The Cardiologist

ISSN: 2691-5685

Case Report

The Diagnosis and Treatment of Anomalous Single Coronary Artery from Pulmonary Artery

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Abstract

The anomalous artery origin of the left coronary from the pulmonary artery is a rare cardiac anomaly with a low incidence and high mortality rate of 1/3000000, and the anomalous artery origin of the single coronary from the pulmonary artery is rarer and the mortality rate is higher. We summarized the current literature reports, a total of 21 cases were reported, including 16 cases of surgical correction, and 9 cases of survival, with the age up to 18 months, we report a 3-year and 4-month-old child with an anomalous artery origin of the single coronary from the pulmonary artery who underwent successful surgery and survived. After 6 months follow-up, the coronary artery blood flow was smooth, the pulmonary artery pressure was normal, but the mitral regurgitation was large.

Keywords: Single coronary; Origin; Diagnosis; Treatment

Case Presentation

The patient was3-year and 4-month-old, male, weighing 15 kg and was hospitalized for pneumonia. Echocardiography indicated anomalous origin of coronary artery and was admitted to our hospital. After hospitalization, electrocardiogram examination suggested ST-T elevation, BNP elevation, and echocardiography suggested LV shumoduan inner diameter 53 mm, adductor end 35 mm, Ef 62%, left atrium 47×61×70 mm, left ventricular endocardial echo enhancement, chordae tendinosus and papillary muscle showed blood deficiency, anterior mitral prolapse, incomplete closure, mitral annulus 41 mm, and massive mitral regurgitation. The right coronary sinus of the aorta showed no definite coronary artery emission, and there was no patent ductus arteriosus.

CT of the coronary artery indicated significant enlargement of the left atrium and left ventricle, and the common origin of the right coronary artery and the left main trunk of the left sinus, however, all-origin anomalies are suspected on imaging (Figure 1).

Coronary angiography showed that the right sinus of the pulmonary artery sent short trunk, which was divided into left and right coronary arteries, and pulmonary artery pressure was measured at 60 mmHg (Figure 2).

Surgical methods

Extra Corporeal Membrane Oxygenation (ECMO) was prepared before operation, blood pressure was measured 97/46 (61) mmHg and pulmonary artery pressure was 43/14 (25) mmHg after thoracotomy. The coronary artery was dissociated and button-shaped coronary artery openings were cut.

Citation: Cao Y, Liu Y, Li L, Wang Y, Zhang W, Fan X, et al. The Diagnosis and Treatment of Anomalous Single Coronary Artery from Pulmonary Artery. Cardiologist. 2019; 1(1): 1004.

Publisher Name: Medtext Publications LLC

Manuscript compiled: October 10th, 2019

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The autologous pericardial suture is tapered, the coronary artery is anastomosed to the side wall of the aorta (Figure 3), and the heart automatically re-jumps. Due to the difficulty of shutting down extracorporeal circulation, ECMO assist is used.

On the 4th day after the operation, the ECMO was withdrawn, the tracheal intubation was removed on the 7th day, and the 10th day was discharged.

At 6 months follow-up, echocardiography showed increased mitral regurgitation, coronary angiography showed smooth blood flow (Figure 4), and right heart catheterization showed pulmonary artery pressure was 30 mmHg (Figure 5).

Discussion

Anomalous origin of coronary artery is a kind of congenital malformation with low morbidity and high mortality, while left or right anomalous origin of coronary artery is rarer. Literature reports are mostly case reports. We reviewed the reported literature, and based on A. Mirza's [2] reports, a total of 21 cases have been reported so far (Table 1) [2-12]. The largest 18 months, the smallest 3 days. Surgical correction was performed in 16 patients, 9 survived and 12 died. Among the 21 patients, the age of operation ranged from 3 days to 18 months. 15 patients were combined with other cardiac malformations, including PAT/DORV/VSD/ASD/PDA, and 5 patients survived surgery with a survival rate of 33.3%. 6 patients without other heart malformations, the oldest was 18 months, and 4 patients survived the surgery with a survival rate of 66.7%. Two deaths occurred in 1983 and 1984. The survival rate of patients without other cardiac malformations was higher than that of patients with other cardiac malformations (Table 2). There were no deaths as technology improved. The low survival rate of patients with other cardiac malformations may be related to the long operative time, inadequate surgical preparation, and the failure of cardiac arrest to detect the abnormal origin of coronary artery for the first time.

Such children generally have one of the following three conditions for survival [3]: (1) Immature pulmonary vascular bed (maintains high and persistent pulmonary arterial pressure). (2) Higher left ventricular diastolic pressure (indirect increase of pulmonary artery pressure). (3) Continuous opening of large arterial catheters (increasing pulmonary artery pressure and increasing coronary oxygen supply).

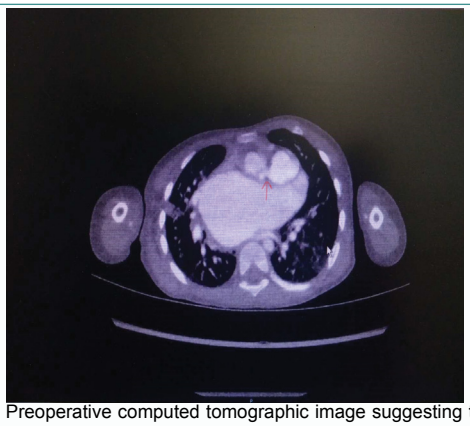

Figure 1: Preoperative computed tomographic image suggesting the single coronary artery from the pulmonary artery.

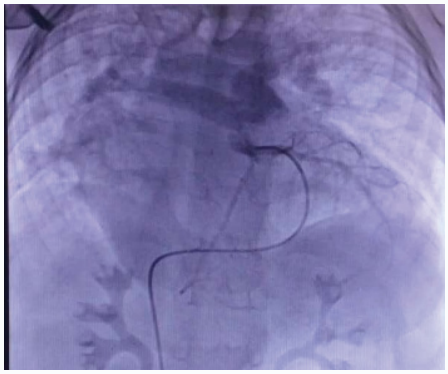

Figure 2: Preoperative Coronary angiography image suggesting the single coronary artery from the pulmonary artery.

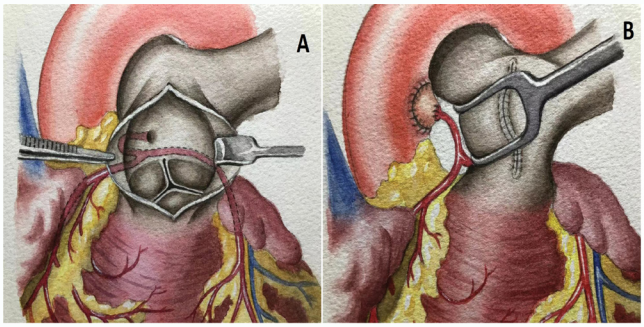

Figure 3: (A) Common coronary trunk arising from the right anterior facing sinus of the pulmonary artery. (B) Tapered autologous pericardial for prolonging coronary trunk was reimplanted into the aorta.

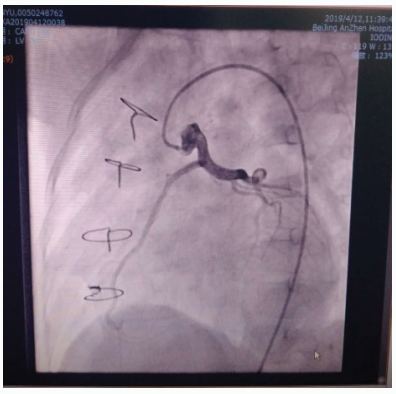

Figure 4: Coronary blood flow.

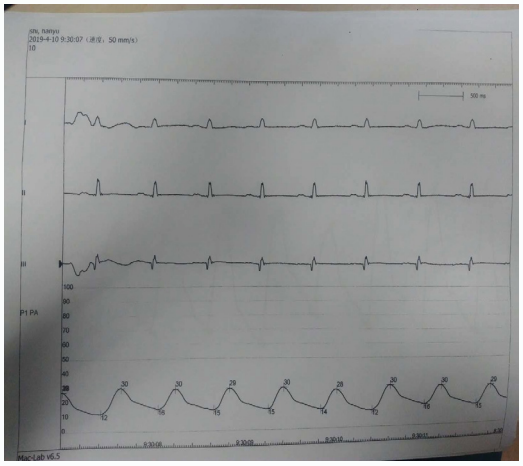

Figure 5: Pulmonary artery pressure.

Table 1: Summary of publications about the anomalous origin of both coronary arteries arising from the pulmonary artery diagnosed in living infants.

First author	Year	Case	Age	Associated pathology	Surgical correction	Survival >1 week
D'Alessandro	1976	1	39 y	PTA	Yes	Yes
Shore [13]	1983	1	23 y	APSD/VSD/PA	Yes	Yes
Keeton [14]	1983	1	5 mo	No	Yes	No
Goldblatt [15]	1984	2	2 mo	No	Yes	No
			3 mo	Pseudo-AC	Yes	No
Heifetz [6]	1986	2	1 w	PA/VSD/PFO/PDA	No	No
			2 w	PA/VSD/PFO/PDA	No	No
Lloyd [16]	1987	1	6 mo	APSD/VSD	Yes	No
Ho SY [17]	1988	1	1 w	PAIS	No	No
Urcelay [4]	1994	2	3 mo	No	Yes	Yes
			6 mo	VSD	Yes	Yes
Santoro [18]	1995	2	1 mo	AC	No	No
			2 mo	AC	Yes	No
Heusch [19]	1997	1	1 mo	VSD	Yes	No
Da Cruz [20]	1998	1	12 d	VSD/PDA/ASD/AC	No	No
Enrique Ochoa-Ramirez [9]	2005	1	2 mo	No	Yes	Yes
Qiang Chen [11]	2006	1	6 w	No	Yes	Yes
Mohsen Karimi [7]	2012	1	7 d	PDA	Yes	Yes
Keti Vianova [5]	2014	1	3 d	COA	Yes	No
Changwei Zhang [12]	2016	1	18 mo	No	Yes	Yes
Katarzyna Januszewska [10]	2018	1	7 d	DORV/VSD/ASD	Yes	Yes

PTA: Persistent Truncus Arteriosus; APSD: Aorticopulmonary Septal Defect; VSD: Ventricular Septal Defect; PA: Pulmonary Atresia; AC: Aortic Coarctation; PFO: Patent Foramen Ovale; PDA: Patent Ductus Arteriosus; PAIS: Pulmonary Atresia with Intact Septum; ASD: Atrial Septal Defect.

Pulmonary artery can provide certain blood and oxygen supply to coronary artery, which is the condition of survival. Patients with other cardiac malformations can guarantee certain blood and oxygen supply because of the existence of intracardiac shunt. Patients without other cardiac malformations rely on sufficient pulmonary artery pressure to ensure the blood and oxygen supply of coronary artery [4], but there are still manifestations of myocardial ischemia and mitral regurgitation. Therefore, preoperative diagnosis of these diseases is difficult, ultrasound is difficult to detect, some patients are found in surgery [5], so coronary angiography is the golden standard, but it is difficult to ensure that each patient has coronary angiography.

Table 2: Survival rate of patients.

Other Cardiac Malformations	Case	Survival	Rate
Y	15	5	33.30%
N	6	4	66.70%

Table 3: Echo cardiographic parameters.

Time	End diastolic diameter (mm)	End systolic diameter (mm)	EF%	FS%
Preoperative	53	35	62	33
1st	40	35	24	11
4th	38	33	40	19
12th	43	31	54	27
19th	40	25	65	35
6 mo	44	31	57	29

Table 4: Myocardial enzymatic parameters.

Time	CK (U/L)	CKMB (ng/ml)	LDH (U/L)	TNI (ng/ml)
1st	3865	93.7	1204	>81
3rd	1337	6.3	1188	16.12
4th	625	3.2	1043	9.3
5th	304	7.6	861	
10th	49	1.5	537	
12th	35	1.7	437	

Paolo Angelini [3], MD, summarized the possible symptoms and abnormalities of preoperative examination in these patients: (1) Shortness of breath 1 to 6 weeks after birth. (2) Feeding difficulties. (3) Crying without obvious inducement. (4) Pale and prone to sweating. (5) X-ray or ultrasonography suggests cardiac enlargement with or without mitral valve disease. (6) Electrocardiogram ST-T changes and Q wave changes. (7) Myocardial enzymes were significantly elevated.

The patient we reported had 5, 6 and 7 of the above symptoms, but no obvious clinical manifestations. This also suggests that for patients with abnormal origin of coronary arteries, preoperative coronary angiography is necessary. Although some children have poor preoperative cardiac function, angiography should also be performed.

This child is the oldest case of pulmonary artery origin of total coronary artery. The characteristics of this case are different from those of the reported case, with the following characteristics: (1) Older, 3 years old and 4 months old, is the oldest survivor reported before surgery. (2) There was no significant decrease in activity and cardiac function, and the cardiac function index was normal. (3) The child had no open arterial catheter and no other cardiac malformations. (4) Pulmonary artery pressure was 60 mmHg by right cardiac catheterization. (5) Massive mitral regurgitation.

We compared the echocardiographic and myocardial enzymatic parameters of the patients before and after operation (Table 3 and 4). The echocardiographic findings showed that the left ventricle shrank gradually after operation, EF and FS increased gradually, but decreased 6 months after operation and aggravated valve regurgitation.

Heifetz et al. [6] reported that about 60% of all deaths occurred two weeks after birth, 70% of the causes of survival were associated with other cardiac malformations, and a few cases survived with lower pulmonary artery pressure and oxygen supply, but only 11 weeks survived. For newborns, dyspnea and other symptoms may occur after birth. The use of prostaglandin E to ensure the opening of the ductus arteriosus wins the opportunity for surgery for patients 7. The relatively high pulmonary artery pressure may be one of the reasons for the survival of the child; otherwise the coronary artery blood supply and oxygen supply cannot be satisfied. However, pulmonary hypertension without other cardiac malformations does not exclude the possibility of primary pulmonary hypertension or indirectly increased pulmonary artery pressure. Therefore, right cardiac catheterization 6 months after operation found that pulmonary artery pressure was normal, excluding primary hypertension. It is proved that the preoperative pulmonary artery pressure of 60 mmHg may be caused by a large number of mitral regurgitation, and that the mitral regurgitation is aggravated half a year after the operation. It is also proved that the valve may be caused by myocardial ischemia, and that the left ventricular diastolic pressure caused by the valve regurgitation will increase the pulmonary artery pressure. Mitral valve regurgitation, left ventricular diastolic pressure and pulmonary artery pressure are exactly balanced, so that the patient can survive Secondly, it may have abundant collateral blood supply of coronary artery to ensure its survival to three years old. Although angiography has not been found, there is a small amount of blood reflux in the coronary artery orifice during the operation.

We believe that: it is difficult to diagnose these diseases and the mortality rate is high. Children with ECG myocardial ischemia, increased myocardial enzymes or mitral regurgitation, when excluding myocarditis and cardiomyopathy, we should consider whether there is abnormal origin of coronary artery.

It is necessary to perform coronary angiography before operation. Increased pulmonary artery pressure is a necessary factor for survival. Keeping the ductus arteriosus open in newborns helps to win the operation opportunity. Surgical prolongation of the coronary artery is conducive to reducing tension and ensuring the supply of blood to the coronary artery. There are problems of low cardiac function and delayed chest closure after operation. The planned use of ECMO during operation can help the smooth recovery of cardiac function. At the same time, the operation effect of patients without other cardiac malformations is better than those with other cardiac malformations. The cardiac function of survived children can be restored to normal after operation, and the prognosis is good.

References

1. Keith JD. The anomalous origin of the left coronary artery from the pulmonary artery. Br Heart J 1959;21:149-61.
2. Mirza A, Bergoend E, Legendre A, Neville P. Successful Surgical Treatment for a Single Coronary Artery Arising From the Pulmonary Artery. Pediatr Cardiol. 2008;29:1107-9.
3. Angelini P. The Miraculous Survivalof an Exceptional Baby: can a heart manage to function with anomalous origin of both coronary arteries from the pulmonary artery? Tex Heart Inst J. 2008;32(3);351-3.
4. Urcelay GE, Iannettoni MD, Ludomirsky A, Mosca RS, Cheatham JP, Danford DA, et al. Origin of Both Coronary Arteries from the Pulmonary Artery. Circulation. 1994;90(5):2379-84.
5. Vitanova K, Cleuziou J, Deutsch MA, Ackermann K, Schreiber C. A Rare Anomaly of a Single Coronary Artery Arising from the Right Pulmonary Artery in a Neonate. World J Pediatr Congenit Heart Surg. 2014;5(3):453-5.
6. Heifetz SA, Robinowitz M, Mueller KH, Virmani R. Total Anomalous Origin of the Coronary Arteries from the Pulmonary Artery. Pediatr Cardiol.1986;7(1):11-8.
7. Karimi M, Hulsebus E, Lutin W. Reimplantation of Anomalous Single Coronary Artery from Pulmonary Artery: Diagnosis and Surgical Management. Ann Thorac Surg. 2012;93(1):308-10.
8. Tavora F, Burke A, Kutys R, Li L, Virmani R. Total anomalous origin of the coronary circulation from the right pulmonary artery. Cardiovasc Pathol. 2008;17(4):246-9.
9. Ochoa-Ramírez E, Valdez-Garza HE, Reyes-González R, Mateos-Corral D, Sánchez-Sigel D, González-López V. Double Anomalous Coronary Origin from the Pulmonary Artery. Tex Heart Inst J. 2005;32(3):348-50.

10. Januszewska K, Kehl HG, Malec E. Single Ostium of the Right and Left Coronary Artery from the Right Pulmonary Artery. Ann Thorac Surg. 2018;105(2):e67-9.

11. Chen Q, Soo E, Tometzki AJ, Parry AJ. Surgical Management of Single Ostium Anomalous Coronary Artery From Pulmonary Artery. Ann Thorac Surg. 2006;81(3):1127-9.

12. Zhang C, Zhang H, Gao H, Wang X, Liu J, Wang Y, et al. Single-Trunk Anomalous Origin of Both Coronary Arteries from Pulmonary Artery: Serendipitous Diagnosis and Successful Surgical Treatment. Ann Thorac Surg. 2016;102(1):e49-50.

13. Shore DF, Ho SY, Anderson RH, De Leval M, Lincoln C. Aortopulmonary septal defect coexisting with ventricular septal defect and pulmonary atresia. Ann Thorac Surg. 1983;35:132-7.

14. Keeton BR, Keenan DJ, Monro JL. Anomalous origin of both coronary arteries from the pulmonary trunk. Br Heart J. 1983;49(4):397-9.

15. Goldblatt E, Adams AP, Ross IK, Savage JP, Moris LL. Single-trunk anomalous origin of both coronary arteries from thepulmonary artery: Diagnosis and surgical management. J Thorac Cardiovasc Surg. 1984;87(1):59-65.

16. Lloyd TR, Marvin WJ Jr, Lee J. Total anomalous origin of the coronary arteries from the pulmonary artery in an infant with aorticopulmonary septal defect. Pediatr Cardiol. 1987;8(2):153-4.

17. Ho SY, Carvalho J de S, Sheffifield E. Anomalous origin of single coronary artery in association with pulmonary atresia. Int J Cardiol. 1988;20(1):125-8.

18. Santoro G, di Carlo D, Carotti A, Formigari R, Boldrini R, Bosman C, et al. Origin of both coronary arteries from the pulmonary artery and aortic coarctation. Ann Thorac Surg. 1995;60(3):706-8.

19. Heusch A, Quagebeur J, Paulus A, Krogmann ON, Bourgeois M. Anomalous origin of all coronary arteries from the pulmonary trunk. Cardiol. 1997;88(6):603-8.

20. Da Cruz E, Carbognani D, Laborde F, Bourgaran J, Dibie A, Le Bidois J, et al. Aortic coarctation, multiple ventricular septal defects, and anomalous coronary artery arising from the right pulmonary artery. J Thorac Cardiovasc Surg. 1998;115(1):244-6.