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Case Report

Congenital anomalous/aberrant systemic artery to pulmonary venous fistula: Closure with vascular plugs & coil embolization

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ABSTRACT

A 7-month-old girl with failure to thrive, who, on clinical and diagnostic evaluation [echocardiography & CT angiography] to rule out congenital heart disease, revealed a rare vascular anomaly called systemic artery to pulmonary venous fistula. In our case, there was dual abnormal supply to the entire left lung as[1] anomalous supply by normal systemic artery [internal mammary artery][2] and an aberrant feeder vessel from the abdominal aorta. Left Lung had normal bronchial connections and normal pulmonary vasculature. The fistula drained through the pulmonary veins to the left atrium leading to 'left–left shunt'. Percutaneous intervention in two stages was performed using Amplatzer vascular plugs and coil embolization to close them successfully. The patient gained significant weight in follow up with other normal developmental and mental milestones.

1. Introduction

After Huber in described for the first time in 1777 about an anomalous pulmonary artery arising from the aorta, several reports of an abnormal vascular supply to the lung were published. Connections between the systemic and pulmonary systems are rare conditions, either congenital or acquired diseases.[1,2]

Here, we report a unique rare case of a congenital systemic artery to pulmonary vein AV malformation/fistula in a small infant which successfully managed with percutaneous intervention. In this condition involved, lung has normal bronchial connections and pulmonary arteries which distinguish it from pulmonary sequestration.

2. Case report

A 7-month-old girl was referred by a General Practitioner with failure to thrive with suspected congenital heart disease for an echocardiography. Anthropometric examination revealed weight of 4 kg [Expected: 8 kg; 5th percentile] and a length of 62 cm [Expected: 68 cm; <75th percentile].

On examination, there was a hyper dynamic pulse at the rate of 102/min and blood pressure measured with appropriate size cuff of 82/38 mmHg. There was no cyanosis or clubbing. Also there were no signs of heart failure.

Cardiovascular examination revealed forceful left ventricular apex impulse with no thrill or other abnormal palpable

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heart sounds. On auscultation there was Grade III/VI continuous murmur over left infra-clavicular and apical regions with the same intensity over the entire back. Hence, the provisional clinical diagnosis of patent ductus arteriosus or large AV malformation was made.

The complete blood profile was normal except mild anemia with hemoglobin of 10.6 g%. The ECG showed left ventricular hypertrophy with left atrial enlargement. Chest X-ray showed mild cardiomegaly with increased vascular markings in the entire left lung field. Echocardiography [Fig. 1a–f] revealed the reversal of flow in the aorta suggestive of hyper dynamic circulation in abdominal aorta in a subcostal view. Also, an aberrant vessel at the level of celiac trunk demonstrating continuous flow could be traced up to the level of the diaphragm. Intra-cardiac anatomy was normal without any shunt lesions. But there was left atrial and left ventricular dilation for her age but a normal ejection fraction. Also Doppler velocities in the left sided pulmonary veins were conspicuously increased. In suprasternal view, left subclavian artery was dilated with torrential flow.

The diagnosis of the case was not clear hence CT angiography was done and 3D volume rendering images were reconstructed using 'Siemens Leonardo DA Vinci' workstation which revealed [Fig. 2a–c] anomalous systemic feeders from the branches of left subclavian [left internal mammary artery] and axillary arteries and an aberrant vessel from the upper abdominal aorta with drainage into the left atrium via left pulmonary veins. There was normal pulmonary arterial and pulmonary venous supply to both the lungs. The left sided pulmonary arteries were hypoplastic and left pulmonary veins were dilated and hypertrophied. The diagnosis of left systemic artery to pulmonary vein arterio-venous malformation was made.

The patient was subjected to cardiac catheterization and angiography for further confirmation of such a rare diagnosis.

Cardiac catheterization was done via trans-femoral approach using 4 Fr. femoral sheath and catheters. The oxymetric and pressure data revealed high cardiac output without any shunt lesion. Angiography of arch of aorta revealed enlarged left subclavian artery giving origin to large aberrant feeder [Fig. 3a–f]. Abdominal aortogram revealed an aberrant vessel originating from the thoracic aorta at the level of celiac axis piercing diaphragm. Both aberrant vessels created a vascular sponge in entire left lung with rapid drainage into pulmonary veins and left atrium.

This case was discussed with vascular and Cardiothoracic Surgeons, Interventional Radiologist, and Cardiac Anesthesiologist. In view of the small size of the child and a vascular nature of AV fistula, the risk of surgery was more and hence percutaneous intervention was planned in stages to improve growth and reduce hyper dynamic circulatory load of left ventricle to prevent heart failure.

Hence, we decided to stage the procedure in view of limitations of contrast volume. The left internal mammary artery was cannulated using 6 Fr. Judkin's right guiding catheter trans-femorally. There were three feeders arising from left internal mammary artery which were closed with embolization of 8 mm, 12 mm and 14 mm Amplatzer vascular plugs [AVP; AGA Medical, Golden Valley, MN, USA] [Fig. 4a–f]. The post procedure course was uneventful except for mild anemia [Hb – 9.8 g%] hence whole blood was transfused as per weight. In the second stage after 2 weeks, the abdominal aorta feeder

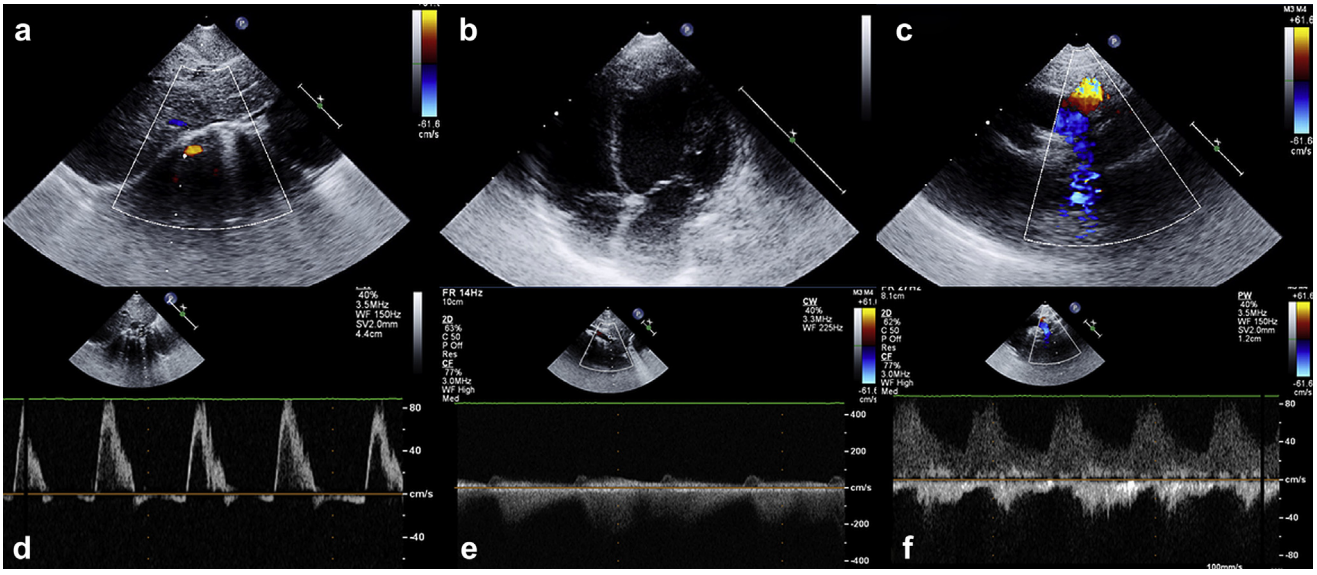

Fig. 1 – a: Subcostal echocardiography to assess Situs Solitus revealed an aberrant vessel with mosaic color pattern with continuous flow pattern (Doppler) originating from the upper abdominal aorta which could trace up to the diaphragm. b: Four chamber view showing dilated left ventricle and left atrium without any evidence of shunt. c: Suprasternal view demonstrated dilated left subclavian artery with turbulent flow. d: Pulse wave interrogation of abdominal aorta which showed diastolic runoff suggestive of hyper dynamic circulation. e: Continuous wave interrogation of the aberrant vessel showed continuous flow with wide pulse pressure. f: Pulse wave interrogation of left subclavian artery showed a continuous flow pattern with diastolic runoff suggestive of hyper dynamic circulation.

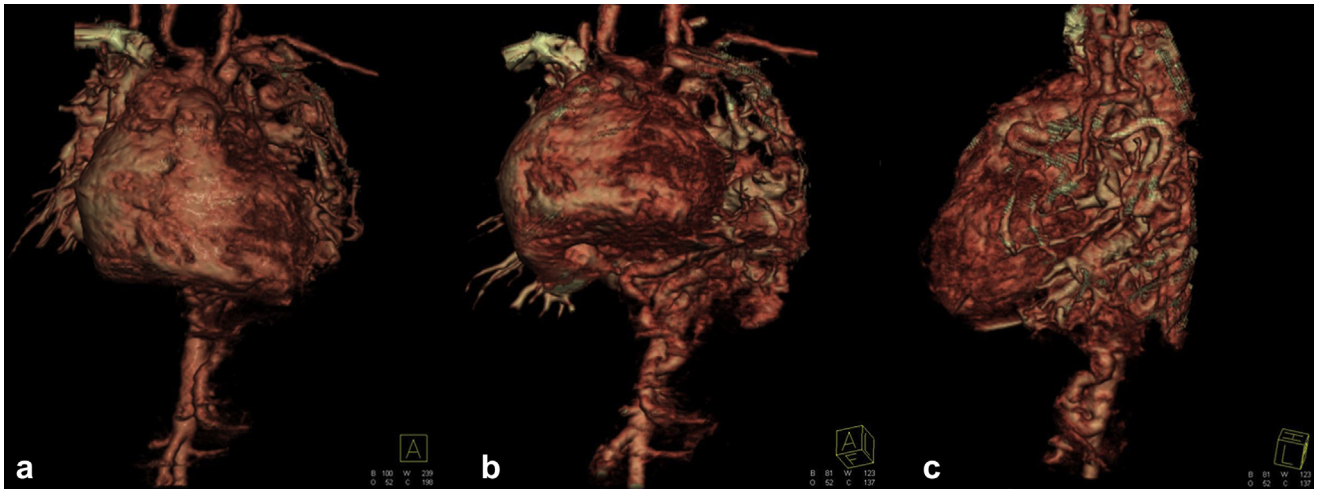

Fig. 2 – a–c: CT angiography with 3D volume rendering showing aberrant vessels originating from the left subclavian artery and abdominal aorta to the entire left lung which drained into the left atrium via normal pulmonary veins.

vessel [inferior phrenic artery] was calculated with 6 Fr. Renal guiding catheter [Fig. 5a–c] and 10 mm Amplatzer vascular plug was embolized with successful closure. The axillary artery feeder was ultimately closed with embolization of 4 × 8 mm Flipper Detachable Embolization Coils (Cook Cardiology, Bloomington, Indiana, USA) using diagnostic 5 Fr. Judkin's right coronary catheter [Fig. 5d–f].

Access site was closed using manual compression and using ultrasound and Doppler study was done every time to confirm hemostasis without complication.

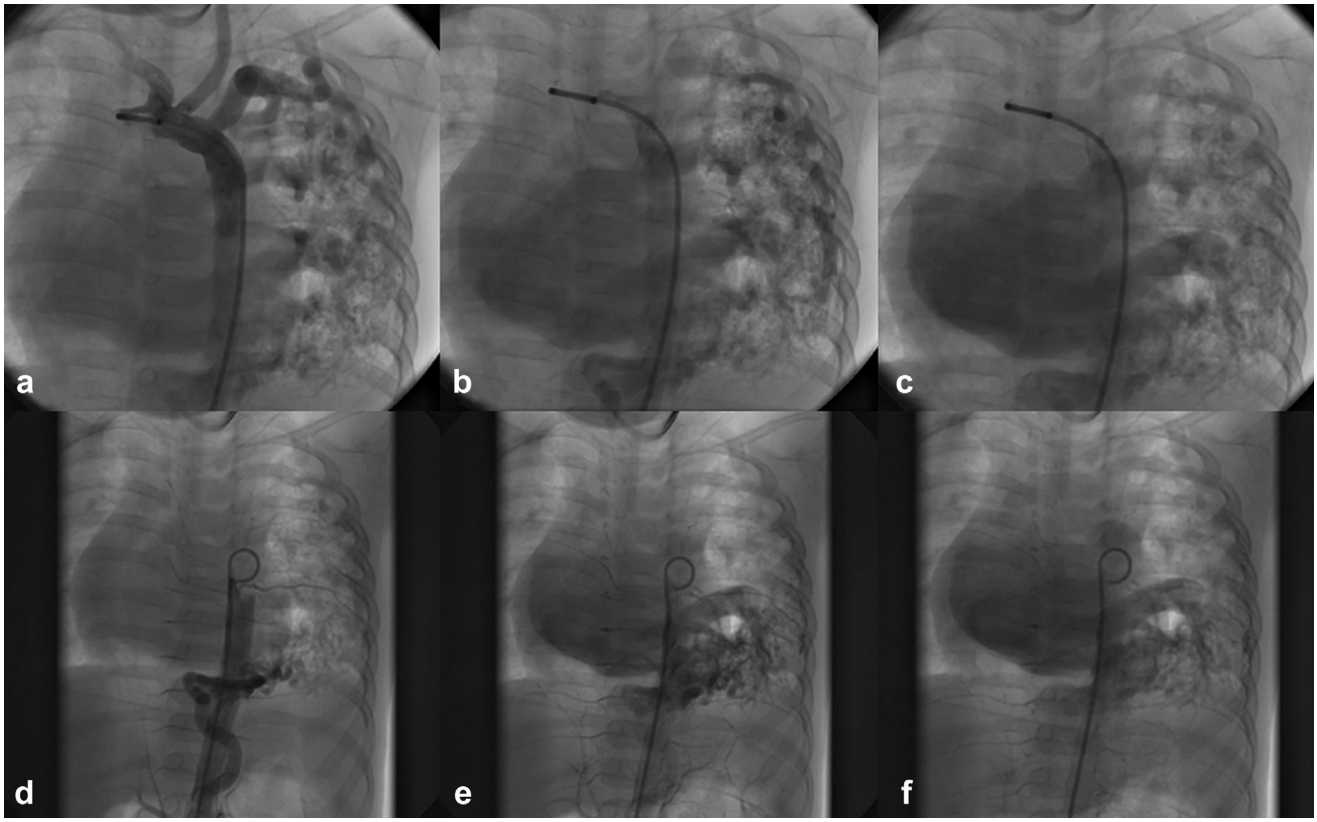

Fig. 3 – a–c: Arch and thoracic aortic angiography using a pigtail catheter showed large aberrant feeder vessels originating from the left subclavian artery and supplying the left lung. Post arterial phase contrast created vascular sponge followed by venous phase showing dilated pulmonary veins draining into the left atrium. d–f: Abdominal angiography showing the aberrant feeder vessel origin from the upper abdominal aorta piercing left hemi-diaphragm and supplying left lower lobe of lung creating similar post arterial vascular sponge and draining into left lower pulmonary vein to the left atrium.

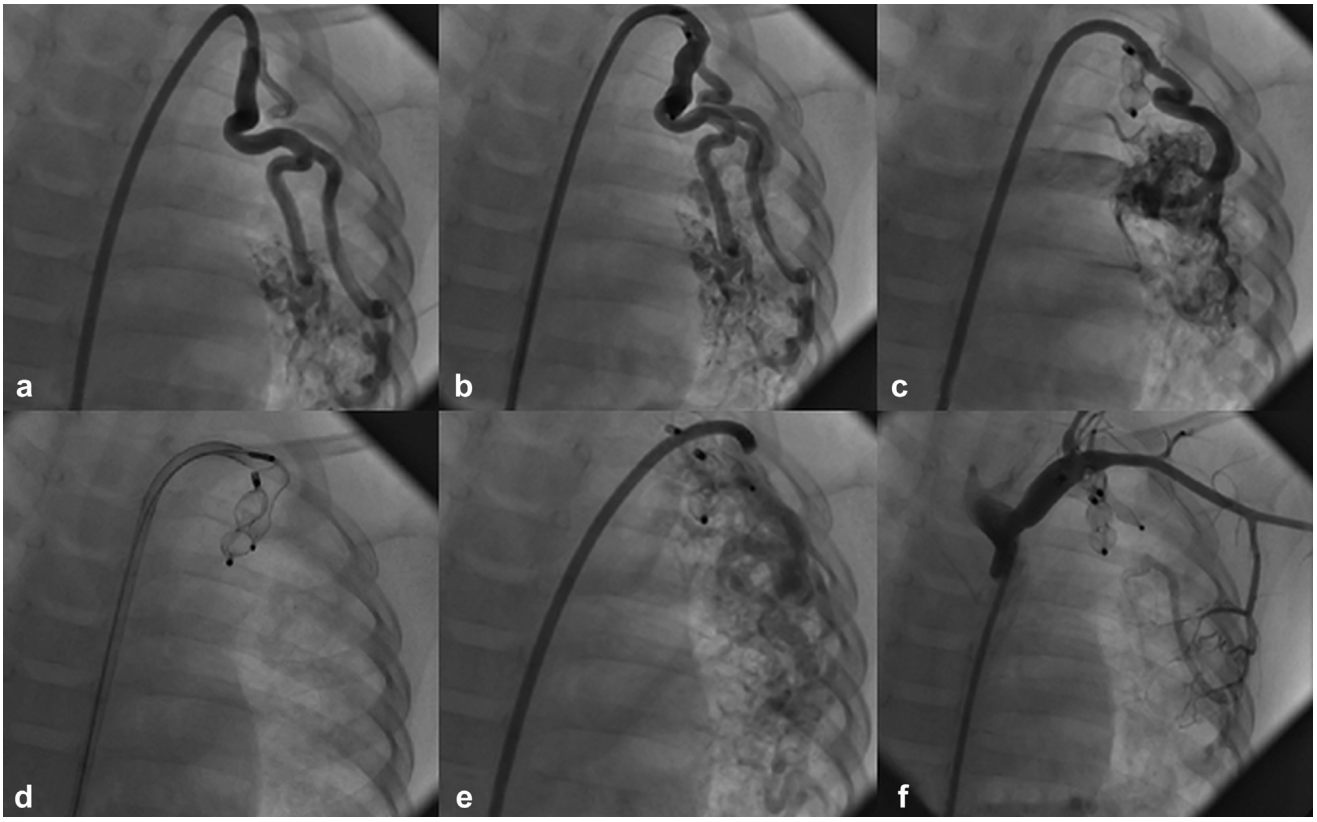

Fig. 4 – a: 1st intervention showing Judkin's right guiding catheter across left subclavian artery and the aberrant systemic artery and angiography check showing pacified medial feeder vessel. b: Deployment of Amplatzer vascular plug into the aberrant feeder systemic artery and guiding catheter engaged into middle aberrant feeder and angiography check showed supplying to left middle lobe of the lung. c: Deployment of Amplatzer vascular plug into middle feeder. d: Guiding catheter now into a lateral feeder vessel opacifying lateral portion of lung adjacent to the chest wall. e: Largest Amplatzer vascular plug was deployed into the lateral feeder vessel. f: Final angiogram using diagnostic Judkin's right catheter which revealed complete closure of left subclavian aberrant feeder systemic arterial supply. It also showed aberrant supply to the left lung from the axillary artery across the chest wall.

The patient tolerated this procedure as well, under General anesthesia and post procedure outcome was uneventful. The patient was discharged in stable condition with palpable left upper limb pulse. The 6-month follow up completed and patient has gained 2.5 kg of weight with improvement in appetite and interest in surroundings. Also, repeat CT angiography with 3D volume rendering images revealed complete closure of all aberrant vessels with normal broncho-vasculature to the left lung [Fig. 6a, b].

3. Discussion

In the normal lung, the only communication between the systemic and pulmonary arterial systems is the connections between the bronchial and pulmonary artery systems that occur at the respiratory bronchioles, where pulmonary and bronchial capillaries freely anastomose.[3]

The absence of parenchymal abnormalities and a normal bronchial supply clearly distinguishes systemic arterial supply without sequestration from true sequestration. Systemic arterialization of the lung without sequestration corresponds to type I of the sequestration complex, which was first described by Pryce et al[4] They are characterized by various features as below in Table 1.[5–7]

Lawler and Fishman have defined systemic supply to the lung as abnormal communications of broncho-pulmonary vascular complex.[8] They are classified as

1. With or without congenital heart disease.
2. Systemic supply through pulmonary artery.
3. Systemic artery supply can be through a normal artery or aberrant artery.
4. Venous drainage may be to pulmonary veins or to systemic veins.
5. It could be sole supply or partial supply to the lungs.
6. Surrounding pulmonary parenchyma may be normal or sequestrated.

Depending upon above classification, there are three types of intrapulmonary arterio-venous fistulas seen clinically [Table 2].[8,9] The normal pulmonary artery branches may or

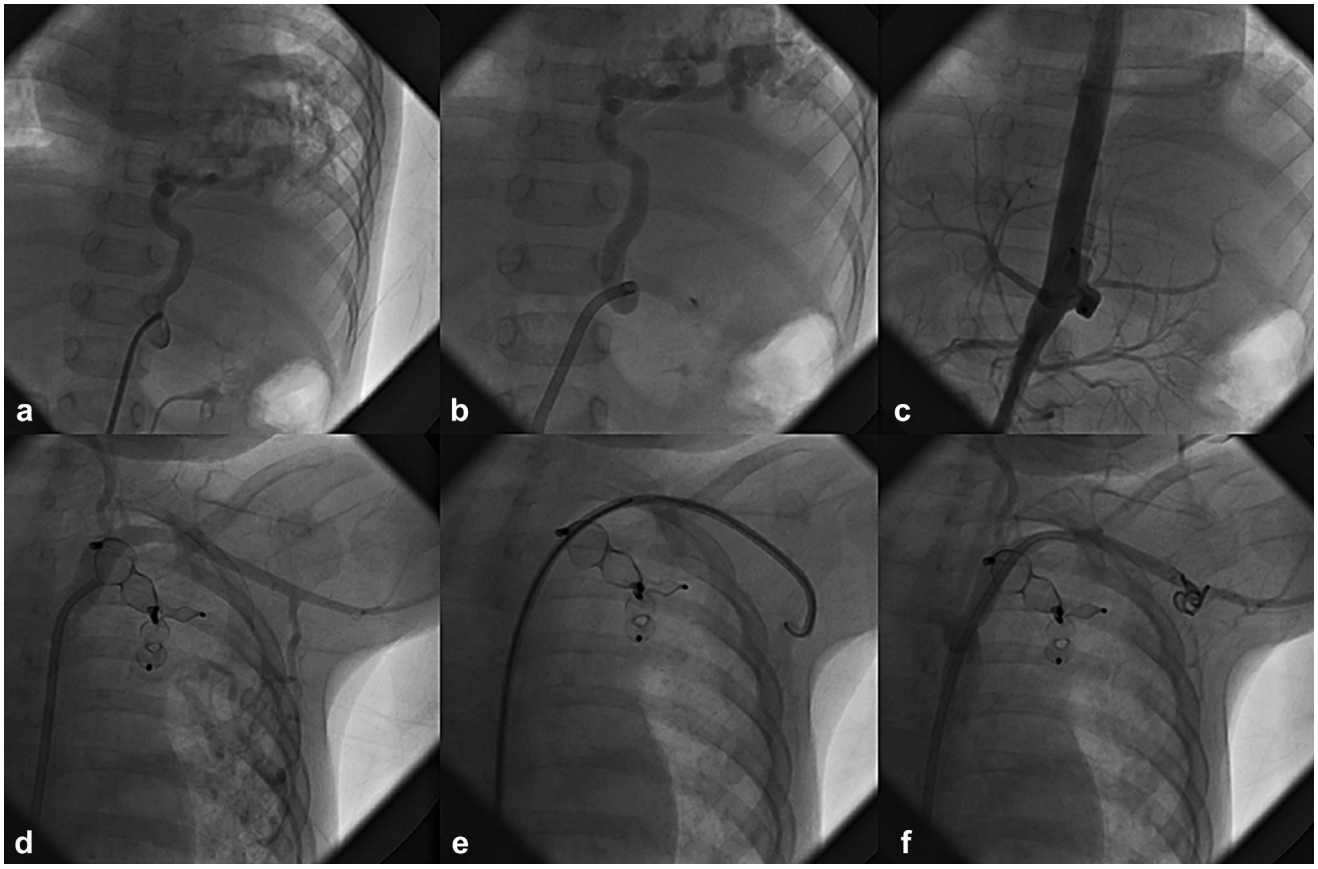

Fig. 5 – a and b: 2nd stage intervention showing angiography using renal guiding catheter of abdominal aberrant feeder systemic artery opacifying left lower lobe of lung. c: Deployment of Amplatzer vascular plug into the abdominal aberrant feeder vessel and angiography check demonstrated near closure with faint streaks of contrast across the device. d and e: Closure of lest feeder vessel originating from the axillary artery using detachable coil which was deployed using deep engagement diagnostic Judkin's' right catheter. f: Final angiography showing complete closure of all aberrant systemic supply to the left lung with previously deployed Amplatzer vascular plugs and a detachable coil with patent left subclavian and axillary artery.

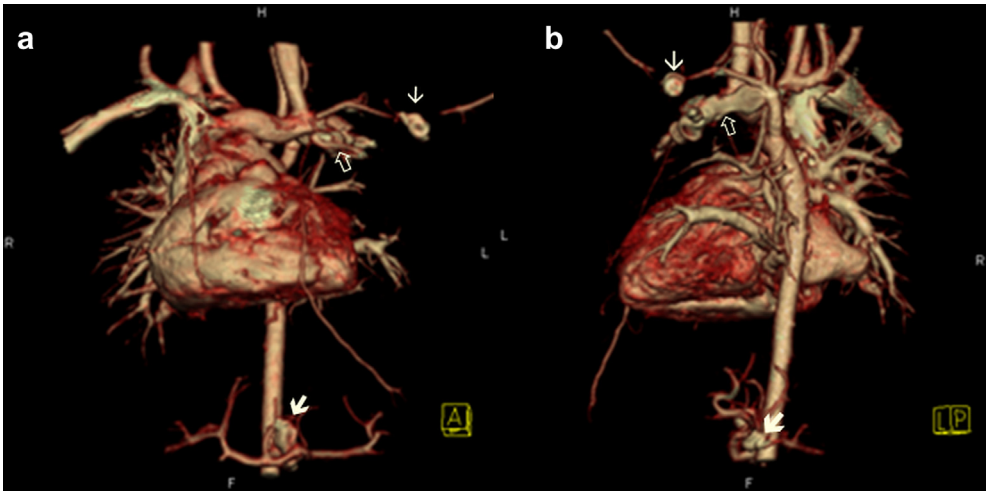

Fig. 6 – a and b: CT angiography with 3D volume rendering in front and back views showing complete closure of all aberrant feeders from left subclavian, axillary arteries and from abdominal aorta with Amplatzer vascular plugs and a detachable coil in situ. It also showed normal vasculature to the left lung and patent left subclavian and axillary artery.

Table 1 – Definition of types of sequestration complex.

True broncho-pulmonary sequestration	Characterized by systemic arterial supply of lung parenchyma with absence of normal bronchial and pulmonary arterial supply. A] Intra lobar – the sequestrated lung parenchyma may be included in the substance of the lobe. B] Extra lobar – the sequestrated lung parenchyma may be anatomically distinct from the remainder of the lung.[5]
Pseudo sequestration	The combination of systemic arterial supply to the lung with normal bronchial connections but absent normal pulmonary arterial supply.[6]
Systemic to pulmonary vein fistula – non sequestration	It is supplied by the aberrant artery, has no parenchymal or bronchial abnormalities and there is a normal connection with the bronchial tree.[7]

Table 2 – classification of arterio-venous fistula/malformation of lungs.

Type	Characteristics
1. Venous – Systemic fistula Fistulous connection between a normal pulmonary arterial branch and the pulmonary venous system.	Causes cyanosis as venous blood is shunted into the systemic circulation.
2. Arterial–Systemic fistula 2A Normal systemic artery (e.g., bronchial, internal mammary, intercostal) to pulmonary veins. 2B An aberrant systemic artery arising from the descending aorta to pulmonary veins.	1. Both are acyanotic lesions. 2. In type 2B, pulmonary parenchyma surrounding the fistulous connection may be normal or sequestered.

may not be present. Sometimes the lung parenchyma is vascularized by both the anomalous systemic artery and the normal pulmonary artery branches.[5]

Our case had a combination of two types [2A and 2B] in a single patient as upper fistula had a feeder vessel from internal mammary artery and lower fistula had a feeder vessel arising from descending thoracic aorta.

Congenital anomalous systemic artery to pulmonary venous arterio-venous malformation/fistula is a left–left shunt without lung sequestration and is an extremely rare anomaly of cardiovascular anatomy.[9] This condition differs from 'typical sequestration syndrome' though there is abnormal or aberrant systemic arterial supply as there is normal bronchial and pulmonary artery supply [Dual supply].

The characteristics of this condition are as described in Table 3[1,2,7,9–21]: Multi-detector CT and three-dimensional volume rendering can deliver high-quality angiograms that show multiple and complex arterio-venous malformations. It creates a road map before the endovascular procedure as we planned in our case.[8]

Campbell et al [1962],[12] Scott et al [1968][13] and Ernst et al [1971][14] reported systemic artery to pulmonary vein fistula with left–left shunts earlier. Later on other isolated case reports of systemic artery to pulmonary vein fistulas, mostly congenital in origin was published which are summarized in Table 4. We reviewed 16 cases of anomalous/aberrant systemic to pulmonary venous fistula in the literature.

Wolf et al detected aberrant systemic artery–pulmonary fistula using 2D echocardiography with contrast injection through the umbilical vein.[11] Robida et al suspected aberrant systemic artery–pulmonary venous fistula arising from the

Table 3 – characteristic features of anomalous systemic artery to pulmonary venous fistula.

1. Fistulas from systemic arteries to the pulmonary vein may be congenital or acquired. But congenital variety being most common diagnosed from newborn to 55 years of age.
2. Hemodynamically significant left to left shunts are associated with a continuous murmur, bounding peripheral pulses and left ventricular enlargement.
3. Patients are not cyanotic clinically.
4. Atypical location of the continuous murmur, lack of evidence of increased pulmonary flow and evidence of localized lesions in pulmonary parenchyma should lead to a suspicion that a left to left shunt is present.
5. The bronchial tree, pulmonary artery branching and affected lung tissues are completely normal.
6. Enlarged feeders with significant shunt volume.
7. At cardiac catheterization left to left shunts are not associated with increased oxygen saturation in the pulmonary artery and may be clearly outlined with the use of arteriography.
8. Rapid passage of contrast into pulmonary vein and left atrium.
9. The same vein also drained the pulmonary circulation.
10. Except one fistula most of them were located in the right or left lower lobes with left lower lobe being most common site.
11. Congestive heart failure or vascular compression or asymptomatic with only murmur and sometimes during imaging of chest were common mode of presentation.

Table 4 – Review of the literature published to date of anomalous systemic artery to pulmonary venous fistula.

Sr. no.	Author	Year of publication	No of cases	Age & sex	Origin of systemic aberrant vessel	Site of drainage of lung	Management
1	Campbell et al[12]	1962	2	35 yrs/male	Abdominal aorta	Right lower lobe	Lobectomy
				14 yrs/male	Descending thoracic aorta	Left lower lobe	Surgical ligation
2	Scott et al[13]	1968	2	6.2 yrs/female	Descending aorta	Posterior basal segment of left lower pulmonary lobe	Surgical ligation and lobectomy
				7.6 yrs/female	Post Potts' descending aorta-pulmonary anastomosis [TOF], descending aorta was inadvertently anastomosed to a pulmonary vein	Left lung	Surgical ligation and intra-cardiac repair of Tetralogy of Fallot's
3	Ernst et al[14]	1971	1	3 yrs/male	Abdominal aorta	Right lower lobe	Surgical Ligation
4	Varma et al[2]	1971	1	7 yrs/male	Descending aorta below diaphragm	Right lower lobe	Surgical ligation
5	Currarino et al[9]	1975	3	2 ½ month/male	From aorta at the level of celiac axis	Posterior basal segment of right lower pulmonary lobe	Surgical ligation
				3 yrs/female	From thoracic aorta at the level of diaphragm	Posterior basal segment of right lower pulmonary lobe	Surgical ligation and partial lobectomy
				5½/male	From thoracic aorta above level of diaphragm	Left lower lobe	Treatment refused by parents
6	Masaoka et al[18]	1978	1	16 yrs/male	Descending aorta [D8 level]	Left lower lobe	Surgical ligation
7	Wolf et al[11]	1985	1	Newborn	Low thoracic aorta.	Postero- basal segment left lower lobe	Surgical ligation
8	Robida et al[15]	1992	1	6 yrs/female	Near the celiac axis, piercing the right hemi-diaphragm	Right Lower lobe of lung	Surgical ligation
9	Brühlmann et al[7]	1997	1	51 yrs/male	Lower thoracic aorta	Left lower lobe	Coil embolization using multiple coils
10	Chabbert et al[19]	2002	1	17 yrs/male	An aneurysmal artery with partial thrombosis arising from the aorta above the celiac trunk	Basal segments of the right lower lobe	Coil embolization using multiple coils
11	Baek et al[17]	2006	1	17 yrs/male	Descending aorta	Lower part of the left lung	Surgical ligation of anomalous artery.
12	Kosutic et al[10]	2007	1	3 month	Two major aberrant arteries with separate origins came from the descending thoracic aorta	Right upper lung lobe	Coil embolized, and the other aberrant artery spontaneously closed after cardiac catheterization
13	Shebani Suhair et al[20]	2007	1	Newborn [7th day]	Descending thoracic aorta	Right upper lung lobe	Surgical ligation
14	Komaki et al[16]	2008	1	55 yrs	Left lateral thoracic artery from the left subclavian artery	Lingular division of left lung	Refused treatment
15	Wong et al[1]	2008	1	10 month/male	Descending thoracic aorta at the level of T8	Lower lobe of left lung	Surgical ligation and lobectomy
16	Singhi et al[21]	2012	2	30 days/male	Descending thoracic aorta	Left lung	Ligation of the anomalous vessel
				90 days/?	Two collaterals arising from the abdominal aorta	Left lower lobe	Amplatzer vascular plug and Gianturco coils

celiac trunk based on Doppler waveform showing retrograde diastolic flow in the proximal abdominal aorta similar to our case.[15]

Wong et al[1] diagnosed anomalous systemic arterial supply to the left lung draining through normal pulmonary veins using 64-slice computed tomographic contrast-enhanced angiography, as in our case while Kosutic et al[10] reported a 3-month-old infant presented with vascular tracheal compression and congestive heart failure which was diagnosed using angiography. Komaki et al reported a case of 55 years old male who was diagnosed during a routine medical checkup with nodular left lung opacity.[16]

Currarino et al treated them surgically by partial lobectomy with ligation of feeder vessels. They had a high incidence of morbidity [infection mainly] and mortality in the pre - intervention era.[9] Wan Ki Baek et al performed a simple division of the anomalous artery has brought a satisfactory result as left lower lobe had dual supply. The presence of normal pulmonary artery to the diseased segments of the lung was hard to define by means of pulmonary angiography, largely because of flows from the anomalous artery.[17]

W. Brühlmann et al described the first trans-arterial therapeutic embolization of systemic arterialization of the lung without sequestration.[7]

Geyik et al described a case of systemic artery to pulmonary artery malformation where the systemic artery feeder was similar as in our case but drainage was in to pulmonary artery arterial.[3] Endovascular intervention with the help of coils, glue and detachable balloon was used to occlude successfully.

But there were some issues before intervention as below

1) Small size of patient and limited use of contrast. 2) Choice of hardware. 3) Choice of percutaneous devices for closure. 4) Risk of bleeding. 5) Anesthesia related and post procedure general care.

Amplatzer® vascular plug (AVP; AGA Medical, Golden Valley, MN, USA) used for trans-catheter embolization in the peripheral vasculature and occlusion of abnormal vessel communications[22] with reported technical success rate of 100%. The only disadvantage is need for relatively large catheters, 5 Fr. at least.[9] In small children as in our case, there is a need for evaluation of femoral artery using ultrasound and Doppler study before procedure to measure the size of vessel and post procedure to ensure normal flow without any complication.

Controlled-release coils are well-known devices consisting of a metallic coil with fibers attached to increase their occlusive effect. The advantage of these coils that can be implanted via 4 Fr. Catheters are used as a delivery system attached to the coil by a threaded mechanism.[23,24]

Girona et al described the successful closure of 51 vascular fistulas in 30 patients aged from 6 days to 28 years (mean, 8.4 years) by percutaneous embolization. They used coils for smaller fistulas and vascular plugs for closure of larger fistulas as in our case.[24]

4. Conclusions

This type of AV malformation is rare and till now only few cases being reported to our knowledge similar to our case. In our case we used vascular plugs and a coil together first time to close which was a complex intervention in a small sized infant without any complications. These conditions carries a higher mortality left untreated. Advances in the percutaneous interventions and devices lead to successful closure of the defect in high risk surgical cases. Further follow up of the case in future will answer the long term prognosis and management of such cases.

Conflicts of interest

All authors have none to declare.

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