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**[Case Report]**

# ANOMALOUS SYSTEMIC ARTERIAL SUPPLY TO THE BASAL SEGMENTS OF THE LUNG

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**Abstract :** We report on a 58-year-old man with an anomalous systemic artery to the basal segments of the left lung. The anomalous systemic artery originated from the descending thoracic aorta, distributed to the basal segments of the left lower lobe, and drained into the normal left inferior pulmonary vein. The pulmonary artery to the basal segments was not recognized. His bronchoscopic findings were normal. He underwent left lower lobectomy with division of the aberrant artery. The patient has remained well without any complications since surgery.

**Key words :** aberrant artery, basal segment, embryological development, anomalous systemic arterial supply, lobectomy

## INTRODUCTION

Anomalous systemic arterial supply to the basal segments of the lung is a relatively rare entity. It was previously classified as one type of sequestration according to Pryce's terminology[1)]. This type of anomaly is currently believed to be distinguished from pulmonary sequestration because of the deficiency of the sequestrated lung. We herein report a rare case of a patient with an anomalous systemic arterial supply to the basal segments of the lung who was treated by curative surgery. We also review the 58 English-language reports to date of an anomalous systemic arterial supply to the basal segments of the lung.

## CASE

A 58-year-old male patient was admitted to our hospital because of an abnormal shadow on a routine chest X-ray film (Fig. 1). He had no clinical symptoms. A reconstructed computed tomography (CT) scan revealed a dilated aberrant artery from the descending thoracic aorta, and the basal pulmonary artery was not detected (Fig. 2-A, B). Blood drained into the left atrium. No sequestrated lung was identified. His bronchoscopic findings showed normal branches. He was diagnosed with an anom-

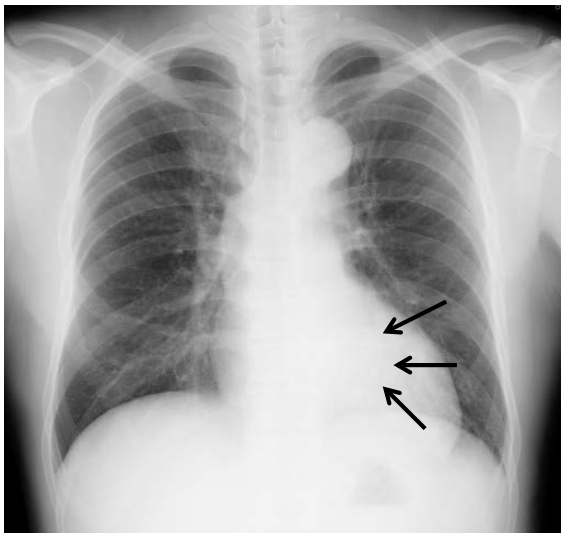

Fig. 1. Chest roentgenogram shows an abnormal shadow in the left lower lung field, which displays a negative silhouette sign with the heart (arrows).

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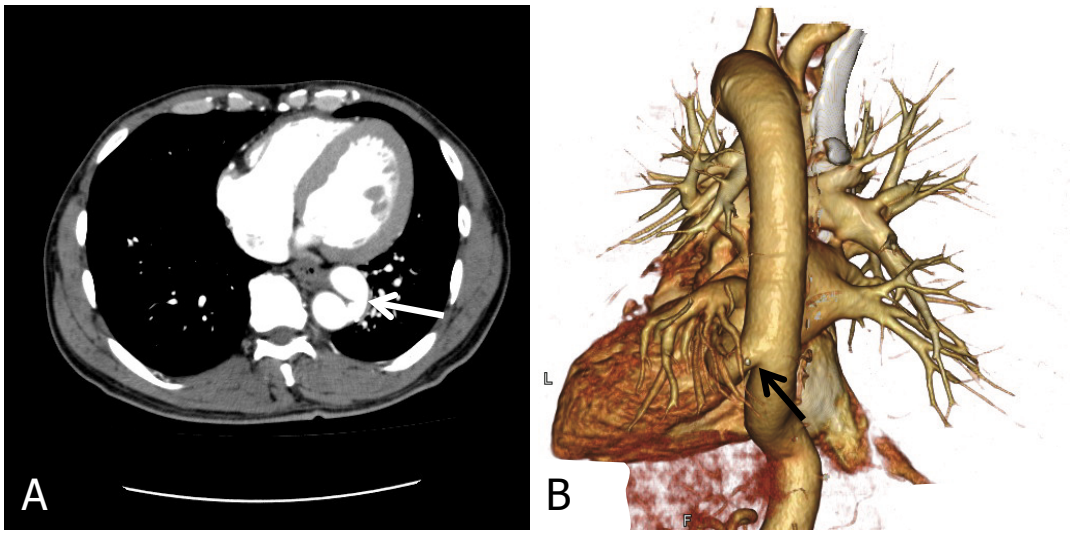


Fig. 2. (A) The aberrant artery arises from the descending thoracic aorta (DTA) (white arrow), (B) 3D-CT also shows aberrant artery from DTA (black arrow) and the basal pulmonary artery is not detected.

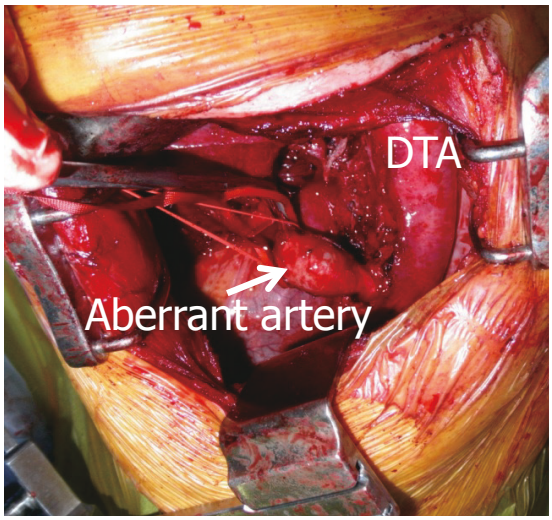


Fig. 3. Aberrant artery, 25 mm in diameter (arrow), arises from DTA.

alous systemic arterial supply to the basal segments of the left lung. He underwent curative surgery because the aberrant artery was dilated like an aneurysm and may have had a risk of rupture. At thoracotomy, the aberrant artery, which originated from the descending thoracic aorta, was identified. The basal part of the left lower lobe had an orange-peel appearance with telangiectatic markings. The diameter of the aberrant artery was 25 mm (Fig. 3), and it was ligated at the base and cut with an autostapler. Left lower lobectomy was then performed. The interlobar pulmonary artery was ligated, and the basal artery was not recognized. The left inferior pulmonary vein and left lower bronchus were treated by autostapling using a routine technique. Pathological examination revealed parenchymal hemorrhage, congestion of papillary vessels, and hypertrophy of the muscle

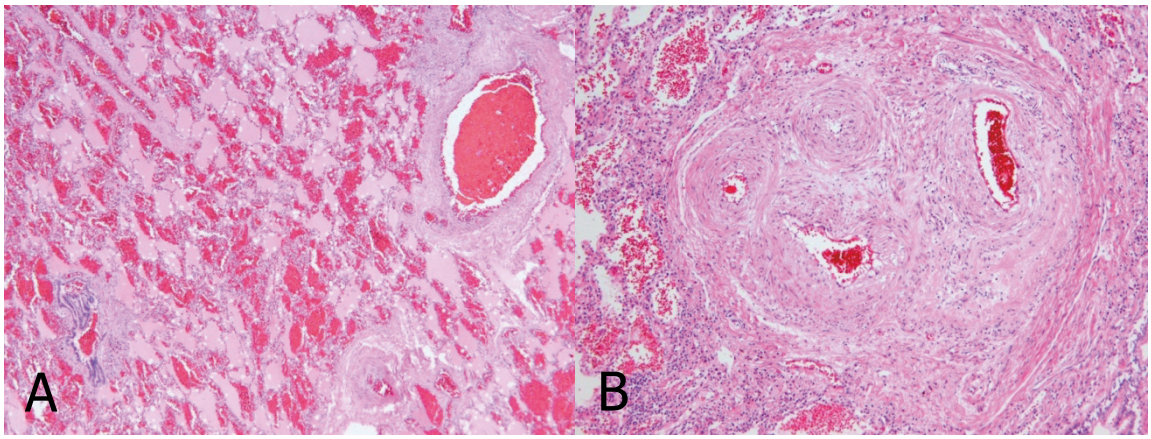


Fig. 4. (A) Pathological examination revealed parenchymal hemorrhage, congestion of the papillary vessels, and (B) hypertrophy of the muscle layer of the pulmonary artery.

layer of the pulmonary artery (Fig. 4-A, B), which suggested the presence of local pulmonary hypertension. The postoperative course was uneventful, and the patient was discharged from the hospital on the eighth postoperative day. He has been well without any complications or symptoms since surgery.

## DISCUSSION

In this report, we described a relatively rare disease of anomalous systemic arterial supply to the basal segments of the left lung. This anomaly is currently classified as an independent disease since Painter *et al*. presented it in 1968[2].

According to our review of 58 cases of anomalous systemic arterial supply to the basal segments of the lung in the English-language medical literature, including our case (Table 1), the major symptoms are as follows : hemoptysis ($n$ = 22, 37.9%), abnormal shadow ($n$ = 9, 15.5%), heart murmur ($n$ = 6, 10.3%), exertional dyspnea ($n$ = 6, 10.3%), and so on. The most common site of this disease is the left side ($n$ = 53, 91.4%), and in these cases, the aberrant arteries arise from the descending thoracic aorta. In cases involving the right side ($n$ = 5, 8.6%), the origin of the aberrant arteries is the abdominal aorta such as the celiac axis. This difference might be caused by the anatomical structure. Interestingly, 47 (81.3%) of the reported cases were East Asian patients, and 11 (18.7%) were Caucasian. The analyzed statistical data suggest a caus-

Table 1. Review of the literature published in English to date of anomalous systemic arterial supply to the basal segments of the lung, including the present case

| | | | |
|---|---|---|---|
| Age (years) | 32.0 ± 20.7 (0-71) | Aberrant arterial perfusion | |
| | | Basal segment | 57 |
| Sex | | Basal and lingular segment | 1 |
| Male : Female | 33 : 22 (Unknown 3) | | |
| | | PA perfusion | |
| Total number of clinical symptons | | S6 | 41 |
| Hemoptysis | 22 | S6 and other segment | 3 |
| Abnormal shadow | 9 | Normal | 2 |
| Heart murmur | 6 | Unknown | 12 |
| Exertional dyspnea | 6 | | |
| Tachypnea | 4 | Drainage vein | |
| Cough | 4 | Inferior PV | 50 |
| Others | 12 | Unknown | 8 |
| Unknown | 11 | | |
| | | Treatment | |
| Side | | Lobectomy | 21 |
| Right : Left | 5 : 53 | Segmentectomy | 3 |
| | | Anastomosis | 2 |
| Origin of aberrant arteries (total) | | Ligation | 2 |
| Descending thoracic aorta | 54 | Embolization | 8 |
| Celiac axis | 4 | Follow-up | 6 |
| Other abdominal aorta | 2 | Unknown | 16 |
| | | | |
| The number of aberrant arteries | | Approach (of 28 surgical cases) | |
| Single | 45 | Thoracotomy | 25 |
| Multiple | 2 | VATS | 3 |
| Unknown | 11 | | |
| | | Ethnicity | |
| Maximum diameter (mm) | 24.9 ± 20.7 (4.4-86) | Asian | 47 |
| | (Unknown, 45) | Caucasian | 11 |

PA : pulmonary artery, PV : pulmonary vein, VATS : video-assisted thoracic surgery

ative factor of East Asian rather than Caucasian ethnicity.

We herein present a new hypothesis about the embryological development of an anomalous systemic arterial supply to the basal segments of the lung. Normally, the embryonic sixth aortic arch interposes between the embryonic primitive pulmonary artery and pulmonary trunk[3]. In the formation of an anomalous arterial supply, the primitive pulmonary artery might be divided into two segments, and the sixth arch might interpose between these two arteries and the pulmonary trunk or descending thoracic aorta. As a result, the unilateral pulmonary artery distributes to the upper/middle lobe and S6 segment, while the aberrant systemic artery distributes to the basal segments.

Patients with this disease are considered to require surgery or radiological intervention because of the risk of hemoptysis due to pulmonary hypertension, heart failure due to a left-to-right shunt, and infection[4, 5], even patients without any symptoms. Accordingly, we decided to perform surgery in this case. The surgical procedure includes lobectomy, basal segmentectomy, simple cutting of the aberrant artery, and anastomosis of the aberrant artery and pulmonary vein. Furthermore, a non-surgical procedure such as catheter embolization of the aberrant arteries is sometimes performed instead of surgery[6-8]. In our case, we chose pulmonary lobectomy because it is still controversial to select a non-surgical treatment as radical treatment for this disease. Furthermore, no long-term outcome data including surgical treatment have been available until now. Therefore, these patients must be followed up for a long period of time to evaluate the value of the procedures. And we should probably consider a minimally invasive treatment such as video-assisted thoracic surgery for this benign congenital disease according to recent reports[9, 10].

## CONFLICT OF INTEREST

None declared.

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