oophorectomy. Arch Gynecol Obstet 2008; 278: 373-376.

[2] Vaquero ME, Magrina JF, Leslie KO. Uterine smooth-muscle tumors with unusual growth patterns. J Minim Invasive Gynecol 2009; 16: 263-268.

[3] Jeon HW, Choi SH, Sung SW, Park JK. Pulmonary benign metastasizing leiomyoma: report of three cases. World J Surg Oncol 2013; 11: 281-284.

[4] Cobellis L, Castaldi MA, Mosca L, Frega V, Ambrosio D, Corvino F, Colacurci N. Benign pulmonary metastasizing leiomyomatosis: case report. Eur J Gynaecol Oncol 2014; 35: 195-198.

[5] Moon H, Park SJ, Lee HB, Kim SR, Choe YH, Chung MJ, Jin GY, Lee YC. Pulmonary benign metastasizing leiomyoma in a postmenopausal woman. Am J Med Sci 2009; 338: 72-74.

[6] Yoon G, Kim TJ, Sung CO, Choi CH, Lee JW, Lee JH, Bae DS, Kim BG. Benign metastasizing leiomyoma with multiple lymph node metastasis: a case report. Cancer Res Treat 2011; 43: 131-133.

[7] Säynäjäkängas O, Maiche AG, Liakka KA. Multiple progressive pulmonary leiomyomatous metastases treated with tamoxifen-a case report with a review of the literature. Acta Oncol 2004; 43: 113-114.

[8] Goyle KK, Moore DF, Garett C, Goyle V. Benign metastasizing leiomyomatosis: case report and review. Am J Clin Oncol 2003; 26: 473-476.
[9] Abramson S, Gilkeson RC, Goldstein JD, Woodard PK, Eisenberg R, Abramson N. Benign metastasizing leiomyoma: clinical, imaging, and pathologic correlation. Am J Roentgenol 2001; 176: 1409-1413.
[10] Horstmann JP, Pietra GG, Harman JA, Cole NG, Grinspan S. Spontaneous regression of pulmonary leiomyomas during pregnancy. Cancer 1977; 39: 314-321.
[11] Tietze L, Günther K, Hörbe A, Pawlik C, Klosterhalfen B, Handt S, Merkelbach-Bruse S. Benign metastasizing leiomyoma: a cytogenetically balanced but clonal disease. Hum Pathol 2000; 31: 126-128.
[12] Lee HJ, Choi J, Kim KR. Pulmonary benign metastasizing leiomyoma associated with intravenous leiomyomatosis of the uterus: clinical behavior and genomic changes supporting a transportation theory. Int J Gynecol Pathol 2008; 27: 340-345.
[13] Kayser K, Zink S, Schneider T, Dienemann H, André S, Kaltner H, Schüring MP, Zick Y, Gabius HJ. Benign metastasizing leiomyoma of the uterus: documentation of clinical, immunohistochemical and lectin-histochemical data of ten cases. Virchows Arch 2000; 437: 284-292.
[14] Arif S, Ganesan R, Spooner D. Intravascular leiomyomatosis and benign metastasizing leiomyoma: an unusual case. Int J Gynecol Cancer 2006; 16: 1448-1450.
[15] Egberts JH, Schafmayer C, Bauerschlag DO, Jänig U, Tepel J. Benign abdominal and pulmonary metastasizing leiomyoma of the uterus. Arch Gynecol Obstet 2006; 274: 319-322.