Left lung hypoplasia with a right tuberculous pleural effusion after childbirth
A case report

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Abstract
Rationale: Unilateral hypoplasia of the lung is a rare congenital condition, the mechanism of which is poorly understood. Primary pulmonary hypoplasia occurring in an adult is extremely rare and we present what is probably the first case of a link to a tuberculous pleural effusion in a young woman after childbirth.

Patient concerns: Herein, we describe a 31-year-old woman with left lung hypoplasia, and she not only survived to adulthood without problems, but was able to deliver a baby in natural labor.

Diagnoses: Left lung hypoplasia, right tuberculous pleural effusion.

Interventions: We initiated an anti-tuberculosis treatment for this patient with dose adjustments to her weight of isoniazid (0.3 g/day), rifampicin (0.45 g/day), pyrazinamide (1.5 g/day), and ethambutol (0.75 g/day) for 2 months then isoniazid and rifampicin for another 4 months.

Outcomes: Ten days later after beginning therapy, she became afebrile and the pleural effusion resolved. No recurrence was observed during a 6-month follow-up period.

Lessons: In clinical practice, if one sees a chest x-ray revealing complete or incomplete opacification of a hemithorax with volume loss and history of repeated respiratory infections, one should consider the possibility of unilateral pulmonary hypoplasia. In such cases, regular close follow-up is important to minimize infections and to prevent development of cor pulmonale or respiratory failure.

Abbreviations: DLCO = carbon monoxide diffusing capacity, ESR = erythrocyte sedimentation rate, FEV1 = forced expiratory volume in one second, FVC = forced vital capacity.

Keywords: after childbirth, computed tomography, lung hypoplasia, tuberculous pleural effusion

1. Introduction
Unilateral hypoplasia of the lung is a rare congenital condition, the mechanism of which is poorly understood. Primary pulmonary hypoplasia occurring in an adult is extremely rare and we present what is probably the first case of a link to a tuberculous pleural effusion in a young woman after childbirth. Commonly patients usually present in childhood with respiratory distress or recurrent infection or haemoptysis and very rarely in adulthood. Our patient not only survived to adulthood without problems, but was able to deliver a baby in natural labor. Owing to its rarity, we hereby describe this case.
nase 58 U/L, lactate dehydrogenase 1020 U/L. Pleural fluid for acid-fast bacilli was negative. An echocardiogram showed 3 tricuspid regurgitation velocity of 3.1 m/s and a pulmonary artery systolic pressure of 44 mmHg. Pulmonary function testing demonstrated a forced expiratory volume in one second (FEV1) of 1.21 L, a FEV1% predicted of 36.2%, a forced vital capacity (FVC) of 1.8 L, a FVC % predicted of 47.2%, a FEV1/FVC of 66.8%, a carbon monoxide diffusing capacity (DLCO) of 5.18 mmol/min/kPa, a DLCO % predicted of 53.6%, a carbon monoxide diffusing capacity/alveolar ventilation (DLCO/VA) of 1.86 mmol/min/kPa/L, a DLCO/VA % predicted of 104.9%, a VA of 2.78 L, a VA % predicted of 52.6%. A contrast-enhanced chest CT scan demonstrated a markedly smaller left pulmonary artery and pulmonary veins, a mediastinal shift to the left and decreased size of the left hemithorax with herniation of the right lung (Fig. 2). The 3-dimensional (3D) reconstruction (minimum intensity projection) with spiral CT showed the left main bronchus was thin and short and the lobar bronchi appeared reduced in caliber (Fig. 3). A 3D volume-rendered image demonstrated an enlarged right main pulmonary artery, a left main pulmonary artery that was obviously smaller than right one, and compensatory increase of right lung vessels (Fig. 3).

We initiated an anti-tuberculosis (TB) treatment for this patient with dose adjustments to her weight of isoniazid (0.3 g/day), rifampicin (0.45 g/day), pyrazinamide (1.5 g/day), and ethambutol (0.75 g/day) for 2 months then isoniazid and rifampicin for another 4 months. Ten days later after beginning therapy, she became afebrile and the pleural effusion resolved. No recurrence was observed during a 6-month follow-up period.

3. Discussion

Schneider and Schwalbe[1] divided lung dysplasia into 3 types in 1912 and Boyden[2] modified in 1955. The classifications are as follows:

Type 1 agenesis: One or two lungs are completely absent, with no traces of bronchial or vascular supply or parenchymal tissue.
Type 2 aplasia: There is a basic bronchus that ends with the blind pouch, but with no evidence of pulmonary vasculature or parenchyma.
Type 3 hypoplasia: There is a certain amount of lung parenchyma, but the number or size of airways, blood vessels, and alveoli reduced.

After that, Monaldi[3] divided lung dysplasia into 4 groups, which are:

Group 1: Only the trachea.
Group 2: Only the basic main bronchi.
Group 3: The main bronchus aplasia after division.
Group 4: Subsegmental bronchi and corresponding lobe segments hypoplasia.
Chest x-ray, CT pulmonary angiography, and 3D volume-rendered image confirm the diagnosis in our case and according to the guidelines, our patient had primary left lung hypoplasia of Schneider Type 2 (Aplasia) or Monaldi group 3. The abnormal structure of the lung increases the chances of infection by common bacteria as well as opportunistic pathogens such as *Mycobacterium tuberculosis*. A hypoplastic lung is often a crippled lung that may not have fully intact local host defense mechanisms and thus will be much more prone to infections.\[^4\]\] Pregnancy also increases the chance of RB infection. At any one time 216,500 pregnant women have active TB in the world,\[^3\]\] which can lead to increased adverse maternal and fetal outcomes.\[^6\]\] In human, cell-mediated immunity by T\(\text{H1}\) lymphocytes \[^7\]\) protects against TB. In pregnancy, however, cell immunity transfer to antibody immunity (T\(\text{H2}\)) \[^8,9\]\] may lead latent TB to reactivate and more easily progress to active disease. For treatment, adult patients with hypoplasia are treated normally with antibiotics for infections, bronchodilators and apophlegmatisant. If the diagnosis of TB is clear, standard anti-TB treatment should be carried out as quickly as possible. To reduce the risk of infection, prophylactic pneumococcal and influenza vaccinations are recommended, especially in the winter and spring.

4. Conclusion
In clinical practice, if one sees a chest x-ray revealing complete or incomplete opacification of a hemithorax with volume loss and history of repeated respiratory infections, one should consider the possibility of unilateral pulmonary hypoplasia. In such cases, regular close follow-up is important to minimize infections and to prevent development of cor pulmonale or respiratory failure.

Figure 2. Computed tomography pulmonary angiogram. The left pulmonary artery is very small as are the ipsilateral pulmonary veins. There is mediastinal shift to the left and a decreased size of the left hemithorax with herniation of the right lung.

Figure 3. The 3D reconstruction (minimum intensity projection) with spiral computed tomography. Left main bronchus is thin and short and the lobar bronchi appeared reduced in caliber. 3D volume-rendered image. The left main pulmonary artery is obviously smaller than enlarged right main pulmonary artery and its hypertrophied and enlarged next several branching generations.
**Author contributions**

All authors diagnosed this disease and collected data, Shan Lin wrote the draft of this article, Wei Guan revised this article. Written consent to publication was obtained.

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