좌측 아폐엽에 궁급하는 비정상적인 체동맥의 치료 (수술과 색전술의 비교)

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Treatment of Systemic Arterial Supply to Lower Lobe of Left Lung (Operation vs. Embolotherapy): Comparison of Two Cases and Literature Review

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Systemic arterialization of lung with/without sequestration (Sequestration/Anomalous Origin of Left Pulmonary Artery, AOLPA) is a rare form of congenital anomalous systemic arterial supply to the lungs. In this anomaly, the arterial supply of one or more arteries of the basal segments of the lower lobe derives from an aberrant vessel arising from the aorta. We report two adult cases of systemic arterialization of normal basal segments of left lower lobe lung with/without sequestration. The one (AOLPA) was treated by left lower basal segmentectomy and the other (Sequestration) by therapeutic angiographic embolization. Based on the favorable follow-up result in our patients, although lobectomy (segmentectomy) is the basic treatment modality, embolotherapy could also be a mode of treatment that could be selectively applied to elderly, infirm patients or high risk patients with poor pulmonary function.

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Key words: 1. Lung, anomaly

2. Pulmonary arteries

3. Embolization

CASE REPORT

1) F/26, Mild Chest Discomfort

A 26-year-old woman visited our hospital due to intermittent discomfort on left lower hemithorax. The past history and family history of the patient were not remarkable. Physical and laboratory findings were within normal limits. A

chest radiograph demonstrated about 2 cm sized well-defined nodular density at left retrocardiac area (Fig. 1). Chest CT with contrast enhancement showed a large anomalous artery arising from lower thoracic aorta supplying basal segments of left lower lobe. No evidence of bronchial abnormalities was noted in the lung (Fig. 2). Pulmonary arteriography showed absence of normal pulmonary artery and vein to basal seg-

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본 논문의 저작권 및 전자매체의 지적소유권은 대한흉부외과학회에 있다.

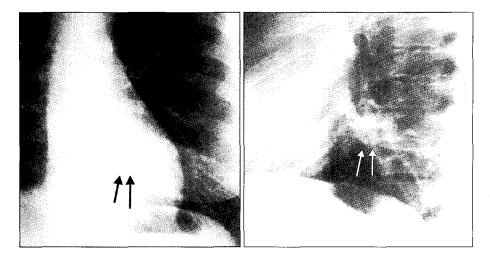


Fig. 1. F/26, Mild chest discomfort. About 2 cm sized nodular density at left retrocardiac area (arrows) in normal lung parenchyme is noted on chest PA and left lateral views.

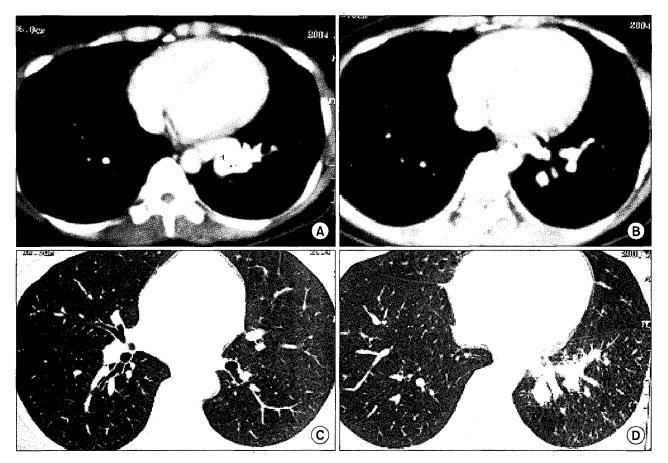


Fig. 2. Well-enhancing nodular lesion at left lower medial lung zone (A), increased vascularity of left lower lobe (B), and normal pulmonary parenchyma and bronchial structure are noted (C, D) in chest CT with enhancement.

ments of left lung. The pulmonary artery supplied entire right lung, left upper lobe and superior segments of left lower lobe and the pulmonary vein drained from concomittent parenchyme. Thoracic aortography showed anomalous systemic artery from left lateral margin of descending aorta, supplying left lower lobe basal segments of lung, a normal parenchymal

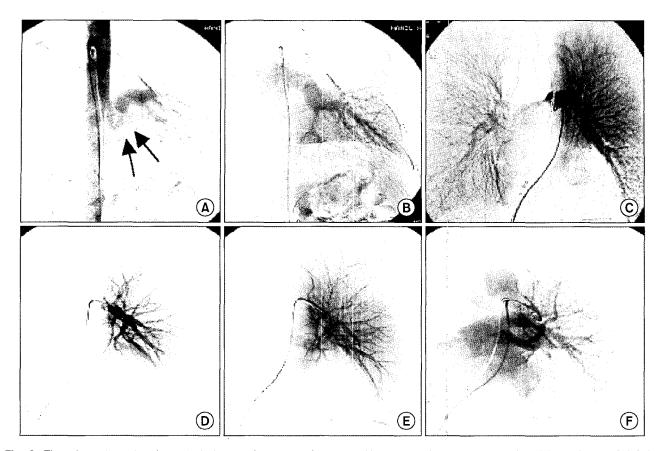


Fig. 3. Thoracic aortography shows typical anomalous systemic artery with swan neck appearance and draining vein to left inferior pulmonary vein from left lower lobe basal segments of lung (A, B). Pulmonary arteriography shows absent segmental arterial and venous branches of left lower lobe basal segments ($C \sim F$).

phase and a draining vein to normal left lower pulmonary vein (Fig. 3). On the diagnosis of anomalous systemic arterial supply to the basal segments of left lower lobe from descending aorta without sequestration, left lower basal segmentectomy with ligation of the abnormal artery was done. Operative findings revealed similar findings to radiographic results, and the involved lung surface showed multiple tiny telangiectases. The anomalous artery was about 1 cm in diameter in the left pulmonary ligament at its origin and became tortuous cranially to the proximal part of lower lobe like a swan neck-shaped curve, and then aneurysmally dilated up to 1.5 cm running along with normal pulmonary artery in the basal segments (Fig. 4). The pathologic examination showed a large anomalous artery of elastic type, normal bronchial communication, and multiple telangiectases in the visceral pleural surface. The patient did well after operation without evidence of complications.

2) M/16, Incidental Lung Nodule

A 16-year-old man admitted to the hospital with acute right chest pain due to pneumothorax. Closed thorocostomy was done. About 1 cm sized nodular density was found incidentally at left retrocardiac area on chest radiograph. Chest CT showed well-enhancing vascular structure at left lower medial lung zone (Fig. 5A), and the fissure density at left retrocardiac area. The findings suggested it to be pulmonary sequestration. The patient's past and family history were not remarkable. On the diagnosis of pulmonary sequestration, thoracic aortography was done. It showed a curved shadow of left lower lung that suggested an accessary fissure artifact of the pulmonary sequestration, and an about 0.7 cm sized anomalous systemic artery from the descending thoracic aorta supplying left lower lobe basal segments of lung. A draining vein to left lower pulmonary vein and no evidence of ab-



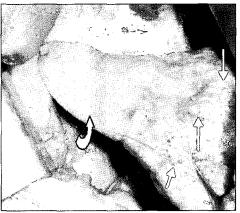
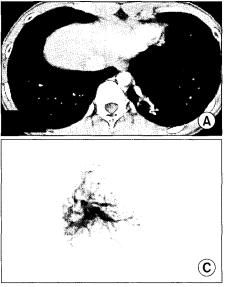


Fig. 4. Operative finding shows a large anomalous systemic artery from descending thóracic aorta, and multiple telangiectases (short arrows) in the surface of basal segments and normal surface of lower lobe superior segment of left lower lobe of lung (curved arrow).



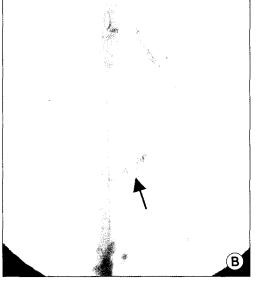


Fig. 5. Anormalous artery is noted at left lower medial lung zone on enhanced CT (A). Aortography shows anomalous artery from left side of lower thoracic aorta (arrow) (B). Selective arteriography shows localized parenchymal lung staining without evidence of fistulous lesion (C).

normal fistula draining to the pulmonary vein was found (Fig. 5B, C). Therapeutic embolization was done with metallic coils and gelfoam particles (Fig. 6). The patient was discharged in a week, and did well after embolotherapy for 2 year follow-up without evidence of lung abscess, pneumonia, or systemic thromboembolism.

DISCUSSION

Anomalous systemic arterial supply to the lungs has been described in bronchopulmonary sequestration, systemic arterialization of normal lung (Anomalous Origin of Left Pulmonary Artery, AOLPA) and the scimitar syndrome. This vessel always supplies one of the lower lobes, frequently one of the

basal segments[1-6]. The absence of parenchymal abnormalities and a normal bronchial supply clearly distinguishes systemic arterial supply without sequestration from true sequestration[4]. In AOLPA, the feeding vessel in systemic arterial supply of normal lung without sequestration arises from the distal part of the thoracic descending aorta, the proximal abdominal aorta or the celiac axis, and it is an elastic type artery[2-6]. The systemic artery can measure up to 1 cm in diameter, is smaller at its origin in the pulmonary ligament, but getting dilated just beyond the site entering the lobe, especially in the left-sided cases, as in our case 1. In most cases atresia of the corresponding pulmonary artery was present[2,4]. There were some cases with normal branching of the pulmonary artery in the involved segment[7,8]. The

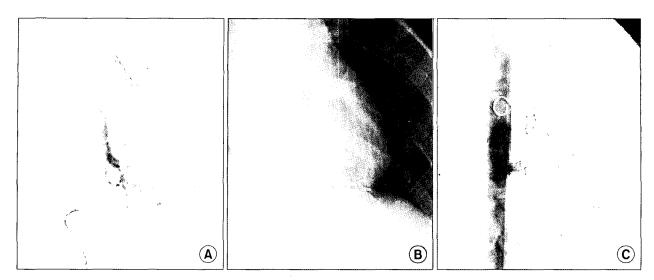


Fig. 6. Venous phase of selective arteriography shows draining vein to normal left inferior pulmonary vein (A). After embolization with microcoils and gelfoam particles, stasis of contrast and embolic material are noted (B). Postembolization aortography shows complete occlusion of anomalous artery with short residual stump at its origin (C).

venous return was always via normal inferior pulmonary vein, causing a left-to-left shunt[2-8].

Symptoms of AOLPA were cough, sputum, fever, and dyspnea, but were usually asymptomatic in most patients. Therefore most of these were diagnosed as an incidental finding on the chest roentgenogram. However, left ventricular failure from left-to-left shunt, or massive hemoptysis may result[2,5,7].

We treated two patients with systemic arterialization of normal basal segments of left lower lobe by basal segmentectomy in the one (AOLPA) and therapeutic embolization in the other (Sequestration) without significant complications in 2 year follow-up. Based on the favourable follow-up result in our patients, lobectomy (segmentectomy) should be performed as the basic treatment modality. However, embolotherapy may be considered selectively an alternative to thoracotomy in patient with systemic arterialization of lower lung to high risk patients of old age or with debilitation, or with poor pulmonary function.

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=국문 초록=

페에 체동맥이 분포하는 선천성 기형은 드문 질환으로 하페엽의 기저분절에 페동맥 대신 대동맥에서 1개 이상의 체동맥이 나오는데 정상적인 기관지와 연결 유무에 따라 페 분획증과 좌측 페동맥 이상 기시증(Anomalous Origin of Left Pulmonary Artery, AOLPA)으로 구분된다. 한일병원 흉부외과에서는 하행 흉부 대동맥에서 시작되는 비정상적인 체동맥을 가진 두 예의 페동맥 기형을 경험하고, 1예는 기저분절절제술을, 다른 예는 동맥색전술을 시행하였고, 2년간 관찰한 결과 양호하였다. 이 결과를 바탕으로 외과적 수술이 이 기형의 근본적인 치료법이지만, 전신상태가 불량하거나 페기능이 저하된 고위험군에서 동맥색전술도 선택적으로 시행할 수 있는 가능한 치료법으로 생각된다.

중심 단어 : 1. 폐, 기형

2. 폐동맥

3. 색전술