

Benign Metastasizing Leiomyomatosis Presenting as Giant Pleural Mass Without Pulmonary Involvement[☆]



Presentación de leiomiomatosis benigna metastatizante como una masa pleural gigante sin afectación pulmonar

To the Editor:

We report a case of benign metastasizing leiomyomatosis (BML) manifesting as a giant pleural mass. To the best of our knowledge, this is the first reported case of thoracic BML with isolated pleural involvement (no pulmonary involvement), and the only report in the literature of pre-operative embolization of this type of lesion to minimize the risk of bleeding during surgery.

The patient was a 48-year-old woman, non-smoker, who presented due to a 1-month history of cough. Her only significant medical history was hysterectomy for uterine myomatosis 9 year previously. A large mass in the right hemithorax was observed on chest X-ray. Chest computed tomography (CT) revealed a large, solid, extraparenchymal mass, 20 cm in its longest diameter, with marked arterial vascularization from the right phrenic, intercostal and bronchial arteries, and right internal thoracic artery (Fig. 1).

A CT-guided core needle biopsy was obtained from the pleural mass and diagnosed as leiomyoma. Histology was identical to that of the previously resected uterine myomas. On the basis of the histology results, a presumed diagnosis of BML was given.

Given the significant systemic vascularization of the pleural mass, we decided to perform preoperative embolization with a view to simplifying resection of the lesion. Selective embolism of the distal branches of the right internal thoracic artery and the right phrenic artery was performed with 500 microparticles, and vascularization of the mass was successfully reduced. Two days later, right posterolateral thoracotomy was performed and the pleural mass was completely removed. Bleeding during the procedure was estimated to be less than 100 ml. Pathological examination of the resected specimen confirmed the diagnosis of BML. No significant complications occurred during the postoperative period. Twelve months after surgery, the patient remains asymptomatic and without radiological signs of local or distant relapse.

BML is a rare disease, characterized by the development of smooth muscle tumors originating from uterine leiomyomas that require hysterectomy in most cases. The mean time from hysterectomy to the appearance of metastasis is 15 years with a range of 3–20 years.^{1,2}

The most widely accepted etiopathogenic theory is that MBL metastases are caused by hematogenous spread from the uterine tumor at the time of surgery, but isolated cases have been reported in which metastasis has appeared even before hysterectomy.^{1,2} Thoracic MBL metastasis occurs most frequently in the lung, but lymph node, cardiac, and vertebral and retroperitoneal spine involvement have been reported.³ Most patients are clinically asymptomatic and MBL is discovered by chance during imaging studies performed for another reason. Some patients with thoracic involvement can have symptoms such as cough, chest pain and dyspnea.^{2,3}

Only one case of BML with pleural involvement has described the scientific literature, but this was associated with lung lesions.⁴ Our case is the only one published to date with isolated pleural involvement and no pulmonary lesions, and is also the largest BML chest lesion reported to date (20 cm).

Treatment of choice is usually surgical resection, often accompanied by estrogen suppression via bilateral oophorectomy or hormone treatment (due to the presence of estrogen and progesterone receptors in both metastatic lesions and uterine leiomyomas).² In our case, we decided to perform preoperative embolization due to the large size and hypervascularity of the tumor. This is the first case of BML with presurgical embolization for reducing the risk of intraoperative bleeding reported in the literature, although this procedure is common in other thoracic tumors.⁵ One case of embolization of a pulmonary MBL has been reported, but in that case, the intention was to treat life-threatening hemoptysis.⁶

In conclusion, this is an exceptional case of giant MBL with isolated pleural involvement in a patient with a history of uterine myomas. The pleural mass was embolized before surgery to minimize the risk of bleeding.

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Fig. 1. Posteroanterior chest X-ray (A) showing a large mass in the right hemithorax. Chest computed tomography with intravenous contrast medium confirming the presence of a solid extraparenchymal mass in the pleura (B) with marked systemic arterial vascularization from the right bronchial, intercostal, internal thoracic and phrenic arteries (C, coronal reconstruction with maximum intensity projection).

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Bronchial Thermoplasty for Severe Asthma: Initial Experience in Chile[☆]



Termoplastía bronquial para el manejo del asma severo: experiencia inicial en Chile

To the Editor:

Bronchial asthma is becoming more prevalent in adults. Around 10.2% of the population of Chile reports suspected asthma, with similar rates between men and women.¹ The recently developed technique of bronchial thermoplasty (BT) has been shown to be a safe and effective option for the treatment of severe asthma, and improves the quality of life of these patients.^{2,3}

We report the first 4 cases of patients with severe asthma treated with BT in Chile. All were receiving high-dose corticosteroids and inhaled β -agonists (fluticasone or equivalent >1000 μ g/day), rescue inhalers >3 times a day, leukotriene inhibitors and continuous systemic corticosteroids. Table 1 shows the baseline characteristics of each patient. They all underwent the first 2 sessions of BT on an outpatient basis, and there were no complications. After the third session, all 4 patients had a severe asthma attack and required hospitalization. The first patient was hospitalized for 48 h, the second for 6 days, and the third and fourth patients for 7 days each. Patient number 4 also required 3 days of non-invasive mechanical ventilation.

The interval between treatments for all patients was 4 weeks or more. Baseline forced expiratory volume in 1 s (FEV1) before each session remained stable. All patients received 50 mg prednisone for 3 days before the procedure, on the day of the procedure and the following day.

BT is a bronchoscopic procedure consisting of 3 sessions of radiofrequency ablation of airway smooth muscle. The right lower lobe is treated during the first session, the left lower lobe during the second, and in the third, both upper lobes are treated. Time between sessions is 4–6 weeks.⁴

A recent systematic review that included the most relevant randomized clinical trials (AIR, AIR II and RISA) found improvements of 0.28 (0.07–0.50) and –0.15 (–0.4 to 0.10) in AQLQ and ACQ scores, respectively; the hospitalization RR during the treatment period was 3.5 (1.26–9.68) with I^2 of 0.0%, and post-treatment RR was

1.12 (0.44–2.85). No improvement was found in the need for rescue medication: –0.68 (–3.63 to 2.28), nor was there any significant change in lung function.⁴

This is the first report of patients receiving BT in Chile, and the second from Latin America.⁵ The baseline characteristics of our patients were similar to those previously described (RISA study). However, the need for hospitalization after the third treatment needs to be addressed. The average rate of asthma attacks per year in our patients before the intervention was high, and they required high doses of corticosteroids, factors that could have been related to the need for hospitalization after the third session. However, after the interventions, all our patients went on to have an excellent clinical response. Two of them were able to completely discontinue systemic corticosteroids and the other 2 reduced their dose by half. They all had fewer, less severe exacerbations. These findings should be taken into account in future studies.

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Conflict of Interests

The authors declare that they have no conflict of interests.

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Table 1

Baseline Characteristics of the 4 Patients Undergoing Bronchial Thermoplasty.

Age (years)	Sex	Baseline FEV1/L (%)	Systemic corticosteroid dose (prednisone)	Number of exacerbations/year
73	Male	1.45 (53.2)	20 mg/day	6
47	Female	1.53 (58.3)	40 mg/day	8
30	Female	1.92 (70.1)	15 mg/day	5
68	Female	1.18 (59.9)	10 mg/day	6

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