Placental transmogrification of the lung presenting as tension pneumothorax: Case report with review of literature

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Supplemental material is available online.

An unusual case of localized giant bullous emphysema with placental histologic features (also known as placental transmogrification of lung) was seen in a patient with bullae admitted for a tension pneumothorax. This report evaluates the clinical, pathologic, and surgical aspects of this unusual histologic subtype of bullous emphysema. Thus far, 12 case reports documenting 23 cases have been identified in the English literature.

Clinical Summary

A 45-year-old Mexican-American man in moderate respiratory distress presented to the emergency department with a vague 1-week history of chest pain and 1-day history of acute onset severe dyspnea. He denied any history of recent trauma, history of smoking, or chemical exposure.

Physical examination revealed decreased breath sounds on the right side, and a chest x-ray was initially interpreted as a tension pneumothorax (Figure 1). Urgent needle decompression was performed, followed by placement of a right chest tube, with only minimal relief of symptoms. A repeat chest x-ray demonstrated relief of mediastinal shift but a lucency consistent with a collapsed lung. A chest computed tomography scan revealed giant bullous disease with a collapsed right lung (Figure 2). No pneumothorax was identified. The left lung was normal on radiologic and physical examination. These findings were suggestive of unilateral bullous disease. No lymphadenopathy or mass lesions were identified. Bronchoscopy demonstrated patency of the airways and no lesions. Bronchial washings were also negative.

The patient was taken to the operating room where he underwent a standard right thoracotomy. A non-anatomic resection of the bullae from all 3 lobes was performed. The remainder of the right lung was grossly normal and readily expanded.

Pathologic evaluation of the tissue showed spongy red lung tissue merging with large bullous cysts. Histologically, the bullous spaces were contiguous with areas that had papillary structures that looked like placental villi (Figure 3). These papillary structures were edematous and had vascular channels. Lymphoid aggregates and follicles were seen in some papillary structures. These findings were consistent with a diagnosis of localized giant bullous emphysema with placental histologic features, also known as placental transmogrification of the lung.

The patient had an uneventful postoperative course. He had total reexpansion of his right lung and was discharged without complications.

Discussion

An extensive review of the English literature was performed that identified 12 case reports describing 23 similar cases.¹⁻¹² This unique pathologic feature has been described as placental bullous lesion of the lung or placental transmogrification of the lung.² The disease was found most commonly in men, usually in their second to fifth decades of life. The pathogenesis of this lesion is still unknown, although there are theories that support carcinogenic or congenital origins.^{3,4}

Horsley and colleagues⁵ concluded that “placental transmogrification of the lung should be in the differential diagnosis in any patient who presents with predominately unilateral bullous disease, especially in those without other risk factors.”

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Figure 1. Chest radiograph at time of presentation demonstrating large right pneumothorax with mediastinal shift and tracheal deviation to the left.

This case is unique because our patient presented with a clinical picture of tension pneumothorax not seen in any of the previously reported cases. Another distinguishing feature is that our treatment differed from the other cases documented in the literature. Traditionally, anatomic resections, including lobectomy and pneumonectomy, have been described for treatment. As Brevetti and colleagues⁶ described, we were able to resect the diseased component with minimal loss of normal lung parenchyma. Our results are consistent with those of Brevetti and colleagues in that our patient had a satisfactory outcome with lung preservation surgery.

Figure 2. Computed tomography of the chest revealing bullous disease of the right lung and abnormal tissue at the right base with a density not consistent with lung tissue.

Figure 3. High-power view of villous-like structure with vascular channels, lymphocytes, and edema (400X).

Conclusions

This rare disease of unclear origin has been encountered in the face of localized bullous emphysema. Resection of the lesion seems to be curative in the reported cases. We recommend that placental transmogrification be included in the differential diagnosis in patients without traditional risk factors who present with significant unilateral bullous emphysema and as a precursor for development of spontaneous pneumothorax. As previous authors have suggested, we believe that these lesions are best treated by minimal resection, leaving as much normal lung tissue and avoiding a lobectomy if possible.

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Surgical correction of postpneumonectomy-like syndrome in a patient with a tuberculosis-destroyed lung

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Postpneumonectomy syndrome is a rare condition that is characterized by dyspnea resulting from an extreme mediastinal shift and bronchial compression of the residual lung after right or left pneumonectomy.¹⁻³ Severe fibrosis of the lung such as is seen in a lung destroyed by tuberculosis (TB) can cause similar clinical features in the absence of pneumonectomy.⁴

We report here a couple of unique cases of postpneumonectomy syndrome without pneumonectomy in which treatment via pneumonectomy and mediastinal repositioning with tissue expanders was successful.

Clinical Summaries

PATIENT 1. A 47-year-old woman had had severe progressive dyspnea for 6 months. She had had pulmonary TB 20 years earlier, which was completely cured by administration of anti-TB drugs. On physical examination, breath sounds were decreased in the left lung field. A simple chest radiograph and computed tomogram (Figure 1, A) revealed that the left lung was almost completely destroyed and the right main bronchus was compressed by the right main pulmonary artery and the vertebral body.

The operation was performed via a left posterolateral thoracotomy. The severe pleural adhesion was dissected, and then left pneumonectomy was performed followed by insertion of two types of tissue expanders into the pleural cavity. One expander was ellipsoid (350 mL) and the other was crescent shaped (250 mL). After insertion of the tissue expanders, normal saline that included antibiotics

was injected into the tissue expanders, and the thoracotomy wound was closed.

Postoperatively, she recovered without any problem. A plain chest radiograph showed that the mediastinum had returned to a normal position (Figure 1, B). In addition, the expiratory wheezing and dyspnea disappeared.

The patient was discharged from the hospital on the 13th postoperative day. At present, 4 years after the operation, she is being observed at the outpatient department and is without dyspnea.

PATIENT 2. A 50-year-old woman had had severe progressive exertional dyspnea for the previous 6 months. She had had pulmonary TB 30 years earlier, which was completely cured by anti-TB medication. Six months before her current admission, the right lung totally collapsed and she had paroxysmal severe exertional dyspnea, which led her to visit our hospital's emergency room. On physical examination, there was no audible breath sound in the right side of the chest. Chest computed tomography revealed a totally collapsed right lung, extreme deviation of the mediastinal structure, and a narrowed left main bronchus, which was compressed between the left main pulmonary artery and the descending aorta (Figure 2, A). We performed a right pneumonectomy via a posterolateral thoracotomy. There was no pleural adhesion. Two types of tissue expanders were inserted, with 320 mL of normal saline being infused into the ellipsoid one and 450 mL into the crescent-shaped one.

Postoperatively, the mediastinum returned to the normal position (Figure 2, B). The patient was discharged on the seventh postoperative day. Five months after the operation, she remains free of expiratory stridor and dyspnea on exertion.

Discussion

Postpneumonectomy syndrome is a rare complication that is characterized by tracheal stenosis and severe dyspnea; these symptoms are caused by mediastinal shifting after pneumonectomy. In the cases we presented, the right or left main bronchus was compressed between the main pulmonary artery and vertebra/aorta not by surgical pneumonectomy, but by auto-pneumonectomy that was due to TB. These 2 cases show that surgical pneumonectomy is not a prerequisite for postpneumonectomy syndrome because this malady occurs as a result of stenosis of the bronchus after vigorous movement and rotation of the mediastinum.

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Figure E1. Chest radiograph at time of presentation demonstrating large right pneumothorax with mediastinal shift and tracheal deviation to the left.

Figure E2. Chest radiograph after placement of tube thoracostomy demonstrating resolution of the shift, incomplete expansion of the right lung and a density in the right lower lung field.

Figure E3. Computed tomography of the chest revealing bullous disease of the right lung and abnormal tissue at the right base with a density not consistent with lung tissue.

Figure E4. Computed tomography of the chest revealing bullous disease of the right lung and abnormal tissue at the right base with a density not consistent with lung tissue.

Figure E5. Computed tomography of the chest revealing bullous disease of the right lung and abnormal tissue at the right base with a density not consistent with lung tissue.

Figure E6. Chest radiograph at discharge showing complete re-expansion of the right lung with normal lung fields.

Figure E7. Low power photomicrograph of lung specimen showing villous-like changes in the region of bullae-placental transmogrification (100 $\times$).

Figure E8. High power view of one villous-like structure with vascular channels, lymphocytes and edema (400 $\times$).