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Adult congenital horseshoe lung with bilateral pulmonary sequestration: A case report

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Abstract

Horseshoe lung (HL) is an infrequent congenital lung anomaly. Its main feature is that the lower lungs on both sides extend behind the pericardium and fuse across the midline, usually accompanied by pulmonary dysplasia. It is reported that 80% of HL is relevant to the abnormal return of some pulmonary veins from the right lung to the inferior vena cava or right atrium (scimitar syndrome). Most patients are within 5 years old, most commonly within 1 year old, but HL may also have no apparent clinical symptoms or mild symptoms. This case is a 36-years-old adult female who developed left chest pain more than a month ago and continued to worsen for 10 days. The patient also had repeated pulmonary infection with cough and expectoration.

Keywords

Horseshoe lung, adults, pulmonary sequestration, CTA

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Case

The patient was a 36-year-old female. The left side chest pain occurred more than 1 month ago and continued to deteriorate for 10 days. She had repeated pulmonary infection, accompanied by cough and expectoration. Her maximum temperature was 38.5°C, without chest tightness and dyspnea. Physical examination: the chest was barrel shaped, and no definite abnormality was found in chest auscultation and palpation. Cardiac examination was normal. The laboratory test of human immunodeficiency virus (HIV) was negative, and there was no contact history of tuberculosis. The patient's abdominal CT plain scan showed no obvious abnormality.

The lung window in axial and sagittal position of multidetector spiral CT showed that the bilateral basal lung isthmus was connected, extending behind the pericardium and crossing the midline for fusion. The left lower lung basal segment had sparse lung markings, with increased transparency, and patchy shadows could be seen near the spine at the bottom of both lungs. There was an obvious pleural interface between the isthmus and the left lung (arrow) (Figures 1 and 2). Transparent imaging (Figure 3) showed the isthmus lung tissue among the lungs (arrow). The three-dimensional volume-rendered imaging of the bronchus (Figure 4) showed that the bronchial branches of the lower lobe of the left lung were thinner than those of the right side. Cystic changes could be seen at the beginning of the lower lobe of the left lung. The outer and posterior basal segments of the lower lobe of the left lung fused at the beginning, and the distal branches were sparse. Thoracic CT

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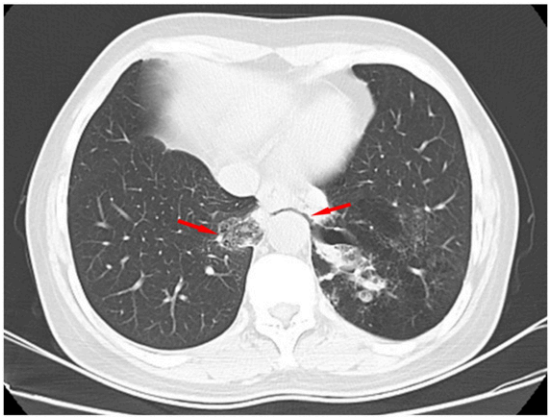

Figure 1. The lung window in axial and sagittal position of multi-detector spiral CT shows that the bilateral basal lung isthmus is connected, extending behind the pericardium and crossing the midline for fusion. The left lower lung basal segment has sparse lung markings, with increased transparency, and patchy shadows can be seen near the spine at the bottom of both lungs.

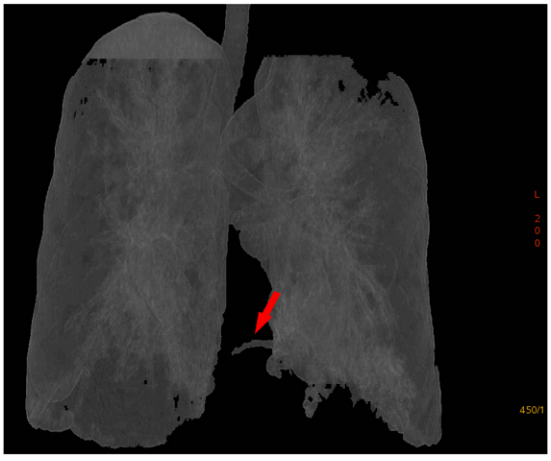

Figure 3. Transparent imaging shows the isthmus lung tissue among the lungs (arrow).

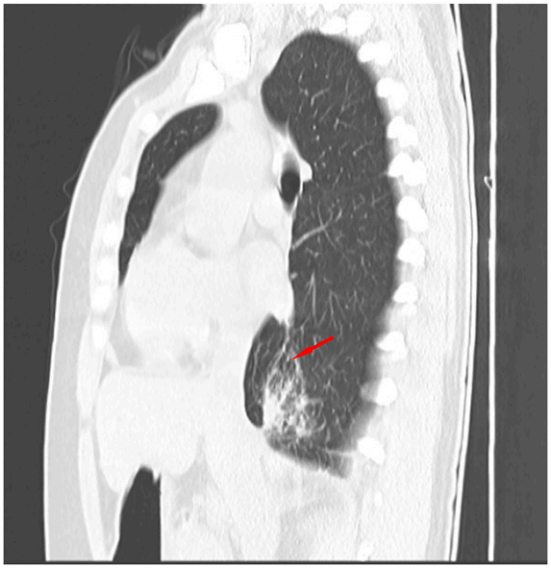

Figure 2. The lung window in sagittal position of multi-detector spiral CT: there is an obvious pleural interface between the isthmus and the left lung (arrow).

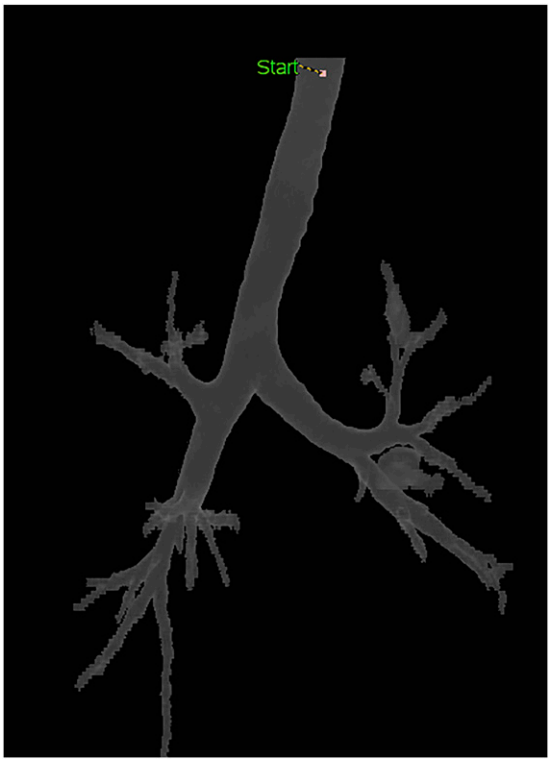

Figure 4. The three-dimensional volume-rendered imaging of the bronchus shows that the bronchial branches of the lower lobe of the left lung are thinner than those of the right side. Cystic changes can be seen at the beginning of the lower lobe of the left lung. The outer and posterior basal segments of the lower lobe of the left lung fuse at the beginning, and the distal branches are sparse (arrows).

angiography scanning (Figures 5 and 6): consolidation shadow could be seen in the basal segment of the lower lobe of both lungs, with the left lung as the focus, and multiple calcified nodules could be seen in it. The blood supply arteries of bilateral lesions directly originated from the adjacent trunk of thoracic aorta (arrows).

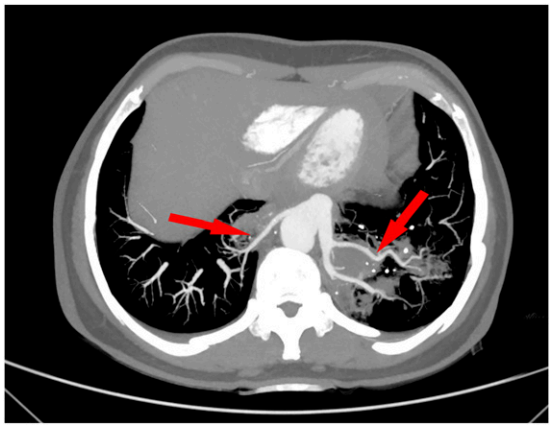

Figure 5. Maximum intensity projection (MIP) can be seen in the basal segment of the lower lobe of both lungs, with the left lung as the focus, and multiple calcified nodules can be seen in it.

Discussion

Horseshoe lung (HL) is a rare congenital pulmonary anomaly. Its main feature is that the lower lungs on both sides extend behind the pericardium and fuse across the midline and connect with the isthmus behind the heart and in front of the spine and esophagus, usually accompanied by pulmonary dysplasia.[1] It is characterized by that all or part of the right pulmonary veins drain to the inferior cavity, liver or portal vein, and includes the right pulmonary dysplasia, which is supplied by the body artery and/or the small pulmonary artery. 80% of HL is reported to be associated with abnormal return of pulmonary veins from the right lung to the inferior vena cava or the right atrium (sickle syndrome).[2,3] But this case is not accompanied by pulmonary dysplasia syndrome, and the patient is older, with bilateral pulmonary sequestration (intralobar type). So it is particularly rare. HL is often associated with unilateral bronchopulmonary dysplasia, which is characterized by unilateral bronchial agenesis, and can change into soap bubbles.[4] Envelope formation can be seen around HL, which is mainly composed of pleura. But there is still a dispute between visceral pleura of isthmus and normal lung tissue.[5] In clinic, HL may suffer from repeated pulmonary infection, left to right shunt of cardiac dysplasia, progressive pulmonary hypertension, and other symptoms. Therefore, most patients are under 5 years old, most commonly within 1 year old, but HL may also have no obvious clinical symptoms or mild symptoms.[6] This case is older and mainly related to the good development of pulmonary tissue in the isthmus and the absence of pulmonary vein malformation. After searching domestic and international literatures, the author has not found about HL cases with bilateral pulmonary sequestration in adults. Repeated pulmonary infection may be related to bilateral pulmonary sequestration. The patient's left lower lobe basal segment bronchus is hypoplasia with cystic changes (Figure 6). It is consistent with the above literature reports.

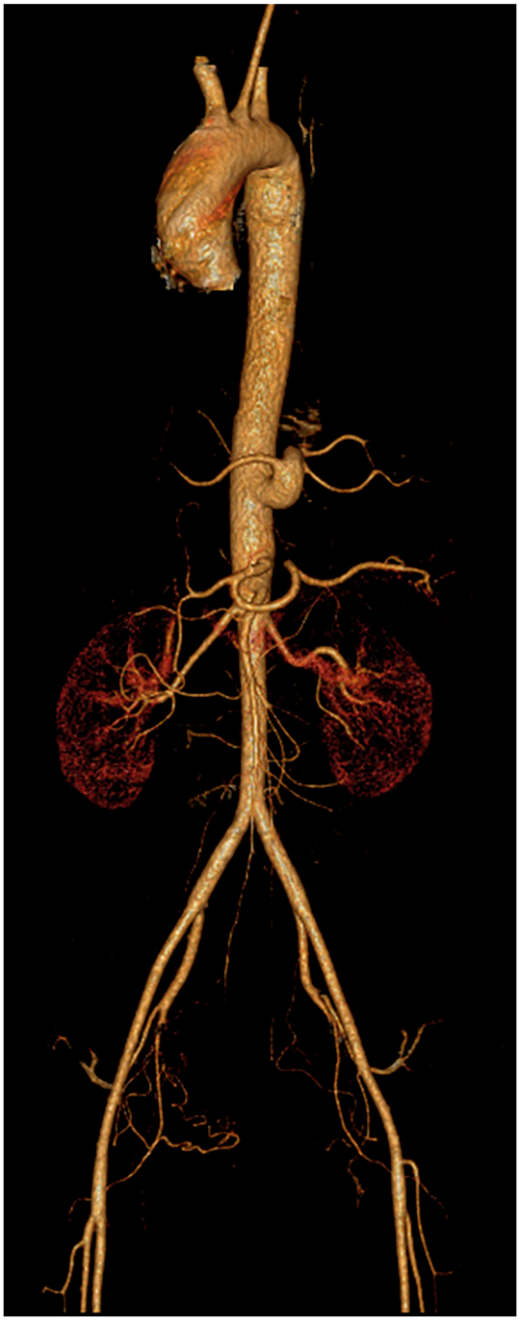

Figure 6. Volume-rendering image of CT angiography (CTA): the blood supply arteries of bilateral lesions directly originate from the adjacent trunk of thoracic aorta (arrows).

The imaging examination of HL mainly depends on CT. Chest X-ray examination mainly shows that the volume of the right chest is reduced, and the mediastinum moves to the right and dextrocardia. Chest X-ray examination can indicate that the X-ray film density is low because the air is trapped in the horseshoe-shaped lung parenchyma.[7] Cicak et al.[8] said that X-ray examination mainly diagnosed sickle lung syndrome and abnormal cardiac position and could not be used to diagnose HL. With the application of multi-slice spiral CT, HL is easier to be diagnosed. At present, multi-slice spiral CT is the best method to diagnose HL. The diagnostic criteria of CT are bilateral basal lung isthmus located behind pericardium, in front of esophagus and aorta. It can also show the situation of the diaphragm. The reconstruction technique can clearly show the lung tissue in the isthmus and the signs of underdeveloped or partially developed bronchi. Enhanced scanning or CTA examination can show the optimal vascular distribution and detect the abnormal cardiovascular and pulmonary vessels.[6] Cardiography is an indispensable imaging examination when diagnosing pulmonary hypertension.[9] This case is diagnosed mainly by multi-slice spiral CT and CTA combined with image reconstruction, and double lobar intrapulmonary sequestration is detected, which is rarely reported in literature.

This case of HL is mainly differentiated from the following diseases: (1) mediastinal pulmonary hernia: the main point of differentiation is that there is no lung parenchyma in the midline of the posterior mediastinum, and the herniated lung parenchyma has a complete pleural envelope. (2) Lung abscess (or necrotic pneumonia): the wall of the abscess is thick and obviously strengthened, and the gas-liquid level can be seen inside.

In conclusion, adult HL with bilateral pulmonary sequestration is extremely rare, which is mostly seen in children under 5 years of age. Multi-detector spiral CT shows the isthmus connection of bilateral lung basal segments as the diagnostic standard. CTA is helpful for a clear diagnosis.

Declaration of conflicting interests

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