J Korean Soc Radiol 2009 ; 61 : 105-107

Bilateral Pulmonary Sequestration with a Bridging Isthmus in an Elderly Person: A Case Report[1]

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Pulmonary sequestration is a rare congenital disorder where a portion of the pulmonary parenchyma has no communication with the tracheobronchial tree and receives the blood supply from a systemic artery. Bilateral sequestration is very rare, and a bridging isthmus is extremely rare. We report here a case of bilateral sequestration with a bridging isthmus in a 54-year-old female patient.

Index words : Bronchopulmonary sequestration
Tomography
X-ray computed

Pulmonary sequestration is a relatively rare disorder where a normal connection is missing with the bronchial tree or the pulmonary arteries. Pulmonary sequestration is classified as either intralobar or extralobar (1). Most extralobar sequestrations are thought be congenital in origin, but competing theories suggest intralobar sequestrations can be either congenital or acquired (2). Bilateral sequestration is very rare, and the presence of a bridging isthmus is extremely rare (3). We report here a case of a bilateral sequestration with a bridging isthmus in a 54-year-old female patient.

Case Report

A 54-year-old female was admitted to our hospital due to a non-productive cough and blood tinged sputum that had persisted for two weeks. A chest radiograph showed the presence of an ill-defined large cavitary lesion with internal air-fluid level at the left lower lung zone (Fig. 1A). The initial blood chemistry, serology and erythrocyte sedimentation rate (ESR) were normal. A sputum acid fast bacilli (AFB) stain revealed one positive result.

A CT scan showed the presence of multicystic lesions in the bilateral lower lobes with a bridging isthmus. The cystic lesions received the blood supply from an aberrant artery, a branch of the abdominal aorta (Fig. 1B–D). The patient was diagnosed as having bilateral pulmonary sequestration with a bridging isthmus and received oral anti-tuberculous medication.

After one month, a follow-up chest radiograph showed a decreased size of the air-fluid level in the cavitary lesion located at the left lower lung zone (Fig. 1E).

Discussion

Pulmonary sequestration is a relatively rare disorder, and is defined as a segment of the lung parenchyma that lacks communication with the tracheobronchial tree and receives an aberrant systemic arterial blood supply. The anomaly can be either intralobar or extralobar (4). Bilateral pulmonary sequestration is a rare anomaly. Srikanth et al. (5) reviewed 57 cases and reported that bilateral sequestrations only occurred in 7% of cases.

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Received February 26, 2009 ; Accepted May 8, 2009
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The presence of a bridging isthmus between bilateral pulmonary sequestrations is extremely rare (3). Bilateral pulmonary sequestrations with a bridging isthmus may mimic a horseshoe lung (3). A horseshoe lung is a rare congenital pulmonary anomaly where the posterobasal portions of the right and left lungs are fused through a

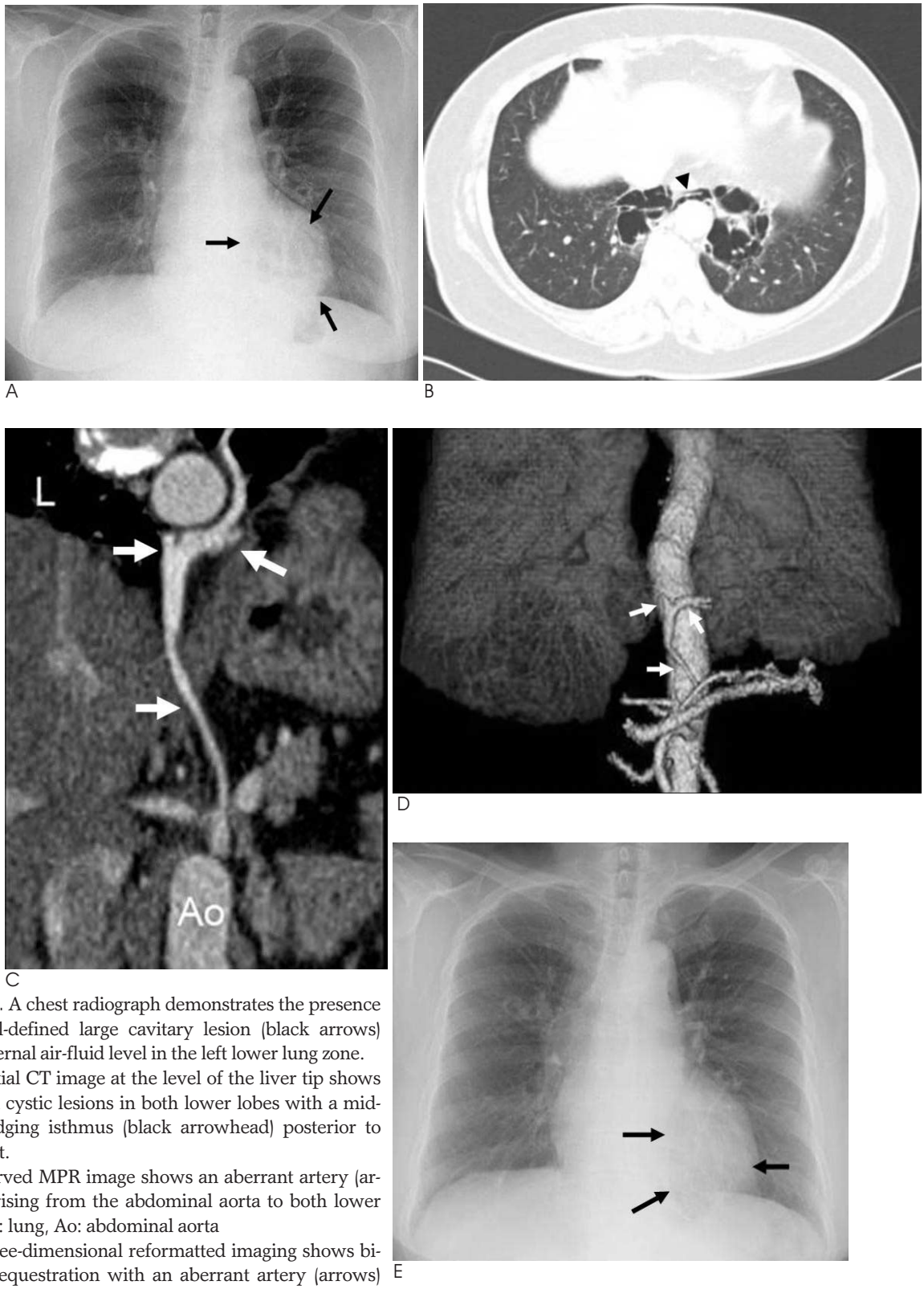

Fig. 1. A. A chest radiograph demonstrates the presence of an ill-defined large cavitary lesion (black arrows) with internal air-fluid level in the left lower lung zone.
B. An axial CT image at the level of the liver tip shows septated cystic lesions in both lower lobes with a midline bridging isthmus (black arrowhead) posterior to the heart.
C. A curved MPR image shows an aberrant artery (arrows) arising from the abdominal aorta to both lower zones. L: lung, Ao: abdominal aorta
D. A three-dimensional reformatted imaging shows bilateral sequestration with an aberrant artery (arrows) from the abdominal aorta.
E. A chest radiograph demonstrates the decreased size of air-fluid level in an ill-defined large cavitary lesion (black arrows).

narrow isthmus between the heart and aorta, and receive the blood supply from a branch of the pulmonary artery (6, 7). Most of the reported cases have been associated with a certain degree of pulmonary hypoplasia (8). The feeding artery of the mass in our case turned out to be an aberrant arterial branch from the abdominal aorta.

Until recently, angiography has been the diagnostic tool for pulmonary sequestration. However, the use of multi-detector row CT (MDCT) has transformed the approach to image the thoracic vessels and airways, especially in infants and children (9). In our case, a CT scan with three-dimensional volume rendering and multiplanar reconstruction showed bilateral sequestration with a bridging isthmus that received the blood supply from an aberrant artery, a branch of the abdominal aorta (Fig. 1).

In addition, cases confined to pulmonary sequestrations with tuberculosis infection are extremely rare. In our case, we could not identify any tuberculous or abnormal lesion with the use of CT imaging other than the sequestration area. Therefore, it was suspected that the sequestration might be infected with tuberculosis. With a positive result of sputum AFB, empirical anti-tuberculous medication was started.

A one-month follow-up chest radiograph showed decreased air-fluid level in the sequestration. M. tuberculosis might have reached the sequestration through the blood stream, through the lymphatic system as a result of progressive primary infection or from adjacent non-sequestrated lung tissue via Kohn's pores (2). Sequestrated tissues also drain poorly due to insufficient interflow between the sequestrated and normal lung airways, and are susceptible to infection.

References

1. Fakhoury KF Seilheimer DK Pulmonology. In Garson A, Bricker JT, Fisher DJ, Neish SR. *The science and practice of pediatric cardiology*, 2nd ed. Baltimore : Williams & Wilkins, 1998:2813-2832
2. Yatera K, Izumi M, Imai M, Ikegami T, Miyazaki N, Kido M. Intralobar sequestration with tuberculous infection confined to the sequestrated lung. *Respirology* 2005;10:685-688
3. Kuo R, Shih SL, Huang JK. On the AJR digital viewbox. Bilateral pulmonary sequestration with bridging isthmus in a boy with Williams syndrome. *AJR Am J Roentgenol* 2006;187:W133-134
4. Carrion-Valero F, Marin-Pardo J. Pulmonary sequestration. *Respiration* 2003;70:528
5. Srikanth MS, Ford EG, Stanley P, Mahour GH. Communicating bronchopulmonary foregut malformations: classification and embryogenesis. *J Pediatr Surg* 1992;27:732-736
6. Figa FH, Yoo SJ, Burrows PE, Turner-Gomes S, Freedom RM. Horseshoe lung—a case report with unusual bronchial and pleural anomalies and a proposed new classification. *Pediatr Radiol* 1993;23:44-47
7. Hawass ND, Badawi MG, Fatani JA, Meshari AA, Edrees YB. Horseshoe lung with multiple congenital anomalies. Case report and review of the literature. *Acta Radiol* 1987;28:751-754
8. Lutterman J, Jedeikin R, Cleveland DC. Horseshoe lung with left lung hypoplasia and critical pulmonary venous stenosis. *Ann Thorac Surg* 2004;77:1085-1087
9. Siegel MJ. Multiplanar and three-dimensional multi-detector row CT of thoracic vessels and airways in the pediatric population. *Radiology* 2003;229:641-650

대한영상의학회지 2009 ; 61 : 105-107

성인에서의 연결 협부를 가지는 양측성 폐 단락: 증례 보고[1]

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손락채 · 이재교 · 황미수

폐 단락은 기관지와의 교통이 없으며 전신 동맥으로부터 혈액 공급을 받는 폐 실질의 한 부분으로서 드문 선천성 질환이다. 양측성 폐 단락은 매우 드물며 게다가 연결 협부를 가지는 경우는 더욱 그러하다. 54세 여자 환자에서 발견된 연결 협부를 가지는 양측성 폐 단락에 대하여 보고하고자 한다.