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Case Report

Bizarre Presentation of Huge Primary Pulmonary Leiomyoma

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Abstract

Primary pulmonary leiomyoma is a rare condition that accounts for about 2% of all benign lung tumors. We report a 38-year-old male patient who presented with shortness of breath and cough. A computed tomography scan showed a right lung mass measuring 13 x 11 cm in diameter that appeared to originate from the pleura. Therefore, a challenging right postero-lateral thoracotomy with lung sparing lobectomy was done with a smooth recovery. To our knowledge, this case is considered the largest reported primary pulmonary leiomyoma according to size with atypical presentation.

Keywords: *Primary Pulmonary Leiomyoma; Huge Leiomyoma*

Introduction

Primary pulmonary leiomyoma is a rare condition that was first reported in 1910 [1]. It represents approximately 2% of all benign lung tumors [2]. The origin of pulmonary leiomyoma is either from the smooth muscle of the tracheobronchial tree or the blood vessels [3]. Common locations for primary pulmonary leiomyoma are: lung parenchyma (51%), bronchi (30%), and trachea (16%) [1].

According to the current literature, this is to be considered the largest described size of primary pulmonary leiomyoma with a peculiar presentation undergoing successful surgical treatment.

Case Report

A 38-year-old male patient, previous smoker, presented to our outpatient clinic complaining of dry cough and shortness of breath for two weeks. Past medical history was notable for bronchial asthma and allergic rhinitis diagnosed ten years ago. The review of other systems was unremarkable. General clinical workup was unremarkable and his physical examination was normal apart from decreased air entry over the right side. Furthermore, a routine chest x-ray followed by a chest-abdomen computed tomography (CT) scan with intravenous contrast was done showing a 13 x 11 cm well-defined solid mass of the right hemithorax suggesting pleural fibroma (Figure 1).

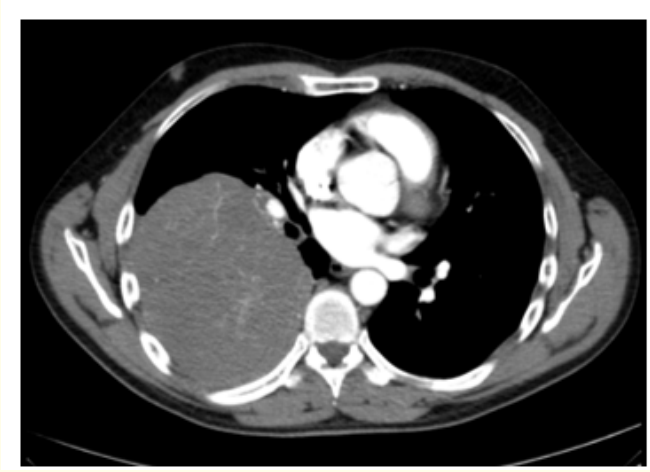

Figure 1: *Chest computed tomography scan showing a right mass in the right lung measuring 13 x 10 cm.*

The patient underwent a right postero-lateral thoracotomy with preserving serratus anterior muscle and fourth rib was excised due to the huge size of the tumor. Intraoperatively, there was a huge right parenchymal mass adherent to the upper lobe (Figure 2) compressing the main pulmonary artery and vein. Therefore, a challenging lung-preserving right upper lobectomy commenced after safely controlling major vessels. Mediastinal lymphadenectomy was done with right phrenic nerve infiltration of lidocaine to obliterate huge dead space.

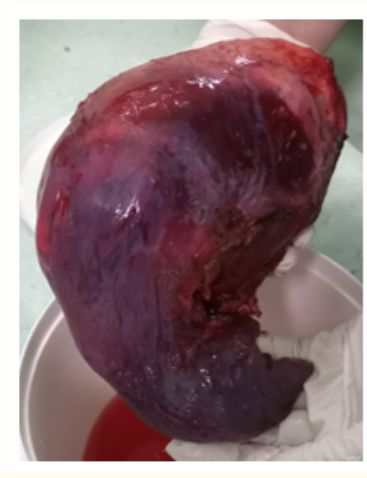

Figure 2: *A gross appearance of the tumor abutting the right upper lobe parenchyma.*

Histopathological studies showed; proliferation of bland spindle cells, and blunt-ended nuclei, mitotic activity 1/10 High Power Field and no pleomorphism or necrosis. Immunohistochemistry for SMA, desmin, S-100, CD34, CD99, TLE.1, ALK, HMB45 and Ki67- revealed

tumor cells were diffusely positive for desmin and negative to other markers. These features are most consistent with leiomyoma excluding other differential diagnosis (Figure 3). Post-operative bronchoscopy was done showing no other endobronchial tumors and the postoperative course was smooth.

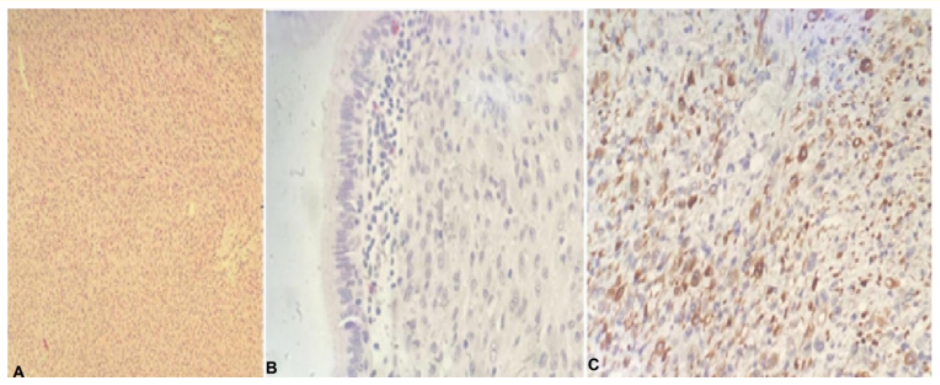

Figure 3: *(A) low power view, H&E stain shows a cellular proliferation of bland spindle cells arranged in fascicles. (B) high power resolution, showing endobronchial epithelium on top of the lesion, indicating origin from the bronchial wall. (C) Desmin immunostain shows diffuse positivity in tumor cells.*

Discussion

Primary pulmonary leiomyoma represents less than 2% of benign lung tumors [2]. They mostly occur in women in their third and fourth decades of life [5,6]. Leiomyoma may be asymptomatic and found incidentally on chest imaging [7,8]. However, endoluminal lesions may cause cough, wheeze, or dyspnea [9]. In many cases, symptoms of primary pulmonary leiomyoma mimic bronchial asthma and most patients would be labeled as asthmatic for many years [10]. Hemoptysis, atelectasis, consolidation, bronchiectasis, or post-obstructive pneumonia might be caused by larger tumors [4,5].

Lesions are discovered via imaging as nodules a few millimeters up to several centimeters (maximum 4.2 cm) in size [11]. However, our patient had triple the size of the largest reported primary pulmonary leiomyoma according to our literature review. Both solitary and multiple lesions are reported. Moreover, computed tomography scans show a homogeneously enhancing airway lesion with intraluminal growth forming an ''iceberg'' appearance (small intraluminal component with large extraluminal component) which explains the asymptomatic giant tumors as in our case [6].

The cornerstone of management of primary leiomyoma depends mainly on the location and size of the tumor [12]. Both bronchoscopic and surgical resection of tumors have been reported [12]. Segmental resections should be considered for symptomatic parenchymal lesions which cause complications such as bronchial compression or bronchiectasis [13]. In a series of 66 cases of operated bronchopulmonary leiomyoma; 17 were treated by pneumonectomy, 35 by lobectomy, two by segmentectomy. Operative procedures without lung resection were performed in 12 cases, of which sleeve bronchoplasty accounted for 8 and removal of the tumor through bronchotomy for 4 [12]. However, there is no current guideline for the extent of surgical resection nor for a conservative approach. Therefore, in our case lung sparing right upper lobectomy was done.

Conclusion

Pulmonary leiomyoma is a rare asymptomatic tumor. It might cause asthma-like symptoms such as cough, wheezing, or dyspnea. On imaging, primary pulmonary leiomyoma represents either endobronchial or lung parenchymal lesions or both. Surgical management mainly depends on the location, size, symptoms, and expertise. The surgical resection margin should be individualized according to the site and size of the tumor.

Conflict of Interest

None.

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