

Pulmonary Benign Metastasizing Leiomyomatosis presenting Giant Bullae with lung function decline

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Introduction: Benign metastasizing leiomyoma (BML) is a rare disorder in which histologically benign smooth muscle tumors metastasize to extrauterine sites. We report a case of pulmonary BML with lung function decline due to secondary bullous change who successfully underwent bullectomy.

Case report: Previous healthy 53-year-old woman was diagnosed with a uterine leiomyoma in 2011. During follow up, multiple lung nodules were diagnosed, and she underwent hysterectomy with bilateral oophorectomy in 2014. Pathology revealed benign leiomyoma. Under diagnosis of BML, hormonal treatment using letrozole was administered for lung nodule. In 2019, she presented with left pulmonary bullae, but remained asymptomatic. However, after she voluntarily stopped taking letrozole and 3 months later, in May 2023, the size of bullae and nodule increased (Fig.1). PFT showed obstructive ventilation defect, including gradual decrease in FVC, FEV1 and TLC with exertional dyspnea. In June, video-assisted thoracoscopic surgery bullectomy was performed. Grossly, there were two giant bullae (up to 10.3x5.8x0.7 cm) surrounding mass. Pathology of the mass showed benign leiomyoma, consisting of spindle-shaped cells without nuclear atypia in fascicular pattern and it was confirmed that smooth muscle cell grew and thickened lining the wall of bullae (Fig.2). On immunohistochemical staining, cells were positive for SMA, Desmin, Ki67, and CD31. She was discharged on the 5th postoperative day without any complication (Fig.3). On the 25th postoperative day, her FVC, FEV1, TLC increased, and RV, as a measure of air trapping, decreased respectively with symptom improvement (Table1).

Conclusion: This case highlights a rare presentation of pulmonary benign leiomyoma with secondary bullous change. BML can regrow after stopping hormonal treatment, even in menopause. The potential mechanisms for the development of bullae are secondary obstruction and check-valve phenomenon of distal airway or cyst-forming tumor growth lining the bullae wall. In case of progressive secondary bullous changes with lung function decline, bullectomy can improve the symptom and pulmonary function.

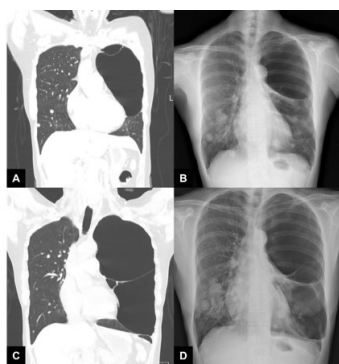


Fig.1 Initial Chest CT and Chest X-ray (A,B) shows multiple lung nodules with left bullae formation. Follow up chest image 3 months after stopping taking letrozole (C,D) shows increased size of bullae and nodules.

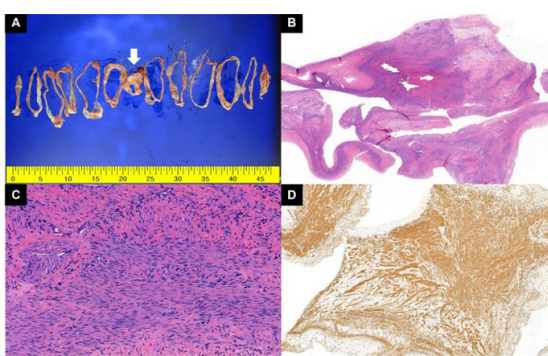


Fig.2 (A) Gross findings of pulmonary bullae surrounding leiomyoma (arrow). (B,C) Hematoxylin and Eosin stain (x50, 200). (D) Immunohistochemical stain for smooth muscle actin was positive (x100)

	20.05.14	22.05.31	23.05.23	23.07.25
FVC	2.71(80%)	2.25(67%)	1.87(56%)	2.92(87%)
FEV1	2.11(70%)	1.78(63%)	1.44(51%)	2.52(89%)
TLC	4.54(93%)	4.33(88%)	4.20(86%)	4.37(89%)
DLCO	82	64	58	66
RV	1.73	2.03	2.26	1.45

Table.1 PFT showed gradual decrease in FVC, FEV1, TLC and increase in RV during follow up (20.05.14~23.05.23). After bullectomy (23.07.25), PFT dramatically improved.



Fig.3 Chest X-ray after bullectomy.