Respir Case Rep 2023;13(1):6-8 DOI: 10.5505/respircase.2024.02223

Is Surgery a Must? A Case of Endobronchial Leiomyoma Removed via Rigid Bronchoscopy

Tek Seçenek Cerrahi mi? Rijid Bronkoskopi ile Çıkarılan Endobronşiyal Leiyomyom

Aytekin İdikut, Gamze Göktaş, Oğuz Karcıoğlu, Ziya Toros Selçuk

Abstract

Pulmonary leiomyoma is a very rare benign lesion that usually develops as a result of bronchial metastasis in women with a history of uterine leiomyoma or myomectomy. Although primary pulmonary leiomyomas are less common than the metastatic forms, they can be detected incidentally or when complaints such as cough and dyspnea are present. The structure of the lesion and its relationship with the bronchial mucosa play an important role in the determination of the treatment option. Endobronchial methods such as rigid bronchoscopy and electrocauterization may be superior to surgery as they are less invasive.

Keywords: *Leiomyoma, rigid bronchoscopy, tracheobronchial tumors.*

Öz

Akciğer leiyomyomu oldukça nadir görülen ve genellikle uterusta leiyomyom veya miyomektomi öyküsü olan kadınlarda bronşa metastaz sonucu görülen benign lezyonlardır. Primer pulmoner leiyomyom, metastatik olana göre daha nadir görülmek ile beraber, öksürük ve nefes darlığı gibi şikayetler olması üzerine ya da insidental olarak akciğer görüntülemelerinde saptanabilmektedir. Lezyonun yapısı ve bronş mukozası ile ilişkisi, tedavi seçeneğinin belirlenmesinde önemli bir rol oynamaktadır. Rijid bronkoskopi ile elektrokoterizasyon gibi endobronşiyal yöntemler kullanılması, daha az invazif olması nedeni ile cerrahiye üstünlük sağlayabilmektedir.

Anahtar Kelimeler: *Leiyomyom, rijid bronkoskopi, trakeobronşiyal tümörler.*

Department of Chest Diseases, Hacettepe University, Ankara, Türkiye

Hacettepe Üniversitesi Tıp Fakültesi, Göğüs Hastalıkları Anabilim Dalı, Ankara

Submitted (Başvuru tarihi): 07.08.2023 ***Accepted (Kabul tarihi):*** 09.10.2023

Correspondence (İletişim): Aytekin İdikut, Department of Chest Diseases, Hacettepe University, Ankara, Türkiye

e-mail: aytekinidikut@gmail.com

Leiomyomas are benign tumors that occur in smooth muscle and that can arise in the airway in the absence of a history of uterine leiomyomas (1). First described by Frokel in 1909, pulmonary leiomyomas are benign tumors originating in the mesoderm (2,3). The optimum treatment of leiomyomas depends on the location of the lesion and the clinical symptoms present (4). In our case, an endobronchial tumor excised by rigid bronchoscopy was diagnosed as leiomyoma.

CASE

A 71-year-old non-smoking male patient with a history of hypertension, type 2 diabetes mellitus and kidney donation were admitted to the Department of Chest Diseases with dyspnea, an occasional dry cough and no other symptoms, and a physical examination revealed no remarkable findings. Pulmonary function test results revealed FEV1 2.6 L (93%), FVC 3.4 L (95%) and FEV1/FVC 76.4%. A 16x12 mm polypoid lobulated lesion occupying the entrance of the left lower lobe bronchi (Figure 1) was identified on thorax CT, while fiberoptic bronchoscopy revealed a polypoid, soft, non-vascular and mobile endobronchial lesion originating in the lateral wall of the end of the left main bronchus that was almost completely blocking the lumen (Video 1, Figure 2). PET-CT confirmed a very low FDG uptake (11x16 mm, SUVmax: 2,7), and no findings compatible with metastasis. Since the lesion had a narrow base and did not extend beyond the bronchi, it was electrocauterized via rigid bronchoscopy using a 30-watt cautery, during which the base of the lesion was captured with a snare and almost all of it was resected (Video 2). The presence of neoplastic cells that stained positively with desmin and H-Kaldesmon and negatively with ALK, CD34 and S100 confirmed the diagnosis of leiomyoma. A thorax CT conducted 1 month after the procedure revealed no lesion within the lumen. The patient was scheduled for follow-up of symptoms with a physical examination and imaging procedures every 6 months.

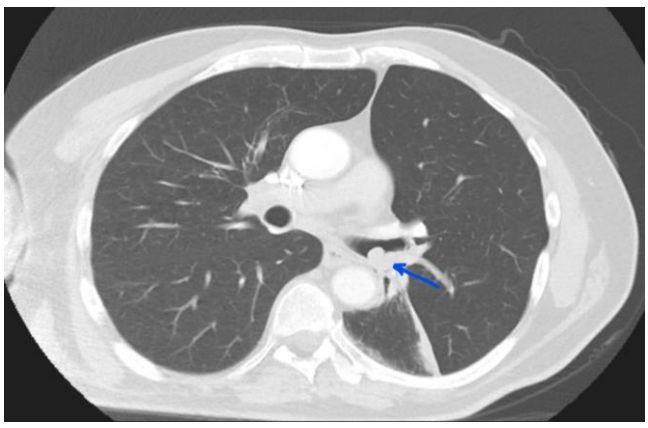

Figure 1: *Tumor in the distal left main bronchus on Thorax CT*

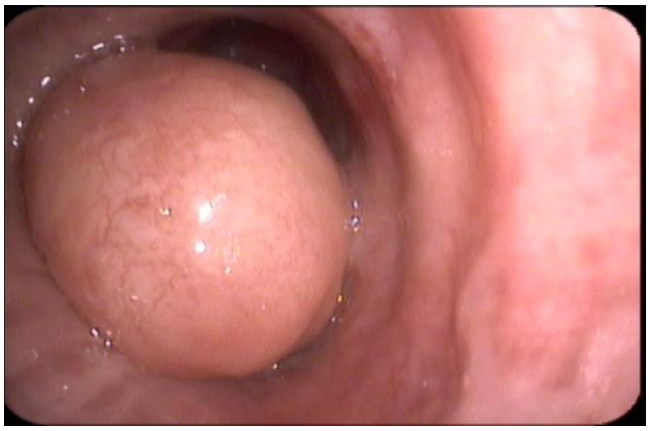

Figure 2: *Appearance of Endobronchial Leiomyoma in Fiberoptic Bronchoscopy*

DISCUSSION

Leiomyomas are benign tumors comprised of smooth muscle cells that are immunohistochemically stained by actin and desmin. They are usually well-circumscribed and larger than 1 cm (5,6) and may also affect the respiratory system in rare cases, originating from pulmonary parenchyma, as well as the airways (7). Leiomyomas occurring only in the lung are referred to as primary pulmonary leiomyomas (PPL), and as benign metastasizing leiomyoma (BML) if found in the lungs of women with a history of uterine leiomyoma (1,4). Although they can radiologically be confused with malignant lesions, they can be pathologically proven to be benign based on a low mitotic index and no evidence of invasion.

Pulmonary leiomyomas are referred to as endobronchial when they emerge in the proximal airways (up to the segmental level), and as parenchymal when they originate in peripheral airway tissue and spread to the rest of the lung (8). The most common symptoms of endobronchial leiomyoma are cough, wheezing, dyspnea, chest pain and recurrent respiratory infections, and may be misdiagnosed as asthma, reflux and post-infectious cough due to such non-specific symptoms. Their local effect can cause atelectasis by blocking the bronchus in their location and can lead to post-obstructive pneumonia (4), and some patients may also experience hemoptysis (4,9,10). In two-thirds of patients with PPL, the condition manifests as an endobronchial lesion in the proximal airways. There are as yet no published guidelines advising on the management of endobronchial leiomyomas. Resections may vary depending on the size and location and the relationship between the lesion's base and the airway wall. Rigid bronchoscopy is a well-established method that facilitates the resection of endobronchial tumors while protecting airway patency (4,11). In cases with narrow-based lesions, advanced bronchoscopic techniques such as electrocautery, laser and argon plasma coagulation may be used during rigid bronchoscopy to resect the tumor (12). Surgical resection is the general-

ly preferred technique for wide-based lesions, endobronchial lesions, tumors located in the distal bronchial tree and parenchymal tumors (4). Prognosis is, on the whole, favorable following the full excision of the lesion (4,10–12), although patients should be followed up at regular intervals to confirm that no recurrence has occurred (7).

CONCLUSION

PPL, as one rare tumor of the tracheobronchial tree, may present with symptoms such as cough, dyspnea, hemoptysis and chest pain resulting from the local effect on the tree, although they may also be detected incidentally on thorax imaging. The bronchoscopic appearance of the lesion should raise suspicion for diagnosis and guide the appropriate treatment option. The endobronchial approach stands out as the optimum approach to narrow-based lesions due to lower tissue loss than with surgery. PPL can be followed radiologically or undergo bronchoscopic or surgical resection, depending on the clinical manifestations. Assessment of treatment options should consider the impact.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - A.İ., G.G., O.K., Z.T.S.; Planning and Design - A.İ., O.K., G.G., Z.T.S.; Supervision - A.İ., O.K., Z.T.S., G.G.; Funding - G.G., A.İ. ; Materials - G.G., A.İ.; Data Collection and/or Processing - A.İ., G.G., O.K.; Analysis and/or Interpretation - A.İ., O.K.; Literature Review - A.İ., O.K., Z.T.S.; Writing - A.İ.; Critical Review - A.İ., O.K., G.G., Z.T.S.

REFERENCES

1. Midthun DE. Benign Lung Tumors. In: Spiro SG, Silvestri GA, Agustí A, eds. Clinical Respiratory Medicine. Fourth Ed. Philadelphia: W.B. Saunders; 2012:810-7. [CrossRef]
2. Allan JS. Rare solitary benign tumors of the lung. Semin Thorac Cardiovasc Surg 2003; 15:315-22 . [CrossRef]
3. Forkel W. Ein Fall von Fibromyom der Lunge. Zeitschrift für Krebsforschung. 1909; 8:390-3. [CrossRef]
4. Nair A, Leong P. The panorama of pulmonary leiomyoma: A tale of two tumours. Respirol Case Rep 2023; 11:e01088. [CrossRef]
5. Bostwick DG, Meiers I. Prostate. In: Weidner N, Cote RJ, Suster S, Weiss LM, eds. Modern Surgical Pathology. Second Ed. Philadelphia: W.B. Saunders; 2009:1121-80. [CrossRef]
6. Dai HY, Guo SL, Shen J, Yang L. Pulmonary benign metastasizing leiomyoma: A case report and review of the literature. World J Clin Cases 2020; 8:3082-9. [CrossRef]
7. Bawaadam H, Ivanick N, AlShelli I, Krishna G. Endobronchial Leiomyoma: A case report with cryoprobe extraction and review of literature. Respir Med Case Rep 2021; 33:101467. [CrossRef]
8. Abu-Hishmeh M, Kobbari G, Shakil F, Epelbaum O. Primary Pulmonary Leiomyoma: A First for Peripheral Diagnostic Bronchoscopy? J Bronchology Interv Pulmonol 2020; 27:e54-e56. [CrossRef]
9. Öztürk A, Aktaş Z, Yılmaz A, Yeşildağlı H, Memiş L. A case of tracheal leiomyoma misdiagnosed as asthma. Tuberk Toraks 2016; 64:306-9. [CrossRef]
10. Stevic R, Milenkovic B. Tracheobronchial tumors. J Thorac Dis 2016; 8:3401-13. [CrossRef]
11. Cárdenas-García J, Lee-Chang A, Chung V, Shim C, Factor S, Tibb A. Bronchial leiomyoma, a case report and review of literature. Respir Med Case Rep 2014; 12:59-62. [CrossRef]
12. Pandey K, Vaidya PJ, Kate AH, Chavhan VB, Jaybhaye P, Patole K, et al. Bronchoscopic and surgical management of rare endobronchial tumors. J Cancer Res Ther 2016; 12:1093-7. [CrossRef]