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A case of incidental pulmonary benign metastasizing leiomyoma

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Informed Consent
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Abstract

Benign metastasizing leiomyoma is exceedingly rare. Pulmonary benign metastasizing leiomyoma is defined as the metastasis of leiomyoma to the lung tissue. It has a benign histological character, but the presence of metastasis is contradictory. First reported in 1937, it is generally seen among women of young reproductive age and premenopausal women who have undergone uterine leiomyoma or hysterectomy, and the most common site of metastasis is the lung. The prognosis is generally very favorable, and it rarely undergoes a malignant transformation. However, various studies suggest that pulmonary and pleural leiomyoma may cause acute respiratory failure. Radiological findings have a wide spectrum ranging from primary malignant neoplasms to metastatic diseases. Benign metastasizing leiomyoma is usually incidentally found on chest X-rays, and it appears as nodules arising from the benign proliferation of smooth muscles. Here, we present a 46-year-old patient who had a hysterectomy 15 years ago due to uterine leiomyoma and was recently diagnosed with benign leiomyoma which metastasized to the lung. It should be kept in mind that benign leiomyomas with pulmonary metastasis may rarely undergo malignant transformation.

Keywords: Leiomyoma, Hysterectomy, Neoplasm, Metastasis

Introduction

Benign metastasizing leiomyoma, a rare condition with different clinical and radiological features, was first reported by Steiner in 1939 [1]. It usually occurs in women of reproductive age and with a history of uterine leiomyoma. Less than 200 cases have been reported in the literature [2]. In the presence of leiomyoma with benign metastasis, a history of uterine leiomyoma should be questioned.

Benign metastasizing leiomyoma can be seen in many parts of the body, including the lungs the most, and the lymph nodes, muscles, heart, bones, and the central nervous system [3]. Clinical symptoms vary depending on the region of involvement. Most patients are asymptomatic, and the lung lesions are detected incidentally [4]. A case of secondary respiratory failure due to cardiac involvement of the cystic leiomyoma and another one due to the involvement of the left lung lower lobe bronchus were reported in the literature [5].

In chest radiographs, benign pulmonary metastatic leiomyoma typically manifests as multiple pulmonary nodules. Unusual appearances, such as a miliary pattern, and a pedicular pulmonary leiomyoma with a giant cystic formation, have also been reported [6]. We reported this case due to its rarity.

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Case presentation

In September 2019, a 46-year-old female patient, from whom informed consent was obtained to publish this case report, visited our outpatient clinic with a severe dry cough complaint for the last 2 weeks. She had undergone a hysterectomy in 2004 due to uterine leiomyoma. Her family's medical history was uneventful. Physical examination including breast examination was normal. Bilateral parenchymal nodules were observed on the patient's chest radiograph (Figure 1). Computed thorax tomography (CT) was requested for a detailed examination. Enlarged lymph nodes were observed in the mediastinum and the bilateral hilar regions in CT. The largest one, located in the lower right paratracheal region had a short diameter of 1 cm. Scattered in both lung parenchyma, the largest lymph node in the left lung was 2x2 cm, located in the lower lobe superior segment. The nodules were compatible with multiple metastases (Figures 2A, 2B, 2C). Positron emission tomography (PET-CT) revealed no pathological hypermetabolic activity in the multiple lymph nodes scattered across both lung parenchyma (Figure 3A, 3B). Wedge resection was performed with the Video-Assisted Thoracoscopic System (VATS). Microscopic examination revealed benign spindle cells without pleomorphism and mitosis, forming bundles around the alveoli (Figure 4). In immunohistochemical staining, estrogen receptor (ER) and progesterone receptor (PR), actin, desmin, vimentin, cytokeratin, and BCL 2 were positive, while CD34, CD10, CD99 were negative (Figures 5, 6). Histopathological examination of the excised specimen revealed a pulmonary metastatic benign leiomyoma.

Our patient was discharged uneventfully 5 days after the operation. There was no change in pulmonary nodules and or mass recurrence in the thorax CT scan 6 months after the operation.

Especially in reproductive and premenopausal women with a history of uterine leiomyoma, a pulmonary metastatic benign leiomyoma should be kept in mind in the presence of nodules in the lung.

Figure 1: Pulmonary nodules on posteroanterior chest radiography

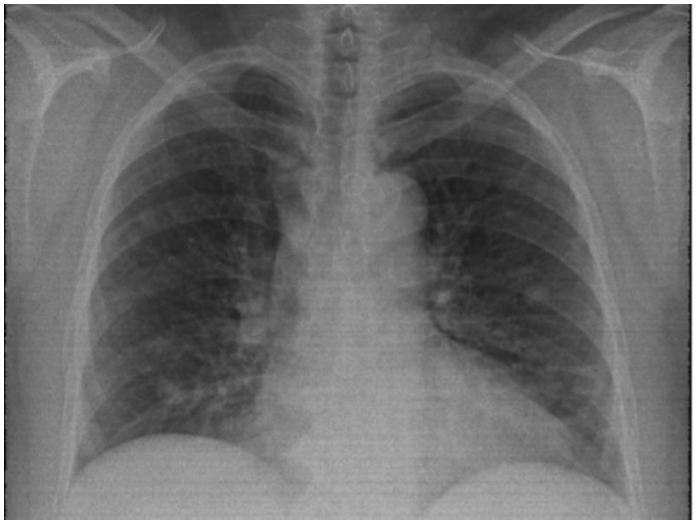


Figure 2: CT image of parenchymal nodules in the lung

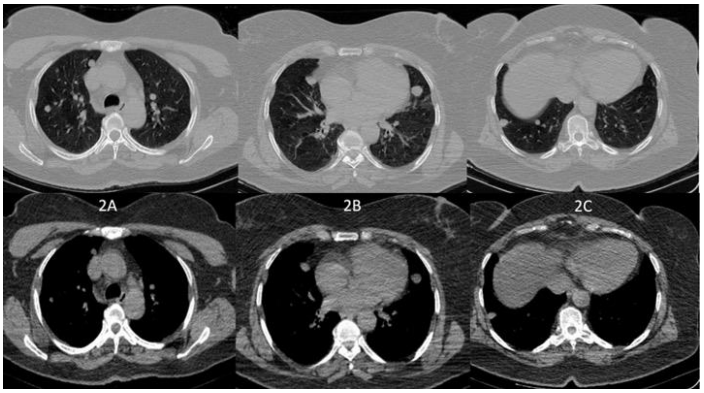


Figure 3: PET-CT view of parenchymal nodules in the lung

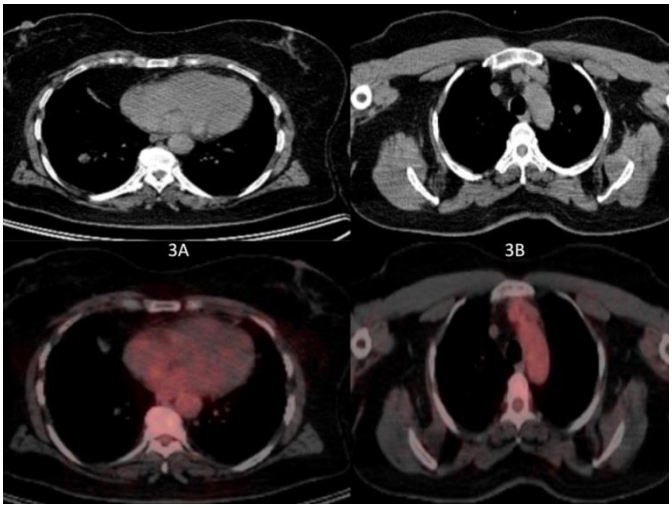


Figure 4: Benign spindle cells forming bundles around the alveoli (HEx200)

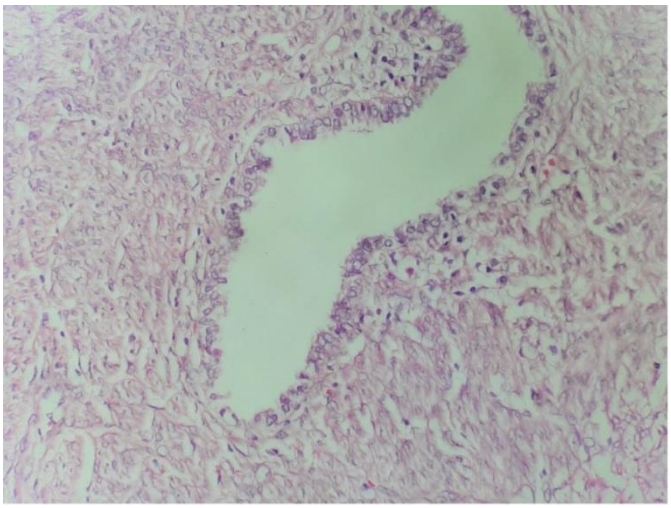


Figure 5: Actin positivity in smooth muscle cells (Actinx400)

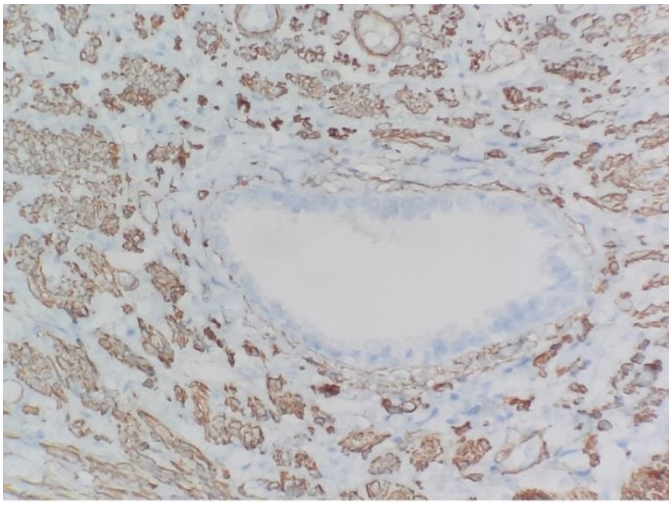
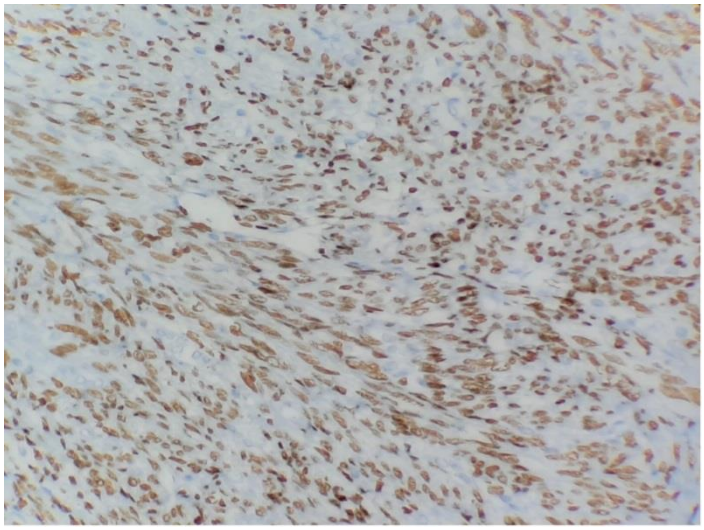


Figure 6: Estrogen receptor positivity in smooth muscle cells (ERx400)



Discussion

Uterine leiomyoma is the most common pelvic tumor in women of reproductive age with an incidence varying between 12-25% [7]. Metastasizing leiomyoma is directly related to uterine leiomyoma and may develop years after myomectomy or hysterectomy. Benign metastasizing leiomyoma is most common among females aged 30-74 years. It may occur in different regions including the lung, liver, heart, breast, and esophagus [8], and rarely undergoes malignant transformation [2].

Approximately 100 cases of malignant transformation of the metastasizing leiomyoma were reported. The pathogenesis and propagation path of PBML is not clear. It is hypothesized that leiomyoma spreads hematogenously during uterine surgery [8].

However, the progress in primary pulmonary leiomyosarcoma is slow. The incidence of sarcomatous transformation of benign uterine leiomyomas is only 0.1-0.8% and is most common among women in their 50s. [9]. Early diagnosis is important and achieving negative surgical margins and wide resection are the standard treatments. The 5-year survival rate is above 50% in patients with complete resection and recurrence is rare. Since lymph node involvement of primary pulmonary leiomyosarcoma is also uncommon, lymph node dissection is unnecessary. Chemotherapy, radiation therapy or both can be planned as adjuvant therapy for advanced lesions [10].

Lung involvement may present with varying images in lung imaging, ranging from unilateral or bilateral nodules to cystic or cavitary lesions of different sizes [11].

PBML has slow growth potential and a limited effect on lung function. However, it can mimic lung cancers or rare benign diseases of the lung, such as tuberculoma, pneumoconiosis, or sarcoidosis [11].

Approximately 200 BML cases were reported [2]. Its pathogenesis is unknown, but the most common theory is the hematogenous spread of leiomyoma during uterine surgery. Excisional biopsy is used for standard diagnosis. SMA, desmin, actin, ER, and PR are positive in immunohistochemical staining of the pathological specimen [12].

Most cases of PBML do not require treatment, but patients with metastasis, increased nodule size and respiratory failure, and hemoptysis should be treated. Surgical treatment should be preferred in these cases. Depending on the size of the tumor and the state of malignancy, radiotherapy, chemotherapy, or both can be administered. In some PBML cases that are positive for ER or PR, reducing hormone release through a hysterectomy, bilateral adnexectomy, and/or hormone therapy can reduce tumor size [13].

Radiologically, it is difficult to distinguish between the malignant transformation of PBML and benign PBML. The only effective diagnostic imaging tool is PET-CT [14].

In our case, no malignant transformation was observed. Microscopic examination of the piece excised from the superior segment of the left lower lobe revealed spindle cell tumor cells without pleomorphism or nuclear atypia and the pathological report was consistent with PBML.

The standard diagnostic procedure for PBML is excisional lung biopsy. Pathologically, the current criteria used to differentiate a malignant smooth muscle tumor from a benign mass are necrosis, high mitotic index (5 mitoses at 50 HPF), and nuclear atypia [15].

Conclusions

In this disease, malignant transformation is very rare. Early diagnosis and total mass resection are of great importance in the case of malign transformation of PBML. If there is an increase in nodule size in CTs during follow-ups, new lesions are detected or there are findings suggesting malignancy in PET-CT, diagnostic procedures should be performed, and surgery should be planned if necessary.

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