

Unusual CT Findings of a Benign Metastasizing Leiomyoma Presenting with Multiple Cavitary Nodules: A Case Report¹

Ji Eun Ahn, M.D., Kyung-Hyun Do, M.D., Eun Jin Chae, M.D., Joon Beom Seo, M.D.,
Jin Seong Lee, M.D., Koun-Sik Song, M.D., Jae Wo Song, M.D.,
Kyu-Rae Kim, M.D.², Tae-Hwan Lim, M.D.

A benign metastasizing leiomyoma is a rare condition that affects women with a history of uterine leiomyoma, usually after a myomectomy or hysterectomy. Typical radiographic findings include well-circumscribed solitary or multiple pulmonary nodules ranging from a few millimeters to several centimeters in diameter and scattered among the normal interstitium. We report a case of a benign metastasizing leiomyoma that manifested with multiple cavitary nodules in a 46-year-old woman with no previous history of a myomectomy or hysterectomy.

Index words : Lung

Uterus

Leiomyoma

Tomography, X-Ray Computed

Neoplasm Metastasis

A benign metastasizing leiomyoma is a rare condition that affects women with a history of a uterine leiomyoma. However, a benign metastasizing leiomyoma without a history of previous surgery has rarely been reported. Typical radiographic findings include well-circumscribed solitary or multiple pulmonary nodules ranging from a few millimeters to several centimeters in diameter and scattered among the normal interstitium. We report a case of a benign metastasizing leiomyoma that manifested with multiple cavitary nodules in a 46-year-old woman with no previous history of a myomectomy or hysterectomy.

To the best of our knowledge, there has been only one

case report of a benign metastasizing leiomyoma manifesting as cavitary lung nodules in the English language literature. Therefore, our case is noteworthy because multiple cavitary nodules are an unusual Computed Tomography (CT) finding of a benign metastasizing leiomyoma, and a benign metastasizing leiomyoma is very rare in a patient without a history of previous surgery.

Case Report

A 46-year-old woman was referred to our hospital for evaluation of a uterine mass. On admission, abdominal and pelvic CT scans as well as preoperative chest radiography were performed. An abdominal and pelvic CT showed a 14 × 10 cm, multilobulating contoured uterine mass in the pelvic cavity (Fig. 1A). Based on the abdominal and pelvic CT scans, the radiological diagnosis was a uterine myoma. Chest radiography showed multiple, small nodules in both lungs and post-inflammatory sequela of pulmonary tuberculosis in both upper lobes

¹Department of Radiology and Research Institute of Radiology, University of Ulsan College of Medicine, Asan Medical Center

²Department of Pathology, University of Ulsan College of Medicine, Asan Medical Center

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Address reprint requests to : Kyung-Hyun Do, M.D., Department of Radiology and Research Institute of Radiology, University of Ulsan College of Medicine, Asan Medical Center, 388-1 Pungnap2-dong, Songpa-gu, Seoul 138-736, Korea

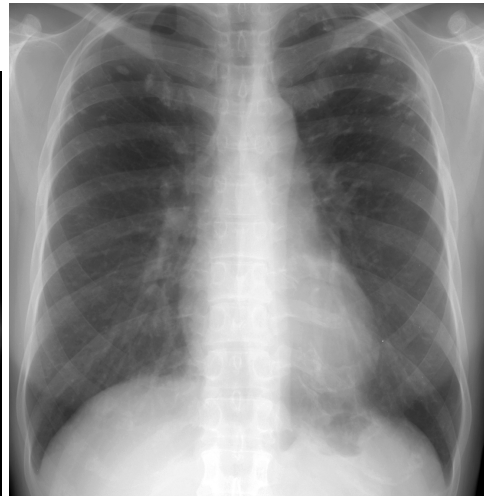
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(Fig. 1B). Clinically, the patient had no respiratory symptoms, and the laboratory tests were within normal ranges. A chest CT was performed for further evaluation. On the chest CT scans, there were multiple, well-circumscribed pulmonary nodules with and without

cavitation, which ranged from a few millimeters to one centimeter in diameter (Fig. 1C - E). There was no pleural change such as pleural effusion or pleural thickening. There was no mediastinal or hilar lymphadenopathy. Although the patient had no history of primary cancer, a



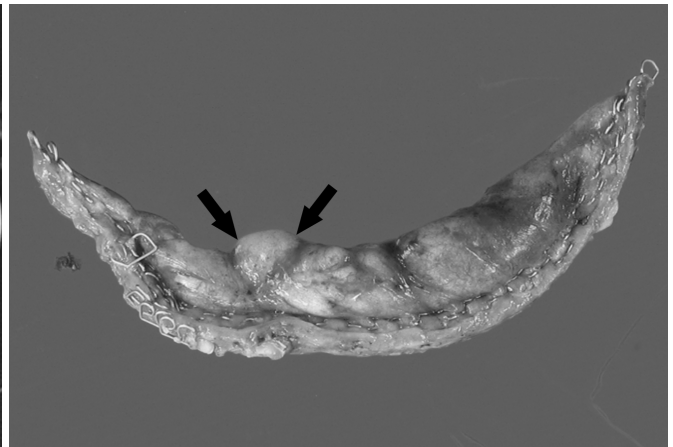
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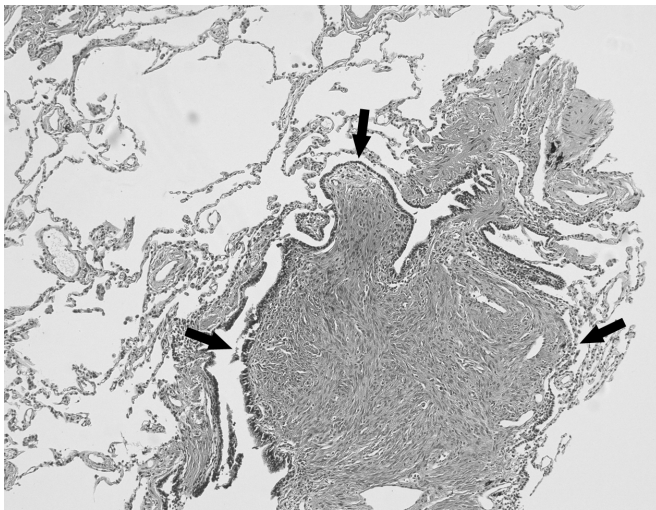
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C



D



E

Fig. 1. A benign metastasizing leiomyoma in a 46-year-old woman with a uterine leiomyoma.

A. A contrast-enhanced CT scan shows a 14 × 10 cm, multilobulating contoured uterine mass in the pelvic cavity (arrows).

B. Chest radiography shows multiple, tiny nodules in the both lungs and post-inflammatory sequelae of pulmonary tuberculosis in both upper lobes.

C. A chest CT shows multiple, well-circumscribed pulmonary nodules with and without cavitation, ranging from a few millimeters to 1 centimeter in diameter (arrows).

D. A photograph of a resected lung specimen shows a well-demarcated, whitish-tan nodule (arrows) approximately 1-cm in diameter.

E. A photomicrograph at low magnification shows that the well-circumscribed tumor mass (arrows) composed of smooth muscle bundles adjacent to the normal alveolar septa (original magnification × 100; haematoxylin and eosin staining).

hematogenous metastasis from an unknown primary malignancy was first considered. A possible fungal infection or septic embolism was also considered. The second radiological diagnosis was a benign metastasizing leiomyoma with cavitation as the patient had a uterine leiomyoma, although this is very rare.

The patient underwent a hysterectomy. Gross examination of the uterine specimen showed a well-defined, huge mass ($3.5 \times 10.5 \times 9$ cm) in the myometrium. The histopathological diagnosis was a benign leiomyoma with no atypical or malignant features. One week after hysterectomy, the patient underwent multiple, wedge resections in the right lung via video-assisted thoracic surgery. Gross examination of the resected lung specimen showed a well-demarcated, whitish-tan nodule, approximately 1 cm in size (Fig. 1F). A histological examination of this pulmonary nodule showed tumor cells composed of benign-appearing, smooth muscle bundles (Fig. 1G - H). Mitosis, cellular atypia or necroses were not found. Following immunohistochemical staining of the pulmonary nodule, smooth muscle actin was strongly positive. The proliferating index was low according to Ki-67 immune staining. Therefore, the histopathological diagnosis of the patient lung was 'benign metastasizing leiomyoma'. Both the histology and the immunohistochemical profiles, i.e., estrogen receptor positive, progesterone receptor positive and very low proliferative index, were identical to those of a uterine leiomyoma.

Discussion

A benign metastasizing leiomyoma is a rare condition that affects women with a history of uterine leiomyoma (1 - 4). Since the first case report by Steiner in 1939, approximately 80 case reports of benign metastasizing leiomyoma have been published. Women that have undergone a hysterectomy for leiomyoma are most commonly affected, and according to the previous reports, pulmonary involvement often occurs several years after the myomectomy or hysterectomy (1 - 4). However, a benign metastasizing leiomyoma without a history of previous surgery has rarely been reported (5).

These patients are usually asymptomatic on presentation, although symptoms such as dyspnea, dry cough or chest pain have been reported (1). Although a leiomyoma is histologically benign, it has the potential to metastasize to distant sites (6). The lung is the most common site of involvement. Skin, bones, mediastinum, lymph nodes, and retroperitoneum can also be involved

(7, 8).

The pathogenesis of a benign metastasizing leiomyoma continues to be a subject of controversy, and several hypotheses have been proposed (9). First, it could be considered as a benign uterine tumor that spreads via a hematogenous route to the lungs or other organs. As a majority of such patients has undergone a myomectomy or hysterectomy, this suggests surgically induced hematogenous spread. Second, it could be a low-grade leiomyosarcoma metastasizing to the lung. Third, it could represent primary pulmonary leiomyomatosis unrelated to but coexisting with the uterine leiomyoma. However, most pathologists now accept the first of these hypotheses, i.e., hematogenous metastases from morphologically benign uterine tumors. In a recent study regarding the pathogenesis of a benign metastasizing leiomyoma, it was suggested to result from the monoclonal, hematogenous spread of a benign-appearing uterine leiomyoma (1). However, in this patient, lung nodules and uterine leiomyoma co-existed prior to the hysterectomy. Although it has rarely been reported, a metastasizing leiomyoma can coexist without a history of previous surgery (5).

A typical radiographic finding of a benign metastasizing leiomyoma is the presence of well circumscribed, solitary or multiple pulmonary nodules (3). Typically, the nodules are not calcified and do not enhance after IV contrast medium administration. Endobronchial and pleural sparing is a characteristic finding of a benign metastasizing leiomyoma. Cavitory change of the lung nodules has been known to represent a rare radiographic finding of a benign metastasizing leiomyoma. To the best of our knowledge, there has only been a single case report of a benign metastasizing leiomyoma manifesting as cavitory lung nodules and interstitial lung disease (10). In this report, there was 5 cm large cavity with an air-fluid level in the left upper lobe of the lung as well as several intrapulmonary and pleura-based, round, mass lesions in both lungs. The investigators explained that the air-filled space represented a dilated and often distorted bronchial tree encompassed in proliferating smooth-muscle cells. Such unique CT features are most likely the combined results of slow tumor growth around the bronchiole/bronchus and dilatation of the bronchial tree secondary to elastic tissue destruction and air trapping.

In summary, we have encountered a rare radiological finding of a benign metastasizing leiomyoma presenting as multiple cavitory lung nodules prior to a hysterecto-

my in a woman with uterine leiomyoma. Although it is very rare, if multiple cavitory pulmonary nodules are seen in a woman with a previous or coincident history of uterine leiomyoma, the possibility of a benign metastasizing leiomyoma can be considered as part of the differential diagnosis.

References

1. Wentling GK, Sevin BU, Geiger XJ, Bridges MD. Benign metastasizing leiomyoma responsive to megestrol: case report and review of the literature. *Int J Gynecol Cancer* 2005;15:1213-1217
2. Barbetakis N, Xenikakis T, Efstathiou A, Vlahveis M, Fessatidis I. Pulmonary benign metastasizing leiomyoma. *J Cardiovasc Surg* 2004;45:399-400
3. Abramson S, Gilkeson RC, Goldstein JD, Woodard PK, Eisenberg R, Abramson N. Benign Metastasizing Leiomyoma: clinical, imaging, and pathologic correlation. *AJR Am J Roentgenol* 2001;176:1409-413
4. Maredia R, Snyder BJ, Harvey LA, Schwartz AM. Benign metastasizing leiomyoma in the lung. *Radiographics* 1998;18:779-782
5. Sabatini R, Ferreri R, Distanti G, Loizzi V, Loizzi P. Benign metastasizing leiomyoma in the lung: a case report. *Eur J Gynaecol Oncol* 2002;23:445-446
6. Takemura G, Takatsu Y, Kaitani K, Ono M, Ando F, Tanada S, et al. Metastasizing uterine leiomyoma: a case with cardiac and pulmonary metastasis. *Pathol Res Pract* 1996;192:622-629
7. Kaplan C, Benirschke K, Johnson KC. Leiomyomatosis peritonealis disseminata with endometrium. *Obstet Gynecol* 1980;55:119-122
8. Kayser K, Zink S, Schneider T, Dienemann H, Andre S, Kaltner H, et al. Benign metastasizing leiomyoma of the uterus: documentation of clinical, immunohistochemical and lectin-histochemical data of ten cases. *Virchows Arch* 2000;437:284-292
9. Patton KT, Cheng L, Papavero V, Blum MG, Yeldandi AV, Adley BP, et al. Benign metastasizing leiomyoma: clonality, telomere length and clinicopathologic analysis. *Mod Pathol* 2006;19:130-140
10. Shin MS, Fulmer JD, Ho KJ. Unusual computed tomographic manifestations of benign metastasizing leiomyomas as cavitory nodular lesions or interstitial lung disease. *Clin Imaging* 1996;20:45-49

2007;57:239 - 242

