Surgical Correction of Congenital Vascular Anomaly of Left Lung, Anomalous Systemic Arterial Supply to Normal Basal Segments of Left Lower Lobe (PALLADA): A Report of Eight Cases

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Abstract

Eight cases of PALLADA were found. Preoperative diagnosis and hemodynamic confirmation were made in all cases except the first case. Surgical correction by anastomosing the abnormal artery to the existing pulmonary is recommended. Lobectomy or division of anomalous shunt is also a choice of treatment in some cases. No operative complications occurred.

Key word : Anomalous Artery, Systemic Arterial Supply to the Lung, Systemic Arterialized of a Lower Lobe, Pseudosequestration, PALLADA

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Congenital anomaly of the pulmonary artery of the left lower lung called PALLADA (Pulmonary Artery of Left Lower lobe Arising from Descending Aorta) is a rare anomaly. After our new clinical criteria were established in 1985, the anomaly has been diagnosed frequently (6 cases in 6 years)[1,2]. In some cases this abnormality could have been misdiagnosed as intralobar pulmonary sequestration preop, intraop and even postoperatively[1]. Both abnormalities are quite different anatomically, physiologically as is the treatment.

At the Central Chest Hospital, eight cases of this anomaly were diagnosed from 1985 to 1998. All cases, except the first case, had been diagnosed preoperatively using our new clinical criteria which was established after carefully studying the first case. We have tried to treat all the patients by transferring the abnormal artery from the aortic origin to the pulmonary artery in order to preserve their lung parenchyma and improve their gas exchange function to a near normal status. Unfortunately, in some cases, we had to remove the lower lobe because

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our findings suggested that the lower lung had changed and not worth preserving.

CASE REPORTS

Case 1.

A Thai female, aged 34 years, was admitted to the Central Chest Hospital in October, 1985. She stated that her pulmonary symptoms had started 1 year ago with fresh blood streak sputum without fever or chest pain. Her chest X-ray showed a faint hazy area behind the left heart shadow which might have been overlooked if the left lateral chest X-ray had not been taken. Fiberoptic bronchoscopy revealed a small amount of blood clot in the lateral basal segmental bronchus of the left lower lobe (LB9) without any other abnormality. Left lower lobe postbronchoscopic bronchography was performed and showed minimal ectatic change of all left lower lobe segmental bronchi.

On the left thoracotomy, there was an abnormal blood vessel, 1 centimeter in diameter, originating from the antero-lateral surface of the lower descending thoracic aorta. The vessel curved 3 centimeters downward then turned upward directly to the left lower lobe lung parenchyma. A mass, 5 centimeters in diameter was palpable. The lung surface of all four basal segments of the left lower lobe, visceral pleura, showed marked dilated pleural capillaries with numerous focal fibrinous hemorrhages seen. In contrast, visceral pleura covering the superior segment of the left lower lobe, looked normal.

Intralobar pulmonary sequestration was diagnosed and left lower lobe lobectomy was performed.

Case 2.

A Thai female, aged 19 years, complained of a tight feelling in the left side of her chest and had slight shortness of breath. Physical examination revealed suspicion of ASD Primum. The patient was referred to our hospital. PA chest X-ray showed right ventricular and right atrial enlargement. The left lower lung field was normal. Left lateral film showed a tubular hazy shadow in the left lower lung field. Echocardiography revealed ASD with minimal mitral regurgitation. No abnormal vessels were detected. Pulmonary angiography showed the pulmonary artery to the left lung supplied all segments of the left upper lobe but only one segment of the left lower lobe (superior segment). Branches to the four basilar segments of the left lower lobe were absent. Aortography showed an abnormal artery, 0.7 to 1.0 centimeters in diameter, arising from the antero-lateral aspect of the descending lower thoracic aorta. The artery curved downward and turned upward before penetrating into the lower lung. In the lung, the artery curved downward and immediately separated into four main branches supplying the four basal segments of the lower lobe.

Operative findings: An abnormal vessel from the descending aorta as described above. Tensed pulsatile abnormal vessels to the basal segments simulated a palpable mass in the lower lung. All the lower lung surfaces showed marked generalized dilated vessels in visceral pleura together with numerous old hemorragic areas except the superior segment of the lower lobe in which normal visceral pleura were observed. The arising artery from the aorta was transfered to and anastomosed with the left pulmonary artery at the level near the interlobar fissure. After the abnormal artery was isolated from the aorta, the palpable mass was significantly diminished in size.

Case 3.

A Thai male, aged 27 years, was admitted with a history of spitting fresh blood off and on for four years. Physical examination was normal. PA chest X-ray showed an abnormal faint hazy shadow in the left lower lung field behind the heart shadow. Left lateral chest X-ray showed the haziness starting at the level of T9 vertebra then separating into three or four unsharped tubular bands at the level of T10-11. Tomography confirmed a vascular liked shadow with downward branching. Bronchofiberscopy was normal. All basal segments of the left lower lobe showed no evidence of external compression. Bronchography showed external compression and deviation of branches of the lower lobe basal bronchi caused by an abnormal vascular shadow. Echocardiography yielded normal findings without evidence of an abnormal vessel from the aorta. Pulmonary angiography demonstrated a small left pulmonary artery to the lower lobe. The artery supplied only the superior segment. On aortography, an abnormal artery arising from the descending thoracic aorta at the level of T9 vertebra was found. The artery, 1 centimeter in diameter, curved 4 centimeters downward and turned upward with a diameter of 1.3 centimeters to the previous level and separated into four downward branches supplying the basal segments of the left lower lobe.

Operative findings were the same as in the previous cases. The abnormal arising artery was divided from the aorta and mobilized upward to anastomose with the left pulmonary artery in the interlobar fissure.

Case 4.

A Thai female, aged 48 years, came to our hospital with a history of occasional hemoptysis for 1 year. She complained of left chest pain. Physical examination was normal. PA chest X-ray showed a round mass, 6 centimeters in diameter, sharp at the border, at the level of T9 vertebra in the left lower lung field surrounded by small hazy shadows. On the lateral chest film, the mass was in the posterior part of the lung superimposed with the lower portion of T8, T9 and upper portion of T10 vertebrae. Bronchofiberscopy showed the left posterior basal segmental bronchus 50 per cent narrowing with oozing hemorrhage. On thoracotomy, the pulmonary artery and the lung surfaces were as described above. There were two abnormal arteries arising from the descending thoracic aorta. A palpable mass like lesion was quite big and the remaining normal lung parenchyma was too small. Left lower lobectomy was performed.

The specimen was carefully studied and it was found that the palpable mass were large hematomas.

Case 5.

A Thai male, aged 33 years, came with a history of intermittent blood streaked sputum and left chest pain for years. Physical examination showed no abnormality. PA chest X-ray demonstrated minimal left ventricular enlargement. A small hazy shadow in the left lower lung field was suspected. Penetrated PA confirmed the mass like lesion. Left lateral chest X-ray also yielded more information about the shadow. Bronchofibrescopy found deviation of all left lower lobe basal segmental bronchi at the location of the abnormal shadow on the chest X-ray. Bronchial mucosa was normal. The deviations were confirmed by post bronchoscopic bronchography. On aortography, at the level of T8 vertebra, there was an abnormal artery arising from the anterolateral aspect of the descending thoracic aorta. The artery, 1.5 centimeters in diameter, curved 3 centimeters downward and turned upward 4.5 centimeters then turned downward by dividing into four branches supplying the four basal segments of the left lower lobe.

This patient refused surgery.

Case 6.

A Thai male, aged 26 years, was admitted with a history of blood streaked sputum intermittently with left chest pain. On auscultation, bruit was detected at the posterior part of the left lower chest. PA chest X-ray showed a hazy shadow in the left lower lung field behind the cardiac shadow. The upper margin of the haziness had a sharp border while the lower part separated as a blurred tubular shadow facing the diaphragm. Lateral chest film confirmed the abnormal shadow. Tomography gave more anatomical details of the vascular lesion. Fiberoptic bronchscopy showed deviation and minimal external compression of all basal segments of the left lower lobe. Post bronchoscopic bronchography confirmed the bronchoscopic findings. Aortography was performed. At the T8 vertebra, there was an abnormal anterior branching, 1.5 centimeters in diameter, from the descending thoracic aorta. The artery curved downward 4 centimeters and turned upward 4 centimeters then divided into four branches while turning downward toward the diaphragm supplying the four basal segments of the left lower lung. CT-chest showed an abnormal shadow in the left lower lung. The attenuation value of the shadow was similar to blood in the heart chamber both before and after contrast enhancement. MRI oblique sagittal scan revealed an abnormal artery arising from the descending thoracic aorta at the level of T8-9 vertebra. Distal to the origin of the artery could not be traced as the artery coursed tortuously. Lung ventilation perfusion scan using Tc99m-MAA intravenous injection, showed no uptake at the basal segments of the left lower lobe. Ventilation scan of the lung by inhaled Tc99m-DTPA showed diminished uptake 7.76 per cent, 6.28 per cent at the superior segment and basal segments of the left lower lung respectively.

The patient refused operation.

Case 7.

A Thai male, aged 63 years, was referred from a medical school with a diagnosis of PALLADA. He had had hemoptysis for months with a faint hazy retrocardiac shadow. Clinical investigations suggested anomalous artery of the aorta to the lung. Aortography and pulmonary arteriography confirmed the diagnosis of PALLADA.

On left thoracotomy, the lung surface of the four basal segments showed dilated vessels. The anomalous vessel, 1.2 centimeters in diameter, originated from the aorta. The vessel curved 2 centimeters downward and turned 2 centimeters upward before entering the lower lobe. In this case, there were small branches of the left pulmonary artery of the left lower lobe supplying the basal segments.

The anomalous vessel was ligated and divided without anastomosing.

Case 8.

A Thai female, aged 25 years, was admitted with a history of spitting blood for years. Clinical investigations suggested the abnormalities. Aortography and pulmonary arteriography confirmed the diagnosis of PALLADA.

Operative findings were the same as in the previous cases. The anomalous artery was 1 centimeter in diameter and 3 centimeters in length. Anastomosis was performed.

DISCUSSION

PALLADA (Pulmonary Artery of left Lower Lobe Arising from Descending Aorta.) is a rare congenital anomaly which might frequently be misdiagnosed as intralobar pulmonary sequestration[1]. In the operative field, a large abnormal systemic artery was found arising from the descending thoracic aorta penetrating into the left lower lobe and a palpable mass in the lung resembling an intralobar sequestrated lung. Both findings have mislead surgeons to the diagnosis of intralobar pulmonary sequestration and lower lobectomy (or segmentectomy) was selected. In our cases, case no 1, also had the same findings as described above and lower lobe lobectomy was performed. From this case the following findings were derived.

1. The pulmonary artery of the left lung supplied all segments of the left upper lobe and only one segment of the lower lobe (superior segment). There were no branches to the four basal segments.

2. On the visceral pleura over the four basal segments of the left lower lobe, there were marked generalized dilated capillaries with numerous areas of previous hemorrhage. In contrary, visceral pleura which covered the superior segment of the left lower lobe looked normal, creating a distinct demarcated zone between the superior segment and the four basal segments. (Fig. 1)

3. The abnormal artery from the descending aorta originated at the level of T8 or T9 vertebra and formed a 4-5 centimeters "u" curve before entering the lung parenchyma. (Fig. 2)

4. The mass like lesion in the lower lung, 3-5 centimeters in diameter was palpable. This mass decreased in size or disappeared after abnormal artery occlusion or discontinued from high aortic pressure.

From additional detailed study of the removed specimens from all cases and preoperative investigations, the following abnormalities were concluded.

1. Bronchus to the lower lobe, especially the four basal segments, were normal. (Fiberoptic bronchoscopy, bronchography, CT and pathohistological study)

2. Abnormal vessels from the aorta, after penetrating the lower lobe, divided into four branches (similar to normal branching of the lower lobe pulmonary artery) supplying the four basal segments along the normal bronchi to the alveoli

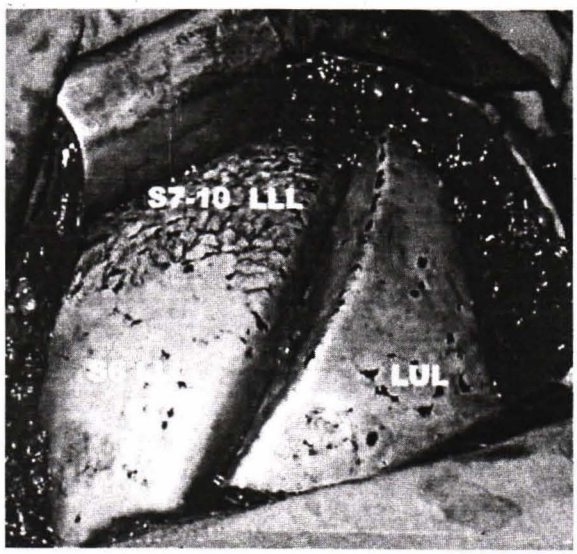

Fig. 1. On thoracotomy: Head of patient is to the right. Left lung was fully inflated. Marked generalized dilated capillaries network over four basal segments (S7-10) of left lower lobe. The abnormality did not appeared over the surface of superior segment of left lower lobe (S6) and left upper lobe.

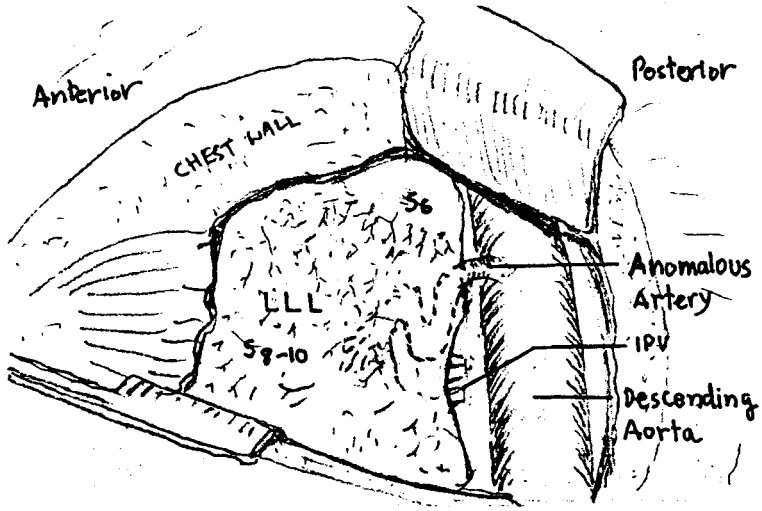

Fig. 2. On thoracotomy; Head of patient is to the top. Abnormal artery arised from antero-lateral aspect of descending thoracic aorta at the level of 9th thoracic vertebra (T9). The artery penetrated the left lower lung 2 centimeters above the level of inferior pulmonary vein.

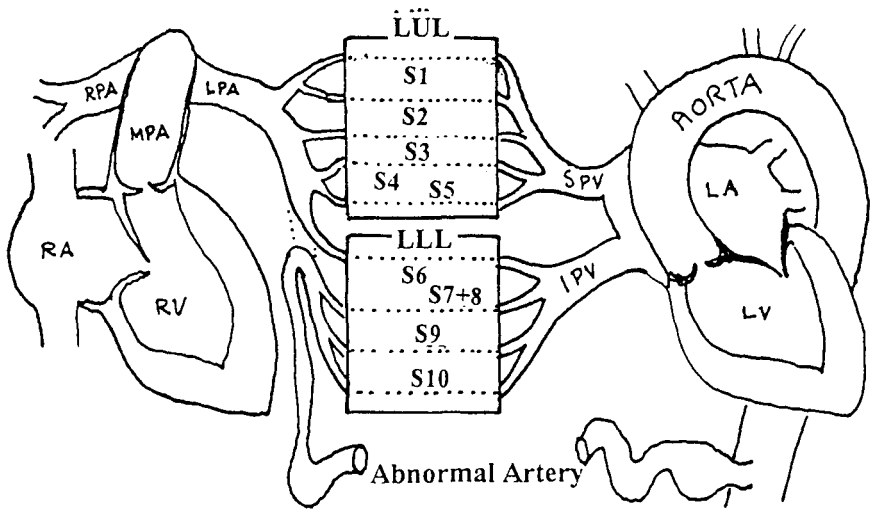

Fig. 3. Simplified diagram shows the abnormal left to left shunt through the four basal segments of left lower lobe *via* the abnormal artery.

with high systemic pressure. This artery drained into the inferior pulmonary vein and left atrium respectively. This abnormal artery created a left to left shunt. (Fig. 3)

3. Lung parenchyma, as a result of systemic high pressure, showed some degree of changes and focal hemorrhages.

From all the described abnormalities, we found that the anomaly was reported as Systemic arterialization of a lower lobe[3]. High systemic origin of the sole artery to the basal segments of the left lower lung[4]. Systemic supply to a normal lung[5]. Absent or hypoplasia of pulmonary with anomalous systemic arteries to the lung[6].

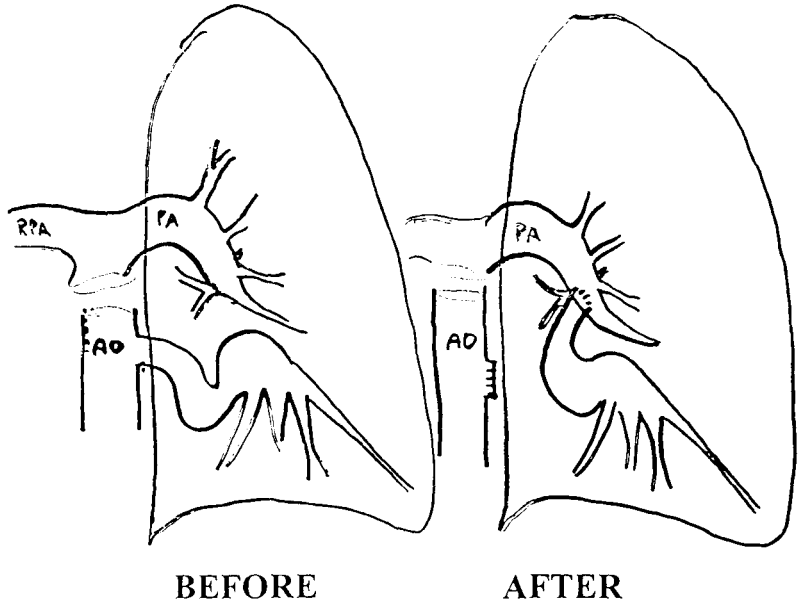

Fig. 4. Drawing demonstrated surgical correction of the anomaly by anastomosing the abnormal artery to the existing pulmonary artery.

Pulmonary Artery of left Lower Lobe Arising from Descending Aorta. (PALLADA)[1,2]. Anomalous systemic arterial supply to normal basilar segments of the left lower lobe of the left lung[7]. Anomalous systemic arterial supply to normal basal segments of the left lower lobe[8]. Systemic arterial supply to the lung without sequestration[9]. Pseudosequestration[10].

Diagnosis of PALLADA and hemodynamic confirmation preoperatively is the most important guide for proper treatment. The patients' complaints of occasional minimal fresh blood spitting. Chest X-ray showing faint haziness on the left lower lung field where haziness is prominent on the left lateral film. Tomography, CT scan or MRI give more details of a vascular like shadow. The definite diagnosis should be confirmed by aortography and pulmonary angiography[1,2].

Surgery is recommended in all cases to discontinue the arteriovenous shunting through the lung. In order to re-utilize the gas exchange function of the previously non functional basal segments of the left lower lobe, transection of the anomaly vessel from the aorta at its origin and bring its proximal end up to the hilum to anastomose to the infero-lateral surface of the pulmonary artery. (Fig. 4) If lung parenchyma of the basal segments is evaluated unlikely to improve gas exchanging function, lobectomy is recommended. In our case no 4, left lower lobectomy was performed because the lung parenchyma was believed to be extensively destroyed which was later pathologically proved to be a big intrapulmonary hematoma.

Dividing shunt without removing lung parenchyma is not recommended as the lung is left non-functioning and it has been reported to be a cause of postoperative ischemic lung changes or subsequent pulmonary complications[3]. Evaluation of structural changes in intrapulmonary arteries exposed to systemic pressure correlated with the diminished gas exchange function (before operation and after the anastomosis) is a practical clue to decide whether anastomosis of blood vessels or lung resection should be done[11]. For the same reason, evaluation or the benefit of anastomosis surgical procedure over lung resection, careful study of lung ventilation perfusion scan is also believed to be a significant tool[12].

SUMMARY

We have described eight cases of PALLADA. Preoperative diagnosis and hemodynamic confirmation were made in all cases except the first case. Surgical correction by anastomosing the abnormal artery to the existing pulmonary artery is recommended. Lobectomy or division of the anomalous shunt is also a choice of treatment in some cases. No operative complication occurred.

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การผ่าตัดแก้ไขความผิดปกติแต่กำเนิดของเส้นเลือดของปอดซ้าย, พัลลาดา ในผู้ป่วย 8 ราย

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ผู้เขียนรายงานผู้ป่วยผิดปกติแต่กำเนิดของเส้นเลือดปอดซ้าย ที่เรียกว่า พัลลาดา จำนวน 8 ราย ทุกรายได้รับการวินิจฉัย และการยืนยันโรคด้วยการฉีดสารทึบรังสีก่อนผ่าตัด ยกเว้นในผู้ป่วยรายแรก ผู้เขียนเลือกใช้วิธีการผ่าตัดย้ายเส้นเลือดที่ผิดปกติ เข้าไปต่อกับเส้นเลือดแดงพูลโมนารีเพื่อเป็นการรักษา นอกจากในบางรายจำเป็นต้องผ่าตัดเอาปอดออกทั้งกลีบ หรือใช้วิธีตัดเส้นเลือดผิดปกติออก ไม่พบอาการแทรกซ้อนจากการผ่าตัด

คำสำคัญ : ความผิดปกติแต่กำเนิดของเส้นเลือดปอด, พัลลาดา, ป็ซูโดซีเควสเตรชั่น

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