

# Anomalous Origin of Left Coronary Artery from The Pulmonary Artery of Left Main Coronary Artery (ALCAPA) In A Pediatric Patient: A Rare Case Report

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## ABSTRACT

**Background:** The anomalous origin of the left coronary artery (LCA) from the pulmonary artery (ALCAPA) is a rare congenital anomaly due to abnormal division of the conotruncus into the aorta and pulmonary artery or persistence of the pulmonary bud with involution of the aortic bud that forms the origin of the coronary artery on embryogenesis. Only a small portion of cases survive more than two months of age. The survived cases can appear as asymptomatic, symptomatic, sudden cardiac death of adulthood ALCAPA. **Case Description:** We present an 8-year-old boy with symptomatic adulthood type ALCAPA. Preliminary chest x-ray examination found cardiomegaly. Unspecific findings were found on Echocardiography including fistula right coronary artery (RCA) and left coronary artery (LCA), with mitral regurgitation, moderate tricuspid regurgitation, and cardiomyopathy. Catheterization confirmed the left coronary artery from the pulmonary artery (ALCAPA) of the left main coronary artery (LMCA) origin. Additional Cardiac computed tomography angiography (CTA) demonstrated the LM originates from the pulmonary trunk accompanied by the presence of collaterals between the RCA and LAD and dilatation and turbulence of the RCA. **Conclusion:** Early diagnosis and prompt surgical intervention can give excellent results and lead to gradual recovery.

**Keywords:** anomalous left coronary artery; pulmonary artery; Catheterization; ALPACA

## INTRODUCTION

The anomalous origin of the left coronary artery (LCA) from the pulmonary artery (ALCAPA) is a rare coronary heart disease affecting heart function profoundly.<sup>1</sup> The incidence of ALCAPA is 1 in 300,000 live births.<sup>2</sup> This congenital anomaly has a high mortality rate in the first year of life.<sup>1-2</sup> If untreated, it has a mortality rate approaching 90% in the first year of life. Only 10% to 15% of cases have extensive intercoronary collaterals with RCA that survive into adulthood.<sup>2,3,4</sup>

The common clinical course of the disease is heart failure and significant mitral valve insufficiency, which usually develops at one to two months of age when pulmonary vascular resistance falls.<sup>5,6</sup> Symptoms that often appear are irritability, eating difficulty, tachypnea, diaphoresis and tachycardia.<sup>7,8</sup> However, in some cases good collateral blood supply from the right coronary artery as mentioned above may be sufficient to survive until adulthood with relatively minor symptoms. Symptoms range from fatigue, dyspnea, malignant ventricular arrhythmias resulting from myocardial scarring or chest pain, and exercise intolerance to sudden cardiac death due to acute ischemia during exercise.<sup>9,10</sup> Here we report childhood onset anomaly of left coronary artery origin from the left main coronary artery pulmonary artery presenting with mild symptoms.

## CASE REPORT

An 8-year-old boy was referred from a regional hospital with suspected congenital heart disease, the patient presented a chief complaint of more easily fatigue when more activity.

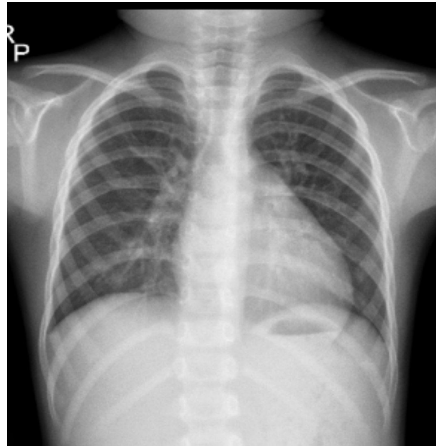
No shortness of breath, tachycardia, retrosternal pain, excessive sweating, bluish, and medical history of syncope. On physical examination, heart rate was 96 per minute, oxygen saturation was 95 % and all peripheral pulses were equal and well felt. The capillary refill time was 3 seconds. The respiratory rate was 20 times per minute with features of mild respiratory distress. Heart sound was a murmur systolic parasternal line IV grade III/VI. There were no signs of cardiac failure. Breath sounds were normal.

Chest x-ray was suggestive of cardiomegaly with a cardiothoracic ratio of 53% (**Figure 1**). An electrocardiogram (ECG) was suggestive of regular sinus rhythm (**Figure 2**). An Echocardiography performance revealed atrial situs solitus, normal systemic and pulmonal vein drain, AV-VA concordant, LAE/LVE, no PDA, no VSD, no ASD, left aortic arch, no CoA, mild MR ec prolapse PMVL, moderate TR, fistula coronary artery RCA, fistula LCA. There was mild mitral regurgitation, moderate tricuspid regurgitation, and cardiomyopathy (**Figure 3**).

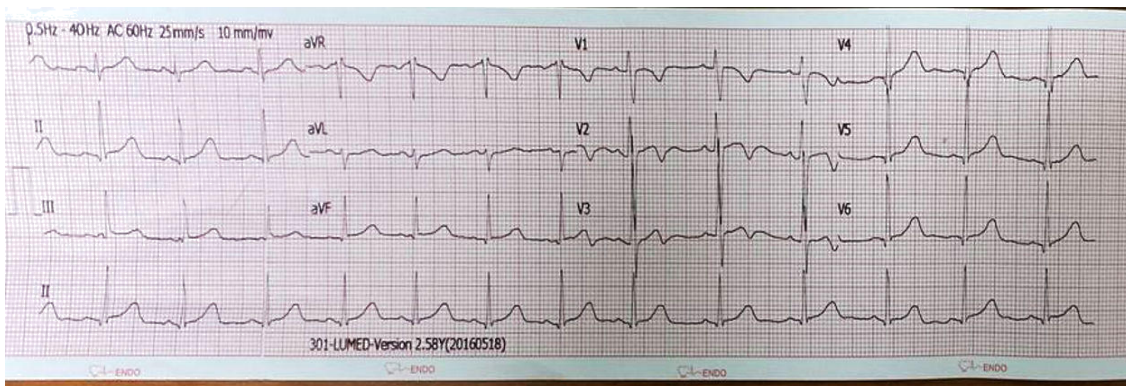
The paraclinical diagnostic methods exhibited a fistula right coronary artery (RCA) and left coronary artery (LCA), with mitral regurgitation, moderate tricuspid regurgitation and cardiomyopathy and planning to heart catheterization with KIV fistula occlusion using ADO II all size. When doing catheterization find this case as anomalous origin of left coronary artery from the pulmonary artery (ALCAPA) of the left main coronary artery (LMCA) origin, enlarged and tortuous right coronary artery (RCA) (**Figure 4**).

Cardiac computed tomography angiography (CTA) examinations were performed either on Siemens Somatom Flash CT scanner using retrospective ECG gating (**Figure 5**) demonstrated the LM originates from the pulmonary trunk

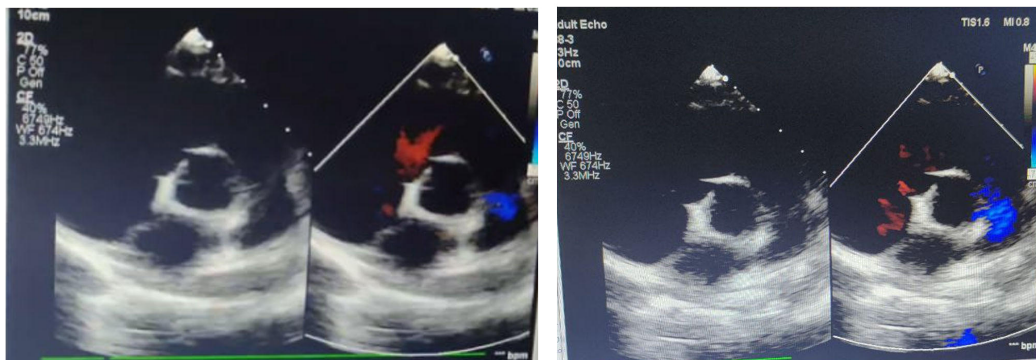
accompanied by the presence of collaterals between the RCA and LAD as well as dilatation and turbulence of the RCA suggesting anomaly of the left coronary artery from the pulmonary artery, Calcified plaque on LAD, LCX, and RCA.



**FIGURE 1:** Rontgen Thorax AP Cardiomegaly (31/12/2021).



**FIGURE 2:** Electrocardiographic examination (05/01/2022).



**FIGURE 3:** Echocardiography Examination Results showed fistula right coronary artery (RCA) and left coronary artery (LCA), with mitral regurgitation, moderate tricuspid regurgitation and cardiomyopathy.



**FIGURE 4:** Pediatric Cardiac Catheterization Result ALCAPA of LMCA origin, Enlarged and tortuous RCA (06/01/2022).



**FIGURE 5:** Cardiac computed tomography angiography (CTA) cardiac showed LM originates from the pulmonary trunk accompanied by the presence of collaterals between the RCA and LAD as well as dilatation and turbulence of the RCA suggesting anomaly of the left coronary artery from the pulmonary artery, Calcified plaque on LAD, LCX, and RCA.

## METHOD

Literature search procedure conducted by browsing the library in Online using databases: Pubmed, Google Scholar, and Science Direct with the keywords: "Anomalous" AND "Left Coronary Artery" AND "Pulmonary Artery" AND "Left Main Coronary Artery" AND "Pediatric". All case report about anomalous origin of left coronary artery from the pulmonary artery of left main coronary artery in pediatric patient are included, with inclusion criteria is case report in English language. Of all articles, 31 cases report articles were obtained related to anomalous origin of left coronary artery from the pulmonary artery of left main coronary artery, then 9 article were ALCAPA in pediatri.<sup>10-22</sup> All of 9 articles were appraised using the Joanna Briggs Institute Appraisal Checklist for Case Report and included in this evidence-based case report (**Figure 6**).

## LITERATURE SEARCH RESULTS

All articles used are presented in Table 1. Each article describes the clinical manifestations, diagnostic tools, corrective treatment and outcome.

## DISCUSSION

This case is a boy aged 8 years old with ALCAPA. Patient came with chief complaints of intolerance to activity. Based on previous report, only about 10%-15% of all ALCAPA cases can survive more than two months.<sup>2,7</sup> Based on its survival and clinical spectrum, ALCAPA is divided into infantile type (less than 12 months age), asymptomatic adult type, symptomatic adult type, and sudden cardiac death.<sup>3</sup> This case, report A 8-year-old boy diagnosed as ALCAPA of left main coronary artery (LMCA) origin.

Clinically, in adulthood type commonly present with signs of myocardial infarction, heart failure, mitral regurgitation, severe pulmonary hypertension, or sudden cardiac death.<sup>3</sup> Manifestations of ALCAPA widely vary depending on extensive coronary artery collateral circulation. Clinical manifestations in patients of non-infant type were atypical. Manifestations may appear as chest discomfort or incidental finding of a heart murmur, or misdiagnosed as coronary heart disease, myocarditis, or Patent ductus arteriosus (PDA).<sup>1,3</sup> Case reports of ALCAPA report respiratory distress (tachypnea or dyspnea or fast breathing) is the most common finding. Meanwhile in cardiac examination murmur frequently found. But, this additional heart sound may due to other associate pathology such as mitral regurgitation or dysfunction of other leaflet.<sup>9</sup> This case, the patient was found to be in a stable condition with minimal symptoms and a grade III/VI parasternal systolic murmur was obtained. In addition, ALCAPA may associate with others syndrome such as Scimitar syndrome, Berry syndrome, and Bland-White-Garland syndrome.<sup>2</sup>

On radiological examination, the most common features were cardiomegaly and pulmonary congestion.<sup>1,6</sup> A study showed the cardiothoracic ratio was from 0.62 to 0.68.<sup>1</sup> In this case also found cardiomegaly with a CTR of 53%. Enlargement of the heart may be caused by cardiomyopathy, either dilated or ischemic cardiomyopathy or ventricular hypertrophy as a complication of this condition.<sup>1,2</sup> One case report mesocardia with complete collapse of the right lung, but this ALCAPA associated with Scimitar syndrome.<sup>2</sup> In this case, chest x-ray was suggestive of cardiomegaly with cardio-thoracic ratio of 53%.

Electrocardiogram usually showed Q waves in anterior leads in 50% of patients and left ventricular hypertrophy in up to 28% of all cases.<sup>2,4</sup> In infantile type, abnormal Q waves with T wave inversion in leads I, avL, and V4-V6, especially in lead avL can be found.<sup>5,6</sup> However, the adult type rarely shows abnormalities on ECG examination.<sup>1</sup> Likewise in this case the ECG picture shows a regular sinus rhythm. Abnormal ECG finding common seen in ALCAPA associate with other abnormalities. Deep Q waves in V5/V6 and ST depressions in the lateral leads found in ALCAPA with mitral regurgitation and ventricular dilatation.<sup>9,7</sup>

On echocardiography, the infantile type often has left ventricular enlargement, reduced left ventricular systolic function, and moderate to large mitral valve.<sup>1</sup> Other possible finding were apical ventricular aneurysm, moderate to severe myocardium infarction (MI), dilated right coronary artery (RCA), coronary artery collateral circulation, and unable showing clear origin of LCA.<sup>7,9</sup> On Doppler examination may found narrow red shunting flow toward the probe at the left or posterior wall of pulmonary artery, and connection of LCA to pulmonary artery (PA) that possibly visualized through two-dimensional echocardiography. Fistula can be found as an initial finding in ALCAPA<sup>1</sup> as in this case. Kumar (2021) also found Coronary Artery-to-Pulmonary Artery Fistula as associated finding to ALCAPA.<sup>3</sup> In this case, on echocardiography found fistula RCA and LCA, with mitral regurgitation, moderate tricuspid regurgitation and cardiomyopathy.

Catheterization and angiography are the standard work up to diagnose ALCAPA.<sup>1</sup> On catheterization, left-to-right shunting can be found and pulmonary artery pressure can also be measured.<sup>3</sup> Angiography found the RCA arising from the aortic root with absence of LCA origin and after opacification of RCA, reverse flow in LCA and PA visualized through collateral circulations.<sup>1</sup>

Classically, ALCAPA is diagnosed by angiography or autopsy. But, by the development of cardiac computed tomography (CT) and magnetic resonance imaging (MRI) has allowed noninvasive evaluation of the coronary anatomy by direct visualization.<sup>3,5</sup>



In Cardiac CT angiography possibly revealed tortuous coronary arteries with many collaterals between the left and right coronary system.<sup>6</sup> Likewise, in this case, collaterals between the RCA and LAD were obtained with dilatation and turbulence of the RCA. Calcification was also found in LAD, LCX, and RCA. This calcification may be caused by fibroelastosis leading to fibrosis and scarring and subsequently induce calcium deposition.<sup>3,7</sup>

The definitive management for this condition is surgery. Early diagnosis and prompt surgical intervention can give excellent results and lead to gradual recovery of the myocardium.<sup>3</sup> ALCAPA with malignant arrhythmias can be treated with drug therapy, ICD implantation or catheter ablation. VT/VF with previous myocardial infarction was superior to ICD implantation compared to antiarrhythmics.<sup>3,8</sup> Most cases report describe underwent Tekauchi repair. Initially, coronary artery incised from pulmonary artery followed by arteriotomies of the ascending aorta and the Main Pulmonary Artery (MPA). The two orifices are stitched together side-to-side to create an aortopulmonary window. Through an adjacent pulmonary arteriotomy, the ostium of the left coronary artery is visualized arising off of the MPA. An intrapulmonary tunnel is constructed from the aortopulmonary window to the ostium of the left coronary artery.

The pulmonary arteriotomy is closed using a patch of homograft or pericardium.<sup>9</sup> Anastomosis made may varied depend on LCA origin. Button transfer with horizontal incision made at the left side of the ascending aorta at the same level of the LCA orifice with the collar of the button around the LCA orifice for the LCA orifice at the bifurcation of the main pulmonary artery and RPA. Anastomosis of the button to the aorta incision for LCA orifice from distal along the RPA from the bifurcation.<sup>8</sup>

Prognosis of ALCAPA is not clear because of rare cases. Eight cases reported were all survived with some complication such as baffle leak (38%), decrease systolic function (25%), mitral valve regurgitation (38%) and need reoperation (38%).<sup>10</sup> Another study of 9 cases found only one patient occurred dehiscence and needs reoperation.<sup>21</sup>

## SUMMARY

This case is an 8-year-old boy with mild clinical manifestations. Chest X-ray showed cardiomegaly and ECG within normal features. Echocardiography found right coronary artery (RCA) and left coronary artery (LCA) fistulas, with mitral regurgitation, moderate tricuspid regurgitation and cardiomyopathy. Catheterization confirmed the left coronary artery from the pulmonary artery (ALCAPA) from the left main coronary artery (LMCA). Additional Cardiac computed tomography angiography (CTA) showed LM originating from the pulmonary trunk accompanied by the presence of collateral between the RCA and LAD and dilatation and turbulence of the RCA. Multiple working diagnostic are required to confirm the diagnosis and detailed characterization of ALCAPA. Anastomoses LMA and Aorta is the main procedure to repair this abnormality. Outcome is not clearly stated, varied on some studies.

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**TABLE 1:** describes of the clinical manifestations, diagnostic tools, corrective treatment and outcome.

Authors (year)	Diagnosis and Associated Anomaly	Subject	Clinical Manifestations	Diagnostic Tools	Corrective Treatment	Outcome
Harrison et al (2022)	ALCAPA, mitral regurgitation, ventricular dilatation	M, 4 months	Progressive labored breathing, tachypnea, RR 80x/minutes, holosystolic murmur apex	ECG : deep Q waves in V5/V6 and ST depressions in the lateral leads; Transthoracic echocardiogram showed a severely dilated left ventricle (Z score +7.7; volumes measured by 5/6 area-length product) with moderate systolic dysfunction and ejection fraction of 42% (age adjusted Z score = -4.1), Catheterization and pulmonary artery angiogram.	Intramural midsection of the coronary artery and adherent to aorta, Annuloplasty	Re operation 15 days after first operation, discharge day 55 day after initial operation, left ventricular dilatation resolved, systolic function normal, EF 60%
Ginde et al, (2012)	ALCAPA, supravulvar pulmonary stenosis	Eight patients, age 5 week until 14.6 years, Gender not described	Consistent with Hearth Failure NYHA class I	Not describe	Tekauchi repair (Arteriotomies made in the ascending aorta and the Main Pulmonary Artery (MPA); The two orifices are stitched together side-to-side to create an aortopulmonary window; Through an adjacent pulmonary arteriotomy, the ostium of the left coronary artery is visualized arising off of the MPA; An intrapulmonary tunnel is constructed from the aortopulmonary window to the ostium of the left coronary artery;The pulmonary arteriotomy is closed using a patch of homograft or pericardium)	All were survived. Moderate stenosis in 2 and severe stenosis in 1 case. Three late survivors (38 %) had a baffle leak. Two patients (25 %) had decreased left ventricular systolic function and 3 (38 %) had developed at least moderate mitral valve regurgitation. Three survivors (38 %) required a reoperation for repair of mitral valve regurgitation, baffle leak, and main pulmonary artery (MPA) stenosis.
Zhang et al, (2016)	ALCAPA, moderate-severe mitral regurgitation, ventricular aneurysm	3 months to 11 years, 3 F and 7 M	Poor feeding, respiratory distress, and fussiness	Echocardiography (n = 10), computed tomography (n = 5), and coronary angiography (n = 4). Echocardiography showed mean left ventricular ejection fraction of 45% ± 10% and mean fractional shortening of 21% ± 7%. Severe mitral regurgitation was found in 4 patients. The left ventricular aneurysm at apex was found in 5 patients with the diameter ranging from 15 to 60 mm. During the operation, confirmed 2 types of LCA orifice: at the bifurcation of the main pulmonary artery and RPA in 3 patients, and more distal along the RPA from the bifurcation in 7 patients.	Two type surgical anastomosis: First, button transfer with horizontal incision made at the left side of the ascending aorta at the same level of the LCA orifice with the collar of the button around the LCA orifice for the LCA orifice at the bifurcation of the main pulmonary artery and RPA. Second, anastomosis of the button to the aorta incision for LCA orifice from distal along the RPA from the bifurcation Four patients with severe mitral regurgitation undergo annuloplasty. Two patients with large aneurysm undergo closing the aneurysmal collar with a Dacron patch and 2 patients with small aneurysm undergo longitudinally plicated without any incision and sutured using 3-0 Prolene with 2 small Dacron patches.	Two patients died due to cardiac failure in early postoperative period. Follow up ranged from 2 months to 6 years found mean left ventricular ejection fraction 67% ± 6% and mean fractional shortening 32% ± 3%. Among the 4 patients who underwent mitral valve repair, mitral regurgitation became mild. Others did not show mitral regurgitation.

Authors (year)	Diagnosis and Associated Anomaly	Subject	Clinical Manifestations	Diagnostic Tools	Corrective Treatment	Outcome
Kumar, (2021)	ALCAPA and Congenital Coronary Artery-to-Pulmonary Artery Fistula	3 months, M	Fast breathing, grade II/VI early systolic murmur at the left upper sternal border,	Transthoracic echocardiogram showed situs solitus and atrioventricular and ventriculoarterial concordance, left atrium and left ventricle were enlargement, dilated LM coronary artery, and a fistulous channel approximately 10 mm in length and 3 mm in diameter, flow reversal in the aortic arch. Coronary CT angiography (CCTA) showed dilated-tortuous LM arising from the left coronary sinus. The RCA was seen arising from main pulmonary artery (MPA); Dilated Left atrium and left ventricle.	Surgical ligation of LM-to-MPA fistula with direct reimplantation of anomalous RCA to aortic root.	The postoperative period was uneventful, and the patient was discharged
Rad et al, (2019)	ALCAPA, left atrial and left ventricular enlargement, severe mitral regurgitation	6 months and one-week-old infant (gender not described)	Tachypnea and tachycardia, and a grade 3/6 regurgitant systolic murmur was heard over the lower left sternal border	Chest X-ray (CXR) showed cardiomegaly and pulmonary congestion. In ECG found abnormal Q waves in leads 1, aVL, V5, and V6. Echocardiographic showed left atrial and left ventricular enlargement, severe mitral regurgitation, left ventricular ejection fraction of 30%, hyperechogenic papillary muscles, and prominent flow in the septal perforators. CT Angiography showed normal coronary arteries. Pulmonary artery-focused contrast echocardiography with supplemental oxygen (PCESO) through injection of agitated saline found ALCAPA. Cardiac catheterization and angiography confirm ALCAPA (delayed back-filling of the anomalous LCA from the pulmonary artery with a high take-off near the orifice of the right pulmonary artery (RPA).	Surgery by these steps: Anomalous LCA harvested from the pulmonary artery and anastomosed into the left coronary sinus of the aorta. Then the left coronary sinus of the aorta and the pulmonary artery were reconstructed using the autologous pericardial patch.	Good clinical condition after 1 week. On Follow up, the left ventricular systolic function had improved, and there was no obstruction in the course of the anastomosed LCA.

Authors (year)	Diagnosis and Associated Anomaly	Subject	Clinical Manifestations	Diagnostic Tools	Corrective Treatment	Outcome
Bhende et al, (2021)	Non-facing sinus ALCAPA, left sided cardiomegaly	5 months, M	Irritability and poor feeding, intermittent episodes of cough and cold, tachypnea, malnutrition, stunting, bilateral crepitations	Two-dimensional (2D) echocardiography showed anomalous origin of the LCA from the pulmonary artery, dilatation of left atrium and left ventricle, mild to moderate mitral valve regurgitation, mild pulmonary valve regurgitation, and a patent ductus arteriosus with no evidence of pulmonary artery hypertension. Ct angiography showed a cardiothoracic ratio of 58.6% and confirmed the echocardiography findings of ALCAPA. Left main coronary artery was shown to be arising from the posteroinferior wall of main pulmonary artery (MPA) having a diameter of 2.0 mm, and the origin was 15.5 mm distal to the pulmonary valve and 9.0 mm proximal to the bifurcation of MPA.	Side-to-side anastomosis of the aorta to the pulmonary artery through medial wall opening. The flap of the anterior wall of the pulmonary artery was displaced posteriorly to cover the back wall of the pulmonary artery and create a passageway within the pulmonary artery from the aorta to the LCA (Takeuchi technique).	25th post-operative day, the baby accepting full feeds and was discharged in stable condition. Five month follow up found no sign abnormality and complication.
Butt et al, (2020)	ALCAPA, 2 cases with moderate-severe MR	1,5-15 years, 3 M and 3 F	Dyspnoea, palpitation, poor feeding, fatiguability, pallor and a murmur of mitral regurgitation	Echocardiography: left ventricular ejection fraction (LVEF) was 64% (range 19-67)	LCA dissected from pulmonary artery, then anastomosed to left side of cut aorta using prolene 6-0. Pulmonary trunk also anastomosed using prolene 6-0. Mitral valve repair for patient with severe MR	No mortality, Supra Ventricular Tachycardia occurred in one patient. Patients were discharged on an anti-failure therapy. In long term, two patients loss to follow up and 4 patients mean LVEF was 66.3%
Karoiczak et al, (2001)	ALCAPA, Bland-White-Garland syndrome	15 years, F	Throbbing in the precordial region and a continuous murmur that could be heard over the mitral valve of 2/6 intensity	Ultrasonography: mitral valve defect (thickening of anterior leaflet), coronary artery fistula ECG: anterior right bundle branch block Echocardiography revealed no abnormalities in location and relations of heart structures and large blood vessels, enlarged left ventricle with normal myocardial contractility, degenerative changes and prolapse of the mitral valve cusps. It was also established that only one (right) coronary artery (RCA), 8–9 mm wide, arose from the aorta Aortography revealed wide RCA arising from the aorta. The left coronary artery, and then the pulmonary trunk, were supplied by distended collaterals between the right and the left coronary artery.	Surgery with Hamilton technique (The aorta and the pulmonary trunk were opened using transverse incisions above the commissural level. Followed by formation of an aorto-pulmonary window (APW), and then a tunnel inside the pulmonary trunk, connecting the APW with the LCA ostium). Takeuchi technique is avoided because brittle and calcified wall of the vessel)	Persistent pericardial exudate (treated with Encorton). Three months follow up, the patient does not report any complaints and echocardiography indicates normal blood flow in the coronary arteries.

Authors (year)	Diagnosis and Associated Anomaly	Subject	Clinical Manifestations	Diagnostic Tools	Corrective Treatment	Outcome
Gribaa et al, (2014)	ALCAPA, dilated cardiomyopathy	6 months, F	Clinical severe heart failure	<p>Electrocardiography (ECG) findings were left ventricular hypertrophy, abnormal Q waves in leads I, AVL, V5, and V6.</p> <p>Chest X-ray: cardiomegaly (CTR 62%).</p> <p>Transthoracic echocardiography (TTE): dilated and hypokinetic left ventricle with an ejection fraction (EF) of 35% and mild mitral regurgitation.</p> <p>Cineangiography: enlarged RCA arising from aorta and a retrograde filling of LCA through collaterals from RCA.</p> <p>ECG-gated CT-angiography: RCA arising from aorta with anomalous origin of LCA emerging from PA.</p>	Aortic reimplantation of left main coronary artery	No perioperative or postoperative complication and discharged 10 days after surgery Six months follow up: good functional status and normal growth and echocardiography show improvement in systolic function (EF of 45% with persisting mild mitral insufficiency).