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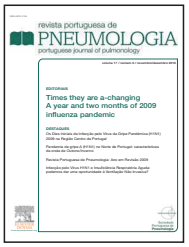

CASE REPORT

Argon-plasma treatment in benign metastasizing leiomyoma of the lung: a case report

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KEYWORDS
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Abstract
Benign metastasizing leiomyomas of the lung are rare smooth muscle cells tumours. We report the case of a 48 year-old female who was evaluated due to persistent cough, progressive dyspnoea and constitutional symptoms. Chest computed tomography revealed a left endobronchial mass, multiple parenchyma nodules and a pleural effusion. Bronchial biopsy histological features were consistent with benign metastasizing leiomyoma. The patient was successfully treated with argon-plasma and mechanical debulking. There was no disease relapse in the last four years.

PALAVRAS-CHAVE
Leiomioma benigno metastizante;
Neoplasias pulmonares;
Diagnóstico;
Broncoscopia;
Tratamento;
Tratamento árgon-plasma

Árgon-plasma no tratamento de leiomioma benigno metastizante pulmonar: um caso clínico

Resumo
Os leiomiomas benignos metastizantes pulmonares são tumores raros de células musculares lisas. Uma doente de 48 anos foi avaliada devido a tosse persistente, dispneia progressiva e sintomas constitucionais. A tomografia computorizada do tórax revelou uma massa endobrônquica à esquerda, múltiplos nódulos do parênquima pulmonar e derrame pleural. As características histológicas da biopsia brônquica foram consistentes com o diagnóstico de leiomioma benigno metastizante. A doente foi submetida a coagulação árgon-plasma e desobstrução mecânica com eficácia terapêutica. Verificou-se estabilidade clínica nos últimos quatro anos.

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Introduction

There are few references concerning pulmonary leiomyomas and it is estimated that they account for less than 2%of all lung benign tumours.[1-8] Although benign, in terms of histology, they behave as malignant tumors based on their metastasizing potential, involving most commonly lung parenchyma, bronchi, distal trachea and more rarely the peritoneum, retroperitoneum structures, lymphatic system and central nervous system.

Case presentation

A 48 yr-old non-smoker female was referred with complaints of fatigue, progressive dyspnoea on effort, non-productive cough, non-pleuritic chest pain, as well as anorexia and weight loss of 8kg which started 12 months previous to initial symptoms. She denied other complaints. Her past medical history included a hysterectomy without oophorectomy for fibroid tumour at age 45.

Blood sample analysis was unremarkable. Thorax CT revealed a complete atelectasis of the left lung due to a left main bronchus mass, homolateral pleural effusion and multiple contralateral lung parenchyma nodules.

Flexible bronchoscopy exposed an endoluminal mass, 2 cm from the main carina, which occluded the left main bronchus (Figure 1). No other tracheobronchial tree changes were visualized. Bronchial biopsies confirmed the presence of a fusocelular neoplasia with no mitotic activity or polymorphisms, with vimentin and actin positivity as well as with estrogen and progesterone receptors positivity, consistent with a benign metastasizing leiomyoma.

Endoscopic treatment was proposed and rigid bronchoscopy performed under general anesthesia. Argon-plasma and mechanical debulking totally restored airway lumen.

There was clinical improvement and the flexible bronchoscopy, performed 7 days after the procedure, confirmed a normal left main bronchus lumen. Control chest CT showed parenchyma nodules stability with minimal pleural effusion (Figure 2). The patient was asymptomatic and refused other treatments. In the last four years there was clinical and radiological stability of the pulmonary lesions.

Discussion

Leiomyomas are smooth muscle cells benign tumors and they are most common in the female genital tract.[2]

Metastasizing leiomyomas occur predominantly in the female sex with a higher incidence around the fourth decade of life, although a third of cases can occur before twenty years of age.[1,6] The existence of a previous uterine surgery is common[1,3,4,6] and our patient confirms it since she had a history of uterine leiomyoma four years before lung presentation.

Leiomyomas of the lower respiratory tract are usually asymptomatic and can be found incidentally on chest radiography or CT scan, performed for other reasons.[3,6] Clinical presentation depends on the size and location of the tumour[3,6] as in the present case, where complaints were related with the nature of involvement—parenchyma, bronchial and pleural. Typical radiographic findings include well-circumscribed solitary or multiple pulmonary nodules, ranging in size from a few millimetres to several centimetres in diameter, scattered along normal interstitium. Classically, these nodules are not calcified or cavitated, do not enhance after IV contrast administration and show little change over time or may even spontaneously regress.

These tumors show low mitotic activity and contain estrogen and progesterone receptors.[2,6] The slow growth along with progressive development of metastasis is characteristic and generates consequent morbidity. The benign histology and malignant behaviour is puzzling and has not been fully clarified. Benign metastasizing leiomyomas have been described as a smooth muscle neoplasm of uncertain malignant potential. For histological reasons they cannot be classified as malignant tumors and can be difficult to distinguish from a heterogeneous group of smooth muscle tumors.[8] The differential diagnosis of leiomyomatosis lesions of the lung includes primary pulmonary leiomyoma, benign metastasizing leiomyoma,

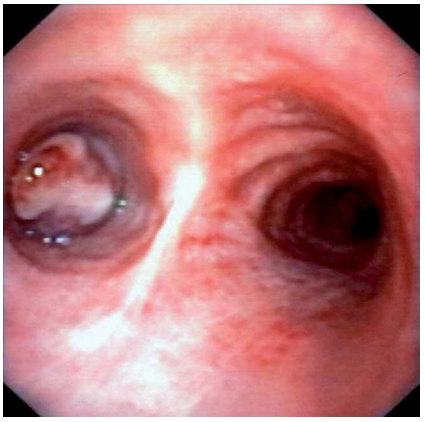

Figure 1 Flexible bronchoscopy reveals an endoluminal mass in the left main bronchus.

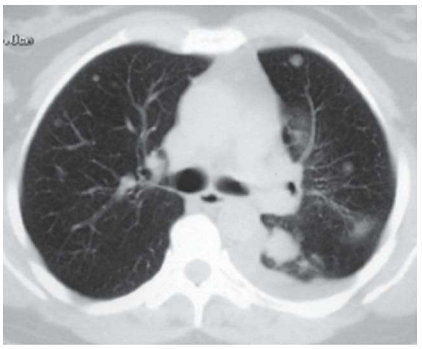

Figure 2 After endobronchial treatment thorax CT scan shows a patent left main bronchus, parenchyma nodules stability and minimal pleural effusion.

leiomyosarcoma, metastatic leiomatosis tumor of an extrauterine source, pulmonary hamartoma and lymphangioleiomyomatosis.[6]

Estrogen and progesterone receptors in the pulmonary lesions allow medical or surgical treatment. There can be a stabilization, slow growth or even regression of the lesions after menopause, oophorectomy or administration of hormone therapy (analogous of gonadotropin-realising hormone, progesterone, tamoxifen, modulators of estrogen receptors and aromatase inhibitors) factors that confirm a hormone-dependent growth.[4,5]

Endoluminal lesions should be removed and interventional endoscopy or surgical treatments are valuable options.[6,7] Argon-plasma and mechanical debulking was our choice and proved to be effective in maintaining airway lumen since there was no relapse in the last four years.

Conclusion

Though uncommon, the diagnosis of a lung benign metastasizing leiomyoma should be suspected when there are compatible radiological features, uterine myomas and the absence of primary malignant illness. Final diagnosis is always histological and long-term follow-up is imperative in view of the fact that relapse may occur.

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