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A Rare Case of Giant Pulmonary Hamartoma

Yuanyuan Zong and Xichao Sun*

Case Report

Department of Pathology, Shandong Provincial Hospital Affiliated to Shandong University, Jinan 250021, PR China

*Corresponding author: Xichao Sun, Department of Pathology, Shandong Provincial Hospital Affiliated to Shandong University, Jinan 250021, PR China, Tel: +86-0531-88392413; E-mail: zong-yy@qq.com

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Abstract

We report a case of huge pulmonary hamartoma with atrophied right lower lobe in a 33-year-old female patient. The pulmonary hamartoma was solid, $30 \times 16.5 \times 14.5$ cm in size, which sectioned surface reveals a thick-walled fibrous cyst with multiple yellow nodules protruding into the lumen. The tumour was compressing the right lung and there was no evidence of infiltration into the surrounding structures. It was successfully treated by surgical resection and final histology was pulmonary hamartoma with predominantly fibrous tissue, cartilaginous tissue and leiomyomatous differentiation.

We believe this is the largest reported case in literatures yet, and this was successfully resected via a median sternotomy.

Introduction

Pulmonary hamartoma composed of an abnormal mixture of mesenchymal elements is the most common benign neoplasm in the lung. Parenchymal lesions are usually an incidental finding and range in size from 1 cm to 8 cm in diameter in various series [1].

Pulmonary hamartoma larger than 20 cm and the cystic variant are also very rare. Pulmonary hamartomas, was also known as mesenchymomas, located in pulmonary parenchymal (80%) or endobronchial (10–20%). The endobronchial tumours usually present with new onset respiratory symptoms most commonly recurrent chest infections or haemoptysis.

Case Report

Chest radiograph of a 33-year-old female patient showed solid mass within the right hemithorax (Figure 1A). Chest computed tomography reveals a huge solid mass containing foci of intralesional calcifications in the right lower lobe of the lung (Figure 1B). But the patient presented with no obvious respiratory symptom.

The patient underwent a median sternotomy whereupon a giant tumour was found arising superficially from the lower border of the right lung and occupying a half of the right chest and extending to the anterior mediastinum. The mass was compressing the right lung with no evidence of local invasion and also no connection with bronchus or vessel. The tumour was found to be adherent to the lower border of the lung and was easily dissected with sharp dissection. The mass had enclosed by a thick-walled fibrous cyst with multiple yellow nodules protruding into the lumen. The final histology of the tumour was a pulmonary hamartoma, measuring $30 \times 16.5 \times 14.5$ cm (Figure 2).

Microscopically, the solid components were composed of columnar epithelium, cartilage, fibrous, leiomyomatous and a little adipose tissue. Also seen were foci of calcification within the sclerotic stroma. The cystic lesions are associated with interstitial fibrous mesenchymal stroma (Figure 3).



Figure 1: Chest radiograph of a 33-year-old female patient showed solid mass within the right hemithorax (A). Chest computed tomography reveals a huge solid mass containing foci of intralesional calcifications in the right lower lobe of the lung (B).

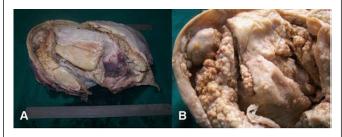


Figure 2: Photograph of the resected hamartoma (A). Gross photograph of solid mass enclosed by a thick-walled fibrous cyst, containing with numerous interstitial cartilaginous small nodules (B)

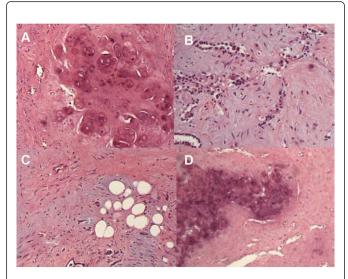


Figure 3: H&E stain, original magnification \times 400. Microscopically, the solid components were composed of cartilage (A), columnar epithelium (B) and a little leiomyomatous and adipose tissue(C). Also seen were foci of calcification within the sclerotic stroma (D).

The patient recovered now and there was no evidence of recurrence for two years after the operation.

Discussion

Depending upon the predominant component, hamartomas can be subdivided into various subtypes; chondromatous, leiomyomatous, lymphangiomyomatous, adenofibromatous and fibroleiomyomatous. Chondromatous hamartomas are the most common subtype and have been divided into endobronchial and intraparenchymal (peripheral) lesions. The onset of the tumor is in adulthood, with the peak age incidence in the sixth decade. Hamartomas may range from 1 to larger than 10 cm in the greatest dimension, but usually are smaller than 4 cm. Up to now, a case of giant pulmonary hamartochondroma 29 cm in diameter has been reported [2].

Hamartomas are frequently discovered on routine chest radiography, cystic ones are very rare. Our case had no bronchial connection to cystic area. Cystic mesenchymal hamartoma refers mainly to neoplasms of children [3,4].

Most notable are mutations in the hamartomas of high-mobilit group (HMG) proteins, a family of non-histone, chromatin-associated proteins, which are important in regulating chromatin architecture and gene expression. Mutations in the regions 6p21 and 12q14-15 are most commonly found [5-7].

In this case, the huge tumour was found to be adherent to the lower border of the lung with no evidence of local invasion and also no connection with bronchus or vessel, but this huge tumor made thoracic surgeons mistake it for malignant tumor and resected right lower lobe. So we believed this report can give thoracic surgeons and young pathologists some useful information.

Conclusion

The hamartoma is composed of cystic nodules with lining of respiratory epithelium and firm cartilagenous tissue without differentiation diagnosis. We believe this is the largest reported case in literatures yet, and this was successfully resected.

The patient recovered now and there was no evidence of recurrence for two years after the operation.

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