A benign pulmonary metastasizing leiomyoma is a recognized clinical entity that has been infrequently reported in the medical literature. We report two cases of a benign pulmonary metastasizing leiomyoma. A 35-year-old woman who underwent myomectomy and a cesarean section approximately 6 years earlier visited our hospital for further evaluation of incidentally revealed multiple lung nodules. A diagnostic percutaneous biopsy was performed. Finally, she was diagnosed with a benign metastasizing leiomyoma. The patient then received LH-RH and has been followed up since. The other 44-year-old woman presented after an initial radiology evaluation revealed the presence of multiple, small-sized lung nodules. She underwent a right middle lung wedge resection to confirm the diagnosis. Finally, she was diagnosed with a benign metastasizing leiomyoma. The multiple lung nodules have been followed up closely.

**Key Words:** Benign Metastasizing Leiomyoma; Lung Nodules; Myomectomy; Hysterectomy

## Case Report

### Case 1

A 35-year-old woman presented after the initial radiology evaluation incidentally revealed the presence of multiple lung nodules. She complained of cough and whitish sputum but she denied dyspnea, hemoptysis or fever. There was no history of malignancy. Her medical history included only myomectomy and a cesarean section performed approximately 6 years earlier. No abnormal sounds were present upon auscultation of the chest. A chest X-ray revealed two lung nodules (Figure 1). A chest CT was subsequently performed, which demonstrated two nodules, one 0.8 cm diameter nodule in the right middle lobe and another 1.0 cm diameter nodule in the left upper lobe (Figure 2). There was no mediastinal lymphadenopathy. A diagnostic transthoracic needle biopsy was performed and she was finally diagnosed with a benign metastasizing leiomyoma (Figure 3). The patient then received LH-RH and has been followed up since.
Figure 1. Chest radiograph shows multiple lung nodules.

Figure 2. Chest CT shows two round, well-defined and poorly enhanced nodules.

Figure 3. (A) Microscopic examination of a percutaneous needle biopsy specimen of the lung nodule shows intersecting bundles of spindle-shaped cells, which are strongly-positive for (B) smooth muscle actin immunohistochemical stain, and they highly express both (D) estrogen and (E) progesterone. However, the tumor cells are negative for (C) C-kit (CD 117) immunohistochemical stain (A, H&E stain, ×400).
Case 2

A 44-year-old woman presented after an initial radiology evaluation incidentally revealed the presence of multiple, small-sized lung nodules. The patient denied dyspnea, cough, sputum production, hemoptysis or fever and there was no history of a malignancy. Her significant medical history included only a total hysterectomy performed approximately 10 years earlier. No crackles nor rales were present upon auscultation of the chest. A chest X-ray showed multiple lung nodules (Figure 4). CT of the chest was subsequently performed, which demonstrated multiple lung nodules ranging in size from a few millimeters to 2 cm in diameter (Figure 5). However, there was no mediastinal lymphadenopathy, pleural effusion or calcification. A diagnostic percutaneous biopsy was performed. The lesions were mainly suspected of being a benign spindle cell tumor. She underwent a right middle lung wedge resection to confirm the diagnosis and was finally diagnosed with a benign metastasizing leiomyoma (Figure 6). The patient did not receive any further treatment, and the multiple lung nodules have been followed up closely.

Discussion

Leiomyomas are the most common benign uterine neoplasms of the myometrium. However, a BML is a rare condition characterized by the occurrence of benign smooth muscle tumors. These lesions are most often located in the lung in association with uterine leiomyomatosis. Other sites of involvement include the pelvic lymph nodes, skull base, spine, brain, heart, ret-
Figure 6. (A) The resected nodule is clearly demarcated from the surrounding lung tissue. (B) It shows the typical features of a leiomyoma with no findings suggestive of malignant potential such as coagulative necrosis, increased mitotic activity or significant atypia. (C) The low cuboidal metaplastic bronchiolar epithelia are invaginated and entrapped between the fascicles of the smooth muscle (H&E stain, A, ×40, B, ×200, C, ×200).

eperitoneum etc. This condition may present in women of any age with uterine leiomyoma, Abramson et al reported that nodules can be observed 3 months to 20 years after a hysterectomy or myomectomy. The clinical course is typically indolent, with the patient mortality commonly occurring from an unrelated disease process. There is little correlation between the extent of the disease and the respiratory symptoms.

The etiology of metastasizing leiomyomas includes the presence of metastatic disease from an existing leiomyoma and the presence of multicentric leiomyomatous growths. Estrogen and progesterone receptors have been identified on the lung lesions, which has led to treatment options based on hormonal therapy through surgery or medical oophorectomy. However, surgery or hormonal therapy may not always be indicated, The effects of natural hormonal changes such as pregnancy and menopause have also been associated with the regression of these lesions.

Almost all patients reported thus far were Caucasians. We report two Oriental patients. One woman had myomectomy performed before the discovery of her pulmonary lesions, and the other underwent a total hysterectomy and myomectomy. Both experienced surgical trauma such as a hysterectomy or myomectomy. These cases further support the higher risk of a benign metastasizing leiomyoma after a surgical procedure on the uterus. In our cases, one woman was closely observed without any management for 5 years. Despite the numerous lung nodules, she has rarely complained of respiratory symptoms such as dyspnea, cough, sputum production and hemoptysis. She has remained stable without any evidence of aggravation.

To our knowledge, there are only 2 other cases reported in Korea. Both underwent a hysterectomy due to a benign uterine leiomyoma. After being diagnosed with a benign metastasizing leiomyoma, one was observed and the other was treated with a wedge resection of all the lung nodules.

In conclusion, benign metastasizing leiomyoma is a rare entity that usually affects women after a hysterectomy or myomectomy for leiomyoma. The lesions are usually discovered incidentally and have an indolent clinical course. The lung is the most common site of metastatic involvement. Therefore, benign metastasizing leiomyoma should be considered for any asymptomatic patient presenting with multiple pulmonary nodules and a history of uterine leiomyoma.

References

3. Abramson S, Gilkeson RC, Goldstein JD, Woodard PK,