Pulmonary Leiomyomatosis After Hysterectomy For Uterine Myoma

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Citation

Abstract
Pulmonary benign metastasizing leiomyoma refers to a neoplasm associated with a previous or coincident history of uterine leiomyomas. A recent study by Sekine et al [1] reports that leiomyomas represent 0.085% of pulmonary benign tumors. Multiple pulmonary leiomyomas represent an exceptional condition that occurs usually in sexually mature women. We present the case of a 49-year old woman with pulmonary benign metastasizing leiomyoma 10 years after hysterectomy for uterine leiomyoma.

CASE REPORT
A 49-year old woman was admitted to our department for investigation of accidentally found space-occupying lesions in both lungs in chest X-ray (Figure 1).

Figure 1
Figure 1: Chest X-ray showing bilateral multiple pulmonary nodules

The past medical history revealed that she underwent hysterectomy 10 years ago for leiomyomas of the uterus. She also reported hyperlipidaemia and smoking 20 cigarettes daily since the age of 18. The patient was free of dyspnea, cough, sputum production, hemoptysis or fever. Physical examination was normal.

Computed tomography of the thorax confirmed the presence of bilateral pulmonary multiple nodules. As multiple metastases due to tumor of unknown origin were suspected, the patient underwent a series of diagnostic tests: computed tomography (brain, abdomen), gastroscopy, coloscopy, bronchoscopy, skeletal scintigraphy and various laboratory tests none of which indicated primary extrapulmonary tumor.

A percutaneous CT-guided biopsy was performed with negative results. Therefore, an exploratory posterolateral mini-thoracotomy through the 5th intercostal space was carried out and histopathological examination of specimens obtained from a resected tumor was performed. The resected tumor revealed bundles of smooth muscle cells without high cellularity or mitosis with nuclear atypism. This was consistent with benign leiomyomas without any evidence of malignancy (benign metastasizing pulmonary leiomyoma – Figure 2). The tumor also showed a reactivity against actin and desmin. Estrogen receptors were present.
Figure 2
Figure 2: Bundles of smooth muscle cells without evidence of malignancy (Benign metastasizing leiomyoma).

The postoperative period was straightforward with no problems. According to the fact that the histologic report showed a high concentration of estrogen receptors, an analogue gonadotropin-releasing hormone (Gn-RH) was given. Ten months later a stable condition in size of the nodular lesions was observed.

DISCUSSION

The etiology of multiple pulmonary nodules is quite complex, with metastatic disease being the most common cause. Other possibilities include sarcoidosis or an inflammatory process, such as fungal infection, tuberculosis, nocardiosis or septic emboli. However, in asymptomatic patients, further considerations include the presence of amyloidosis, hamartomas, rheumatoid nodules, arteriovenous malformations and leiomyomas [2].

Benign metastasizing leiomyoma is a rare cause of a space-occupying pulmonary lesion with unclear pathogenesis, predominantly affecting middle-aged women after hysterectomy for uterine myoma [3]. Frequently, the pulmonary involvement appears several years after myomectomy or hysterectomy with radiographic appearance of slow-growing solitary or multiple lung nodules. Although most of these patients are asymptomatic, presenting symptoms such as dyspnea, dry cough or chest pain have been reported [4]. The most common sites of metastases include the lungs and lymph nodes. A metastatic focus involving the anterior papillary muscle of the right ventricle causing obstruction of the main pulmonary trunk has been described [5].

The pathogenesis remains unclear, hormone-dependent tumor growth being discussed as a possible mechanism. Against it, is that extrapulmonary locations are too rarely observed. The still open pathogenetical question has no therapeutic consequence. Whenever technically possible, a radical parenchyma-saving surgical therapy should be the first choice [6]. Anti-oestrogen administration is the treatment of choice to achieve remission and effective prevention of recurrences [7].

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References

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