Journal of Pulmonary Medicine

Research Article A SCITECHNOL JOURNAL

Unilateral Giant Lung Bulla: Placental transmogrification should be in Mind

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Abstract

Placental transmogrification is a peculiar clinical entity of the lung of uncertain etiology. We report two cases of pulmonary placental transmogrification in two patients of different nationalities. Both of them have no history of smoking or chronic lung disease. The main presentations were dyspnea and chest pain. Radiologic studies showed unilateral giant bulla in both patients; additional pneumothorax was present in only one patient. They were subjected to surgical bullectomy. Histopathologic studies revealed the presence of intracystic placenta like villous structures and a diagnosis of placental transmogrification was made. Placental transmogrification should be considered in cases of unilateral bullae.

Keywords: Emphysema; Lung bulla; Placenta transmogrification.

Introduction

Bullous emphysema is a form of emphysema characterized by the presence of bullae. The main predisposing factors for its occurrence are tobacco smoking and α1-antitrypsin deficiency. In advanced cases the pathologic process is usually diffuse and affects both lungs. However, bullae may occur in lungs that are otherwise normal [1].

Unilateral bullae occupying most of the hemithorax are seldom seen. In 1979, Mc Chesney described a rare congenital form of giant bullous emphysema; he coined the term pulmonary placental transmogrification (PT) because of the morphological resemblance of intracystic papillary or villous structures to immature placental chorionic villi. In fact, PT has no biological or biochemical relations to placenta [2].

Since that time several case records were reported with variable clinical and radiological presentations. Meanwhile pathologists tried to postulate explanations to the pathogenesis of this lesion.

Case Report

We present 2 cases of PT of the lung. Data were collected from patients' files; the study was approved by hospital review committee. The patients were nonsmokers and have no history of chronic chest disease.

Case 1

A 25-year-old African female presented with acute onset chest pain and shortness of breath. Chest radiography gave impression of loculated pneumothorax. Computed tomography of the chest verified the picture as right side pneumothorax, right lower lobe large bulla (Figure 1a) and multiple upper lobe variable sized bullae. Chest drain was inserted and on a later date the patient underwent surgical resection of the lower lobe bulla (Figure 1b), ablation of upper lobe bullae and mechanical pleural abrasions. We started with VATS then converted to small muscle sparing lateral thoracotomy with the assistance of thoracoscopy camera due to marked adhesions of the upper lobe. She is fine 1 year after surgery.

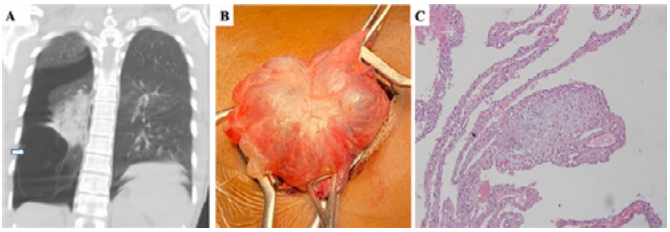

Figure 1: a) The picture as right side pneumothorax, right lower lobe large bulla. b) The patient underwent surgical resection of the lower lobe bulla. c) Diagnosis of placental transmogrification of the lung

Case 2

A 28-year-old Jordanian female presented with right side vague chest pain and shortness of breath of two weeks duration. She has average body built; vital signs and oxygen saturation were within normal ranges. Chest x-ray was initially interpreted as right side pneumothorax but computed tomography of the chest gave clue to diagnosis of right side giant bulla with compression effect on the lung and shift of mediastinum (Figure 2a and 2 b). Patient underwent right side VATS bullectomy (Figure 2c) and mechanical pleural abrasions. She is doing well 8 months after surgery.

Histopathologic examination of the resected bullae of both patients showed intracystic proliferation of papillary structures; the papillary cores contain congested blood vessels and surrounded by hyperplastic alveolar pneumocytes; a diagnosis of placental transmogrification of the lung was made (Figure 1c and Figure 2d).

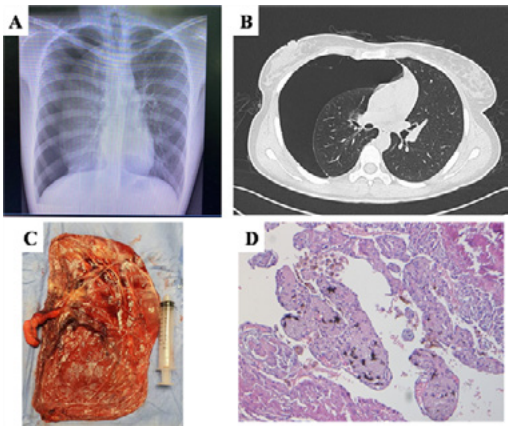

Figure 2: a,b) Compression effect on the lung and shift of mediastinum. c) Patient underwent right side VATS bullectomy. d) Diagnosis of placental transmogrification of the lung

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Received: 16 September, 2020; **Accepted Date:** 30 September, 2020; **Published Date:** 07 October, 2020

Discussion

Placental transmogrification is a rare benign lung malformation. Actually, Less than 40 cases were reported in English literature. In this short report we present two cases diagnosed in a single center within a period of 1 year. This raises some issues about the actual prevalence of the condition. It is possible that the diagnosis is missed or ignored in some cases. Also, reluctancy in publication of new cases could be another cause of the low prevalence of the condition. We encourage good communications between the surgeon and the pathologist when the clinical diagnosis of PT is suspected.

In his first report, Mc Chesney considered PT as an unrecognized hamartoma [2]. After all these years the exact etiology of PT is still controversial. In view of the frequent occurrence of PT with emphysema, some authors consider it as a variant or a complication of bullous emphysema [3]. Whereas, Xu et al [4]; confirmed the presence of PT in 6 cases of pulmonary fibrochondromatous hamartoma, and advocated PT as a hamartomatous lesion in agreement with Mc Chesney [2]. On the contrary, Cavazza et al using immunohistochemical, ultrastructural and molecular studies, identified genotypic alterations in the clear cell component of PT, but not in the adjacent normal tissue; accordingly, they attributed PT to benign proliferation of immature interstitial clear cells with secondary cystic changes, rather than being a variant of emphysema [5]. Another proposal was introduced by Yang et al; they reported PT in a newly developed pulmonary nodule. Accordingly, they consider PT as benign morphologic changes that may be encountered in both congenital and acquired lesions rather than being an independent disease [6].

Clinically, the disease is more common in middle aged men. Patients can be asymptomatic or may present with dyspnea, chest pain, pneumothorax (possibly tension pneumothorax), hemoptysis or a combination of all of the above. In very rare instances the condition may be present in association with lung cancer [7,8].

The radiologic studies may show unilateral bullous lesions, cystic changes with or without associated mass, or non solid lung nodule containing several small round-shaped air spaces [9].

In some cases the differentiation between giant bulla and pneumothorax in chest radiograph can be difficult; in this situation chest CT is useful and gives important aid to diagnosis. Waitches et al suggested that the presence of air outlining both sides of the bulla wall which is parallel to the chest wall gives a clue to diagnosis of simultaneous presence of both pneumothorax and bulla; they called this as the double-wall sign. The absence of double-wall sign provides confidence against the diagnosis of pneumothorax. One should be careful when two large bullae are adjacent to one another producing an apparent double-wall sign; in this situation the bulla wall will not be parallel to the chest wall [10].

Criteria for surgical resection of bullae are generally determined by degree of dyspnea. However, resection is also advised for asymptomatic patient with bulla occupying more than one third of the hemithorax to avoid potential complications. During surgery, resection of the pathologic lesion with preservation of healthy lung tissue is recommended.

Conclusion

Placental transmogrification should be considered in the diagnosis of patients with large unilateral bullae without traditional risk factors for emphysema. Chest CT should be done before chest drain insertion in stable patient when differentiation between Lung bulla and pneumothorax is not clear. Surgical resection with preservation of normal lung tissue is recommended and seems to be curative.

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