Clinical Case Report

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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, FEV_1 = 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.

Editor: N/A.

Compliance with Ethical Standards

The authors report no conflicts of interests.

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2. Case presentation

A 31-year-old woman, nonsmoker, was admitted with the chief complaint of dry cough and dyspnea for five months, fever accompanied with right-sided chest pain for 1 month. In this same time period, because she was midway through a pregnancy she could not undergo any radiological imaging. She delivered a healthy child by natural labor. One month later, her dyspnea worsened and she began to experience fever and right-sided chest pain. The fever as high as 39°C often occurred in the afternoon or at night. She noted night sweats and had lost 10 kg in weight over the month. Examination revealed that she was febrile (axillary temperature 38.8°C) with an oxygen saturation of 89% by pulse oximetry. In the left thorax, we could not hear breath sounds and the cardiac apex was felt in the left seventh intercostal mid-axillary line. Her heart rate was 120 beats per minute, but heart sounds were normal and cardiac souffle was not heard. She had dullness to percussion below the right eighth rib infrascapular line. Chest computed tomography (CT) showed mediastinal displacement to the left side with a cut off the left main bronchus and a right-sided pleural effusion (Fig. 1). Routine blood work was within the normal limits including a white blood cell or 5.32×10^9 cells/L. Other biochemical tests showed: erythrocyte sedimentation rate (ESR) 34 mm/h, C-reactive protein 40.6 mg/L, a positive interferon gamma release assay, and a positive purified protein derivative skin test of 10 mm of induration. A right-sided thoracentesis was performed, which centesis to the right chest and the pleural fluid analysis showed a lymphocytic predominant exudative effusion with an albumin 27.1 g/L, adenosine deami-

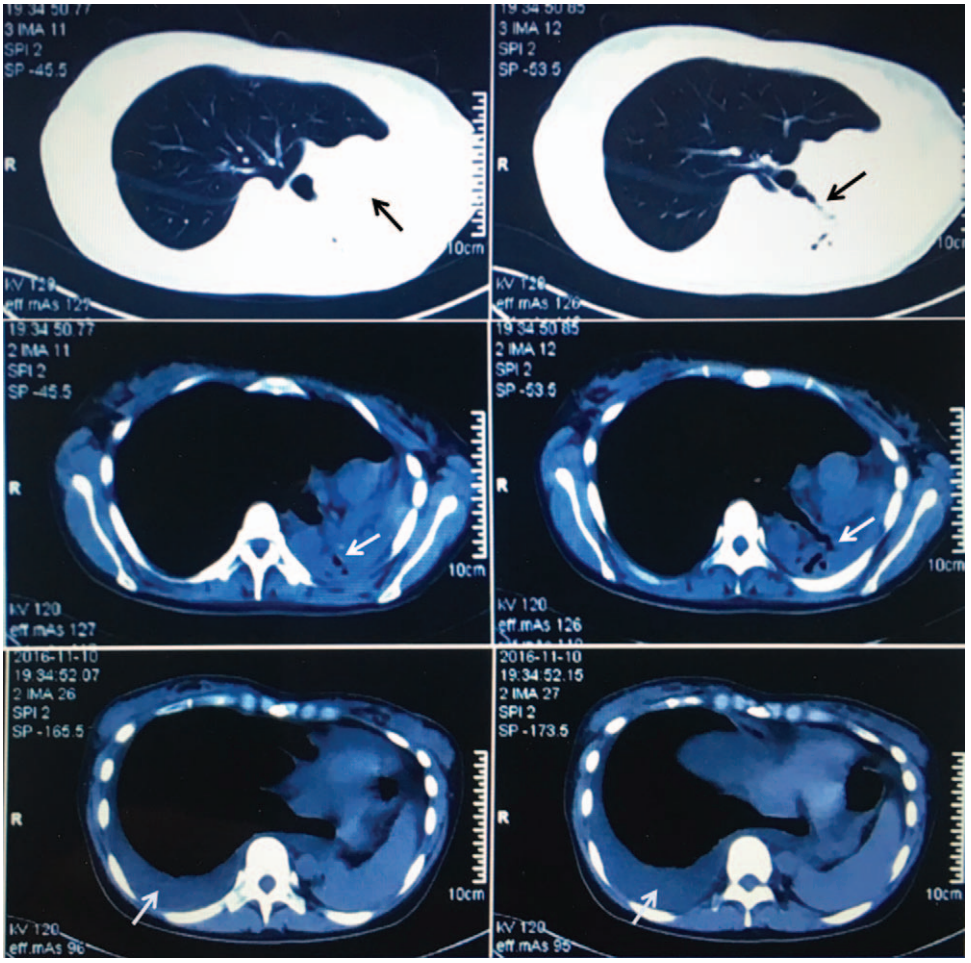

Figure 1. Chest computed tomography. Left main bronchus was cut-off, there was mediastinal displacement to the left side and a right-sided pleural effusion.

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 (FEV_1) of 1.21 L, a FEV_1% predicted of 36.2%, a forced vital capacity (FVC) of 1.8 L, a FVC % predicted of 47.2%, a FEV_1/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.

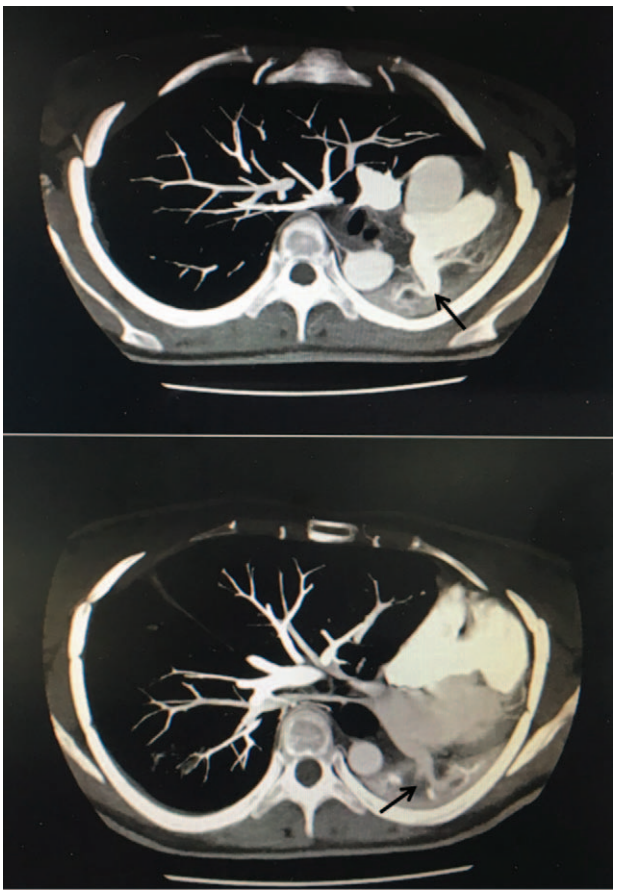

Figure 2. Computed tomography pulmonary angiogram. The left pulmonary artery is very small as are the ipsilateral pulmonary veins. There is mediastinal shift to the left and a decreased size of the left hemithorax with herniation of the right lung.

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,[5] which can lead to increased adverse maternal and fetal outcomes.[6] In human, cell-mediated immunity by TH1 lymphocytes [7] protects against TB. In pregnancy, however, cell immunity transfer to antibody immunity (TH2) [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.

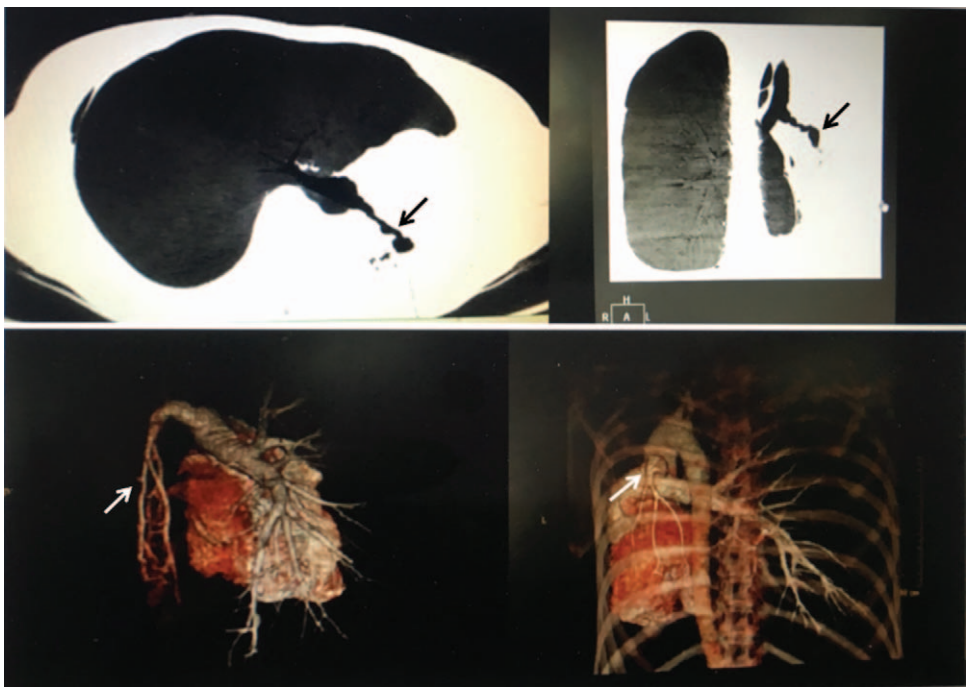

Figure 3. The 3D reconstruction (minimum intensity projection) with spiral computed tomography .Left main bronchus is thin and short and the lobar bronchi appeared reduced in caliber. 3D volume-rendered image. The left main pulmonary artery is obviously smaller than enlarged right main pulmonary artery 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.
Data curation: cuomao lazhou, yingqing shi.
Writing – original draft: shan lin.
Writing – review & editing: wei guan.

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