Placental transmogrification of the lung associated with unilateral pleural effusion: A case report with a comprehensive review of the literature

N. Narula  
_Northwell Health, nnarula@northwell.edu_

S. Ngu  
_Northwell Health, sngu@northwell.edu_

D. Sharma  
_Northwell Health, dsharma2@northwell.edu_

F. Siddiqui

M. Chalhoub  
_Zucker School of Medicine at Hofstra/Northwell, mchalhoub1@northwell.edu_

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Placental transmogrification of the lung associated with unilateral pleural effusion: A case report with a comprehensive review of the literature

Naureen Narula\textsuperscript{a,∗}, Sam Ngu\textsuperscript{b}, Dikshya Sharma\textsuperscript{b}, Faraz Siddiqui\textsuperscript{b}, Michel Chalhoub\textsuperscript{b}

\textsuperscript{a} Internal Medicine, Staten Island University Hospital, Staten Island, NY, 10305, USA
\textsuperscript{b} Pulmonary and Critical Care, Staten Island University Hospital, Staten Island, NY, 10305, USA

ABSTRACT

Placental transmogrification of the lung (PTL) is a rare benign pulmonary lesion resembling chorionic villi. With fewer than 40 cases reported in literature, associations have thus far been made with bullous emphysema, pulmonary fibrochondromatous hamartomas and adenocarcinoma of the lung. Typically presenting as unilateral solitary cystic or bullous lesion, we report the first case of PTL presenting with unilateral pleural effusion. A 70-year-old male presented with recurrent unilateral pleural effusion that failed to resolve with multiple thoracenteses. He underwent thoracoscopic excision and biopsy of a cystic mass identified on computed tomography (CT) scan which revealed characteristic villous and papillary changes. We describe the case and review the literature on this benign but rare pulmonary disease entity.

1. Introduction

Placental transmogrification of the lung (PTL) is a rare benign cystic or bullous pulmonary lesion morphologically resembling placental tissue. However, it is not functionally related to placental tissue. First described by McChesney in 1979, it is a histologic diagnosis and often incidentally found. It has thus far been associated with bullous emphysema, pulmonary fibrochondromatous hamartomas and adenocarcinoma of the lung [1]. Radiographically, the majority of reported cases of PTL describe bullous emphysematous changes. The etiology of these lesions remain unclear; however, proposed mechanisms range from lymphovascular proliferation in the setting of an emphysematous lung to congenital malformation. A study analyzing immunohistochemical profiles proposed that dysregulated growth of respiratory lining epithelium of fibrochondromatous hamartomas results in the placental villi-like structures [5]. Although typically asymptomatic, they can present as cough, chest pain, dyspnea, pneumothorax and emphysema. In this report, we describe the first report of PTL presenting as recurrent right-sided pleural effusion in a 70-year-old male. The diagnosis was confirmed by histopathological examination of the thoracoscopically-excised cystic mass.

2. Case report

A 70-year-old male presented to the emergency department (ED) with progressively worsening shortness of breath over 2 months. He has a past medical history significant for essential hypertension, diabetes mellitus type 2, coronary artery disease and obstructive sleep apnea. He is an active smoker with 30-pack-year. On examination, patient had decreased breath sounds on the right. He otherwise denied fever, chest pain, palpitations, cough, sputum production, orthopnea, nocturnal dyspnea, swelling of the lower extremities or weight change. Blood work showed a normal blood count, comprehensive metabolic panel, cardiac biomarkers, brain natriuretic peptide and D-dimer. A chest x-ray (Fig. 1) showed moderate unilateral right-sided pleural effusion. Lower extremity doppler was negative for deep venous thrombosis in bilateral lower extremities.

A right-sided thoracentesis was performed, draining 800 cc of amber-colored fluid. Pleural fluid analysis showed an exudative effusion with lymphocytic predominance and high amylase and lactic acid dehydrogenase (LDH) levels. No malignant cells were seen. A post-thoracentesis chest x-ray (Fig. 2) showed decreased size of the effusion on right side.

He was subsequently discharged home the following day with outpatient pulmonary follow-up in 1 week. Outpatient chest radiograph performed 1 week later revealed improved right-sided pleural effusion. Computed tomography (CT) chest, abdomen and pelvis with contrast were ordered along with rheumatologic workup for further characterization of effusion. CT chest (Fig. 3) showed a loculated small right-sided pleural effusion with rounded atelectasis of the right middle lobe. No suspicious intrathoracic mass or lymphadenopathy was noted. Basic rheumatologic screen was negative. As the patient was asymptomatic at the time, he refused further diagnostic work-up. The pleural effusion eventually...
completely resolved within 5 months of follow-up.

However, the patient returned to ED six months post-discharge with similar complaint of shortness of breath. Chest radiograph showed worsening right-sided pleural effusion. A repeat CT scan (Fig. 4) of the chest showed a 13 × 7 × 7 cm located right-sided pleural effusion with right lower lobe atelectasis. A CT-guided thoracentesis with drainage of 1200 cc of pleural fluid. Analysis again supported exudative fluid with lymphocytic predominance, without evidence of infection or malignancy.

Positron emission tomography (PET) scan showed no fluorodeoxyglucose (FDG) uptake. He underwent a thoracoscopic excision of the mass to further characterize the etiology. A fluid-filled cystic structure adhered to the chest wall, diaphragm, and pericardium was found and subsequently removed in its entirety with symptomatic resolution of the pleural effusion. Gross examination of the mass revealed an 18 × 12 cm cyst with multiple fragments of fibromembranous tissue. Microscopically, prominent epithelial folding were present in a stroma of myxoid fibroadipose tissue forming papillary projections resembling chorionic villi. Post discharge from hospital, patient was followed upto 6 months in the office with no recurrence of pleural effusion (see Figs. 5 and 6).
3. Discussion

Placental transmogrification of the lung (PTL) is a rare benign disease with striking placentoid morphological features. There are papillary projections with minimal inflammatory infiltrate and absent pulmonary fibrosis [1]. It predominantly affects men between 24 and 44 years of age. First described histologically in 1979, it is generally an incidental finding with a large number of cases being asymptomatic. Clinically, it can lead to dyspnea, cough and hemoptysis. PTL has also been reported to present as tension pneumothorax, lung nodules and mediastinal mass. Fewer than 40 cases have thus far been reported in literature. It has been associated with fibrochondromatous hamartomas and pulmonary lipomatosis [2,7]. There is not enough data available to make an association with smoking. Available reports do not appear to show predilection of laterality or lobe.

Radiological findings include bullous emphysema, mixed cystic lesion and nodule and rare solitary nodule. Unilateral bullous lesion have been reported as the most common presentation of PTL. It is differentiated from bullous emphysema by its unilateral origin in a relatively younger population with no history of smoking or alpha-1 antitrypsin deficiency. Diagnosis can be confirmed by gross and microscopic examination which characteristically shows cystic structure with myxoid, lipomatous and edematous changes bordered by low-columnar respiratory epithelium mimicking placental villi [1,3]. However, a high degree of clinical suspicion is required for diagnosis.

The etiology of these lesions remain unclear; however, proposed mechanisms range from lymphovascular proliferation in the setting of an emphysematous lung to congenital malformation. A study analyzing immunohistochemical profiles proposed that dysregulated growth of respiratory lining epithelium of fibrochondromatous hamartomas results in the placental villi-like structures [5]. Other authors proposed that PTL arises from lipomatosis due to expression of fat tissues inside the villi [4]. Still others described interstitial clear cell proliferation as the primary inciting event followed by emphysematous changes as a secondary phenomenon; however, this theory is limited by only 2 reported cases [6].

Horsley et al. recommended considering PTL as an important differential in patients with unilateral bullous lesion, specifically in

Fig. 4. CT scan of the chest with contrast showing 13 × 7 × 7 cm loculated right-sided pleural effusion (arrow).

Fig. 5. Villus-like structures admixed with remnants of alveolar walls, with core of structures being filled by mature adipose tissue.

Fig. 6. Hematoxylin-eosin–stained slide of the cystic mass showing pulmonary villus papillary projections in a myxoid stroma.
circumstances where emphysema is unlikely [6]. Surgical excision of the cystic mass is usually curative, even in patients with severe pulmonary symptoms. Our case highlights the rare presentation of PTL associated with unilateral pleural effusion.

4. Conclusion

Our case highlights the first reported case of PTL presenting with recurrent unilateral pleural effusion. Although PTL is thought to be a rare and benign pulmonary condition, clinicians should promptly diagnose and treat the condition as it has the propensity for worse outcomes if untreated. Surgical excision have both diagnostic and therapeutic advantages.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.rmcr.2018.11.018.

References