Cardiology in the Young

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A neonate with crossed pulmonary arteries: a case report and literature review of 115 cases worldwide

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Review

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Abstract

Malposition of the branch pulmonary arteries (MBPA) is an unusual malformation characterised by anomalous origin of both pulmonary arteries from the main pulmonary trunk. To date, only few cases have been reported. Herein, we present the first case report of a 3-day-old, full-term male neonate with the lesser form of crossed pulmonary arteries in Saudi Arabia detected by echocardiography and confirmed by cardiac CT. Crossed pulmonary arteries is not a rare anomaly, but it is a somewhat underreported anomaly, and their recognition is important because it is usually associated with other CHDs, airway obstruction, extra-cardiac anomalies, and certain genetic syndromes.

Crossed pulmonary arteries is the classic form of malposition of the branch pulmonary arteries, which was first described by Jue KL and associates in 1966 in an infant with the trisomy 18 syndrome,[1] then Becker et al. in 1970 described the lesser form of this anomaly.[2] Since then, several cases have been reported.[3–5] The exact prevalence of malposition of the branch pulmonary arteries is not known; Hui Liu, in his hospital-based study, found approximately 0.06% in the population who underwent chest CT or CT angiography.[6] Jue KL et al[1] suggested that this condition could result from a faulty differential growth within the pulmonary trunk during the partitioning of truncus arteriosus with a counterclockwise rotation of normally formed main and branch pulmonary arteries.[1,7] It is postulated that this malposition occurs at approximately 41st day of development when the right and left pulmonary arteries ostia migrate towards each other.[7]

Becker et al.[2] classified this malformation based on the presence or absence of crossing. The typical/classical form of crossed pulmonary arteries, which is more common, has an origin of the left pulmonary artery superior to the right of the origin of the right pulmonary artery. Thus, the two branches crisscross one another along with their courses to their related lungs.[2,4] Furthermore, the postulated developmental mechanism is the counterclockwise rotation of the normal origins of the branch pulmonary arteries secondary to abnormal differential growth within the pulmonary trunk.[5,8] Rabail Raza[9] reported reversed crossing, a rare variant of the typical type of crossed pulmonary arteries in which the right pulmonary artery ostium lay superior to the left of the left pulmonary artery ostium, which the authors attributed to clockwise rotation of branch pulmonary arteries instead of the usual counterclockwise rotation.[9] However, the theories regarding clockwise or counterclockwise rotation during the partitioning of the truncus arteriosus are not yet proven. The "lesser form" of MBPA, also called the atypical, is characterised by a direct superior–inferior orientation of pulmonary arteries branches ostia without crossing the branch pulmonary arteries, more commonly, left pulmonary artery openings directly above the right pulmonary artery.[2,4] Jing Zhang reported a rare case with right pulmonary artery openings directly above the left pulmonary artery, the only one in the literature.[10] Moreover, the sixth aortic arch forms the distal pulmonary arteries; this phenomenon may explain why crossed pulmonary arteries is frequently observed with aortic arch abnormalities.[1,11] Furthermore, in Babaoglu et al[11] study, 45% of his cases (9 out of 20 patients) were associated with a genetic syndrome. Recto et al.[4] also showed 4 of 10 patients with microscopic deletion of the chromosomal region 22q11.

Case report

A 3-day-old full-term male neonate with intrauterine growth restriction was referred to the cardiology team because of cardiac murmur. The findings of general physical examination were found to be normal. Cardiovascular examination revealed normal first and second heart sounds with pansystolic murmur, grade III/VI, and maximum intensity at the left lower sternal border.

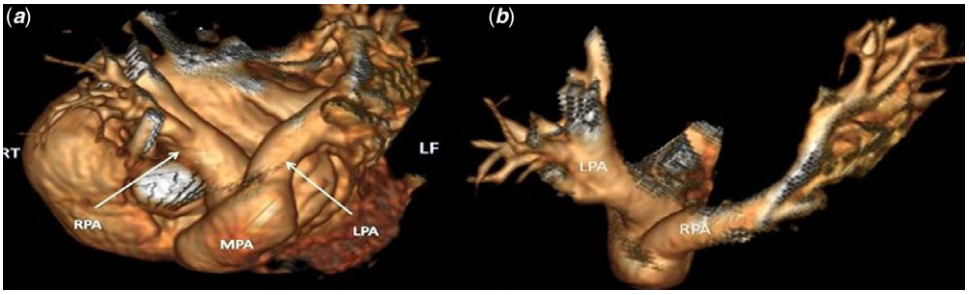

Figure 1. Volume-rendered reconstruction shows the lesser form of malposition of branch pulmonary arteries. The origin of left pulmonary artery (LPA) from the main pulmonary artery (MPA) in a plane superior to that of the right pulmonary artery (RPA), and then the two pulmonary arteries proceeded to their respective lungs without crossing. ***a***) From anterior and ***b***) from posterior.

Chest: Good air entry bilateral with no added sounds. Abdomen: soft, non-tender, and no organomegaly. Central nervous system examination: conscious, alert, normal tone, power, reflexes, and primitive reflexes. A chest roentgenogram showed cardiomegaly with a cardiothoracic ratio of 0.65. An Echocardiography showed situs solitus, levocardia, normal systemic and pulmonary venous drainage and connections. There was a large secundum atrial septal defect with left to right shunt. Competent atrioventricular valves. Large mal-aligned ventricular septal defect with left to right shunt. Mild pulmonary valve stenosis with peak gradient of 28 mmHg. Small patent ductus arteriosus with left to right shunt. Good size branch pulmonary arteries were not seen in the same level. Unobstrcted left aortic arch. The Cardiac CT angiography was performed for further evaluation of the pulmonary trunk and its branches. It showed that branch pulmonary arteries originate from the pulmonary trunk but in superior–inferior relation. The left pulmonary artery arises superior to the right pulmonary artery, then each branch pulmonary artery travels to their respective lungs without crossing (Fig 1: a and b).

Results

We review all the cases with a diagnosis of malposition of the branch pulmonary arteries as per available literature. Most of the cases were diagnosed in the infancy period. The male and female ratios are equal to 1:1. About 42.6% of cases have chromosomal abnormalities with around 20% of DiGeorge syndrome. Two-thirds of the cases were diagnosed by echocardiography and one-third by CT cardiac. Around 53% of cases required surgery for cardiac anomalies, whereas 5% required operation due to airway compression (Table 1).

The typical (classic crossing) type of malposition of the branch pulmonary arteries accounts for 70% of cases, whereas the lesser type (common variant with left pulmonary artery superior to the right pulmonary artery) accounts for an exact of 30%. The isolated malposition of the branch pulmonary arteries (without cardiac defects) accounts for 13% of total cases and the rest (87%) with the association of cardiac lesions. Simple cardiac lesions are around 70%, whereas 30% are with complex CHDs. The most common simple cardiac defects are ventricular septal defect, secundum atrial septal defect, and patent ductus arteriosus with 64%, 59%, and 33%, respectively. The most complex CHDs are tetralogy of Fallot, CoA, and interrupted aortic arch as shown in Table 2 and Table 3.

Discussion

Malposition of the branch pulmonary arteries is a condition that is underdiagnosed and underreported. As per available literature, we found that the classic type was about two times more common than the lesser type, which is similar to the result reported by Hui Liu.[6] The patients with isolated crossed pulmonary arteries are usually asymptomatic. They did not require special intervention unless if there is stenosis of pulmonary arteries branches or CHDs requiring repair. Malposition of the branch pulmonary arteries does not cause mechanical airway obstruction by itself. If it occurs, it is related to associated cardiovascular anomalies like pulmonary arterial sling.[5,6,11,12] Crossed pulmonary arteries must be distinguished from pulmonary artery sling and anomalous origin of one pulmonary artery[3,5,10] and also from the abnormal course of the left pulmonary artery in the setting of a horseshoe lung.[8]

Echocardiography is the primary method used to demonstrate crossed pulmonary arteries in most of the reported cases, as it was a safe and convenient economical image technique.[7,10,11] Modern imaging methods, such as cardiac magnetic resonance angiography and CT, help to diagnose this disease precisely and may aid in correct cardiac repair planning.[13] Many of the authors believe that echocardiography is enough to diagnose crossed pulmonary arteries in most patients, and the cardiac CT is indicated in special cases, for example, poor echo window suspected associated aortic arch anomalies or need to have more information about the course and location of the pulmonary artery branches and related anatomic structures, especially with complex cardiac anomalies.[11] On the other hand, Zhang et al and Jin Chen supported the use of CT in the diagnosis of crossed pulmonary arteries if there is any doubt by echocardiography as they thought that many cases are missing.[10,14]

The disease is found to have a male predilection as per the reports available in literature.[4,6,7,9–11,15] A good number of cases were reported mainly from China.[5,6,10,16–19] Genetic syndrome was detected in 45% cases reported in literature; the commonest reported syndrome was DiGeorge syndrome. Other chromosome abnormalities include trisomy 18, partial monosomy Xq, partial trisomy 1q, Holt–Oram, Noonan, Turner, Castello, and Frankter Haar syndrome.[12,20]

As per the available literature, the classical crossing[2,4–8,11–14,16,17,21–31] is found in majority of cases reported in literature as compared to reverse crossing[9] among typical variety. The typical variety is classified into crossed pulmonary arteries and reverse crossed pulmonary arteries. The crossed pulmonary arteries is associated with left pulmonary artery origin superior to the right of the right pulmonary artery, and the two branches crisscross one another along with their courses to their respective lungs, whereas the reverse crossed pulmonary arteries is described as right pulmonary artery origin superior to the left of the left pulmonary artery, and the two branches crisscross one another along with their courses to their respective lungs. Among the lesser type, common variant left pulmonary artery superior to the right pulmonary artery is seen more commonly.[2,6,10,13] The lesser form or atypical variety is further described as common variant and rare variant.

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Table 1. The demographic data, associated chromosomal abnormality, method of diagnosis, and surgical outcome

Patient characteristics	Number	Percentage	References
Age			
Prenatal	2/70	2.8%	19, 20
Neonate (< 1 month)	29/70	41.4%	2,4,5,6,7,10,11,16,21,22,23, our case
Infant (1–12 months)	26/70	37.1%	2,3,5,6,7 8,9,10,11,13,16,17,24,25,26
> 1-year-old	13/70	18.6%	5,6,15,16,17,27,28,29,30
Undetermined	45/115	64.2%	6,10,14
Sex			
Male	51/115	44.3%	4,5,6,7,8,9,10,13,16,17,23,24,28, our case
Female	52/115	45.2%	2,4,5,6,7,8,10,15,17,20,21,22,26,27,29,30
Undetermined	12/115	10.4%	2,11,14,19,25
MBPA associated with chromosomal abnormality	23/54	42.6%	2,4,5,7,8,9,11,17,25, our case
MBPA associated with DiGeorge syndrome	11/54	20.3%	4,5,8,11,17,25
MBPA associated with arch anomaly and DiGeorge syndrome	7/11	6%	4,5,8
Method of MBPA diagnosis:			
Echocardiography	72/115	62.6%	5,6,7,8,10,11,14,17,21,22,23,25,29,
CT cardiac	33/115	28.7%	4,9,10,11,13,15,16,24,28,30
Prenatal echo	2/115	1.7%	19,20
Cardiac catheterisation	3/115	2.6%	3,26
Others (MRI, surgery)	2 /115	1.7%	17
Outcome			
MBPA with successful surgery for cardiovascular anomalies	53/115	46%	1,4,5,6,7,8,10,15,17,23,24,29,30, our case
MBPA require operation for airway compression	5/115	4.3%	6,7,16,29
Not require surgery, died patients or unavailable data	56/115	48%	2,3,4,5,8,9,10,13,14,19,20,21,22

MBPA = malposition of the branch pulmonary arteries.

Table 2. MBPA types, associated cardiovascular anomalies, and airway obstruction in 112 reported cases

Characteristics	Number	Percentage	References
Type of MBPA			
Typical type	80/115	69.5%	
Classical crossing	79/80	98.7%	1–8,10,11,13–17,19–30
Reverse crossing	1/80	1.3%	9
Lesser type	35/115	30.4%	
Common variant (LPA superior to RPA)	34/35	97.1%	2,6,10,17, our case
Rare variant (RPA superior to LPA)	1/35	2.9%	10
Isolated MBPA (no cardiac defects)	15/115	13%	10,20,26
Associated cardiovascular anomalies	100/115	87%	1–11,13–17,19,21–25,27–30
Simple cardiovascular anomalies	69/100	69%	1,5–8,10,11,13,15,17,21,23–25,29,30, Our case
Complex cardiac anomaly	31/100	31%	2,3,4,5,6,7,8,9,10,11,14,17,19,22,23,27,28
Aortic arch anomalies	47/100	47%	2,4,5,7,8,9,11,17,22,23,26,28,29,30
Associated with air way compression	8 /115	6.9%	6,7,16,29
Due to complete vascular ring	3/8	37.5%	6,7,29
Due to compression without complete vascular ring	5/8	62.5%	6,16

LPA = left pulmonary artery; MBPA = malposition of the branch pulmonary arteries; RPA = right pulmonary artery.

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Table 3. MBPA and the associated cardiovascular anomalies in 115 patients

References	Cardiac anomaly	Number	Percentage
Babaoglu K; Wolf WJ and Area C.	A)Isolated CPA	15/115	13%
	B) Simple cardiac anomalies	69/100	69%
Chen B; Liu H; Zimmerman FJ; Raza R; Babaoglu K; Hernandez LE; Cheng J; Fu F; Agarwal A; Miyahara Y and our case.	Ventricular septal defect	44/69	64%
Liu H; Zimmerman FJ; Raza R; Babaoglu K; Hernandez LE; Cuturilo G; Fu F; Agarwal A and our case.	Atrial septal defect – II	41/69	59%
Recto MR; Chen B; Liu H; Zimmerman FJ; Raza R; Zhang J; Babaoglu K; Cuturilo G; Agarwal A and our case.	Patent ductus arteriosus	23/69	33%
Chen B; Liu H; Raza R; Fu F and our case	Pulmonary stenosis	18/69	26%
Liu H; Zimmerman FJ; Babaoglu K; Hernandez LE; Fu F.	Bicuspid aortic valve with aortic stenosis	7/69	10%
Liu H and Babaoglu K	Cor triatriatrum	2/69	3%
Cuturilo G and Batti S El	Pulmonary vein stenosis	2/69	3%
Batti S El, et al	Single atrium	1/69	1%
Liu H; Raza R; Fu F and Gynecol UO.	Others (PAPVC, ALCAPA, AORL, AVSD, AP window)	5 /69 (one for each lesion)	7%
	C) Complex CHD	31/100	31%
Chen B; Liu H; Zimmerman FJ; Raza R; Chen J; Altun G and Sivakumar K	Tetralogy of Fallot	10/31	32%
Becker AE; Recto MR; Chen B; Liu H; Fu F and Piluiko VV.	Interrupted aortic arch	10/31	32%
Becker AE; Liu H; Raza R; Hauck AL and Siwik ES.	Truncus arteriosus	7/31	23%
Zimmerman FJ; Zhang J and Babaoglu K.	Double-outlet right ventricle	4/31	13%
Zimmerman FJ and Hernandez LE.	HLHS	3/31	10%
Chen B; Raza R; Hernandez LE and Fu F.	Others: (Ebstein's anomaly, TAPVC, TGA, tricuspid atresia, right isomerism)	5/31 (one for each lesion)	16%
	D) Aortic arch anomalies	47/100	47%
Chen B; Liu H; Zimmerman FJ; Raza R; Fu F; Altun G; Hauck AL; Sivakumar K and Ersek S.	Right-sided aortic arch	18/100	18%
Liu H; Zimmerman FJ; Raza R; Zhang J and Hernandez LE.	CoA	11/100	11%
Becker AE; Recto MR; Chen B; Liu H; Fu F and Piluiko VV.	Interrupted aortic arch	10/100	10%
Recto MR; Chen B; Liu H and Ersek S.	Aberrant left subclavian artery	10/100	10%
Zimmerman FJ; Raza R and Hernandez LE.	Hypoplastic aortic arch	4/100	4%
Liu H; Zimmerman FJ; Mch ST and Ersek S.	Vascular ring	4/100	4%
Chen B; Liu H; and Area C.	Aberrant right subclavian artery	3/100	3%
Chen B; Liu H; Hernandez LE and Shunmugasundaram, P	Other: (cervical arch, ovain trunk, hypoplastic LSCA, LARDA, DOA)	5/100	5%
	E) Acquired heart disease	1/100	1%
Babaoglu K, et al	Kawasaki disease	1 case with isolated CPA	1%

AORL = anomalous origin of the right coronary artery from left coronary sinus; CPA = crossed pulmonary arteries; LARDA = left-sided aortic arch and right descending aorta; LSCA = left subclavian artery; MBPA = malposition of the branch pulmonary arteries; PAPVC = partial anomalous venous connection; TGA = transposition of the great arteries; ALCAPA = anomalous left coronary artery from the pulmonary artery; AVSD = atrioventricular septal defect; HLHS = hypoplastic left heart syndrome; TAPVD = total anomalous pulmonary venous drainage; DOA = double aortic arch; CoA = coarctation of aorta.

The common variant has left pulmonary artery openings directly above the right pulmonary artery, and the two branches do not crisscross one another whereas in the rare variant right pulmonary artery openings directly above the left pulmonary artery, and the two branches do not crisscross one another.

Malposition of the branch pulmonary arteries is not always associated with other cardiovascular anomalies,[4–6,11] as we found a total of 15 cases with isolated crossed pulmonary arteries, but however associated CHD is very common as it had been reported in 86.8% of cases and 30% were complex cases, and this result is comparable to the 20% reported by Babaoglu et al. and 40% reported by Hui Liu.[6,11]

Since Becker E. et al. in 1970 reported the association of crossed pulmonary arteries with truncus arteriosus, many authors

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postulated the link of crossed pulmonary arteries with conotruncal anomaly.[4,13] However, according to the available data, we found ventricular septal defect is the most common cardiac defect associated with crossed pulmonary arteries representing 38% (43 cases) of all patients with malposition of the branch pulmonary arteries (ventricular septal defect accompanied with complex CHD not taken in the account), and this result is near to that of Babaoglu et al.,[11] which show ventricular septal defect represent 55% (11/22) of all cases, while in Hui Liu et al.[6] study, ventricular septal defect represents 80% (12/15) and in Jing Zhang[10] study represents 13.7% (4/29). Isolated crossed pulmonary arteries generally is asymptomatic. However, they will express symptoms secondary to the associated heart abnormalities.[11] Many of the previous reports showed that malposition of the branch pulmonary arteries does not cause mechanical airway obstruction,[5,11] while Hui Liu's study showed that 33% of his patients were manifested with airway compression secondary to external compression. Three of them received successful surgery to relieve the external airway compression.[6] Only seven cases had found to be reported in our survey, and this may be because most of the authors did not mention this in their reports, or most of the cases were diagnosed by echocardiography. In the literature coexisting anomalies of the aortic arch, such as interrupted aortic arch, right-sided aortic arch, aberrant left subclavian artery present in 71% of cases which have reported by and Recto MR and 80% reported by Hui Liu.[4,6,13] In our review, aortic arch anomalies were detected only in 42% of malposition of the branch pulmonary arteries patients, which is close to the results of Babaoglu et al.[11] with 35% (7/20) and the difference could be attributed to the use of echocardiography in most of the cases rather than cardiac CT angiography. The prognosis and surgical outcome depend on the associated cardiac anomalies, extra-cardiac malformations, and genetic defects.[11] As per literature available, 53 patients (46% of the total) underwent successful surgery for their associated cardiovascular anomalies. In four patients who underwent pulmonary artery banding, all of them developed severe right pulmonary artery stenosis secondary to band impingement.[4,12] That is why complete early repair is preferential in this setting, rather than palliative procedures.[22] Therefore, a preoperative diagnosis of crossed pulmonary arteries and recognition of this variant may influence management decisions.[23–31]

There are some limitations in this study. Firstly, this is a retrospective review based on available literature. Hence, there may be certain lacunae in the information. Secondly, we believed that this condition is underdiagnosed and underreported, so describing its association precisely with various CHDs could be deceptive until further studies are performed. Thirdly, there is a variation in the methods employed for diagnosis in various studies which may give variable results.

Conclusion

In conclusion, we presume that MBPA is not a so rare anomaly as we thought before. It is usually associated with other CHDs and genetic syndromes, and sometime with airway obstruction. The careful echocardiography can confirm the diagnosis in most of the cases, but still a cardiac CT angiography may be necessary for more clarification of pulmonary artery anatomy, associated arch anomalies, and airway obstruction. The typical type of MBPA is two third of the atypical type. Ventricular septal defect and aortic arch anomalies are the most common reported cardiovascular abnormalities, and the prognosis depends on the associated congenital heart lesions and genetic syndromes.

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Conflicts of interest. None.

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