Surgical strategies for anomalous origin of the left coronary artery from the right pulmonary artery with an intramural aortic course: A report of 10 cases

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ABSTRACT

Objective: Few data are available regarding the surgical strategies for an anomalous origin of the left coronary artery (LCA) from the right pulmonary artery (RPA) with an intramural aortic course. We reviewed our experience in a case series of 10 children.

Methods: From 2007 to 2014, 10 patients (7 boys and 3 girls, aged 3 months to 11 years, median 21 months) underwent surgical repair. Before the operation, echocardiography showed the mean left ventricular ejection fraction $45\% \pm 10\%$ and mean fractional shortening fraction $21\% \pm 7\%$. Moderate to severe mitral regurgitation was found in 4 patients and left ventricular aneurysm in 5 patients. The intramural aortic course of LCA was not diagnosed preoperatively in any of the patients.

Results: During the operation, the LCA orifice was seen and 2 types were identified: 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. In the first type, direct coronary button transfer was performed. In the second type, button transfer with unroofing of the intramural course was performed. Annuloplasty of the mitral valve was performed in 4 patients and the aneurysm was repaired with plication technique in 2 patients. Postoperatively, 2 patients died of cardiac failure. Others showed significantly improved left ventricular function at follow-up as compared with preoperative measures (mean ejection fraction $67\% \pm 6\%$, mean fractional shortening $32\% \pm 3\%$, $P = .01$ for both).

Conclusions: Careful attention should be paid to the extremely rare association of an aortic intramural course before and during an operation when dealing with anomalous LCA from the RPA. Surgical strategies for aortic reimplantation include coronary button transfer and unroofing of the intramural segment. The outcomes are encouraging. (*J Thorac Cardiovasc Surg* 2017;153:648-53)

Operative illustrations of the button transfer and unroofing of intramural segment.

Central Message

We reviewed our surgical experience in a case series of 10 children with an anomalous origin of the left coronary artery from the right pulmonary artery with an intramural aortic course.

Perspective

Few data are available regarding the surgical strategies for an anomalous origin of the left coronary artery from the right pulmonary artery with an intramural aortic course. The case series of 10 children is the largest to date. Our surgical strategies included coronary button transfer with unroofing of the intramural segment, mitral annuloplasty, and plication of left ventricular aneurysm.

See Editorial Commentary page 654.

The most common anomaly of the anomalous left coronary artery (LCA) is from the main pulmonary artery (ALCAPA), or from the left or right pulmonary artery (RPA). This anomaly was first described anatomically in 1885 by Brooks¹ and clinically in 1933 by Bland et al.² ALCAPA is a rare congenital malformation, accounting for 1 in

300,000 live births and 0.25% to 0.5% of all congenital heart disease.^{3,4} It represents one of the most common causes of myocardial ischemia and infarction in children, and if left untreated, results in a mortality rate of up to 90% within the first year of life.^{5,6} Most ALCAPA cases present during infancy in the early months after birth, and approximately 20% of patients present in childhood or even as adults.^{5,7,8} Typically, LCA arises directly from the main pulmonary artery adjacent to the normal aortic origin as an isolated malformation. Rarely, it is from the

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Received for publication March 4, 2016; revisions received Sept 22, 2016; accepted for publication Oct 3, 2016; available ahead of print Dec 4, 2016.

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0022-5223/\$36.00

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<http://dx.doi.org/10.1016/j.jtcvs.2016.10.063>

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Abbreviations and Acronyms

- ALCAPA = anomalous left coronary artery from the pulmonary artery
- LCA = the left coronary artery
- RPA = the right pulmonary artery

RPA and can be associated with other congenital heart defects, such as ventricular septal defect, tetralogy of Fallot, and hypoplastic left heart syndrome.⁹⁻¹² Even rarer is the association with an intramural aortic course but without other congenital heart defects. Such coronary anatomy requires special consideration for the surgical procedures to be selected.¹³ A review of the published data revealed isolated case reports and a small series of 4 cases.¹³⁻¹⁵ Therefore, we sought to review our diagnostic and surgical experience in a case series of 10 children.

METHODS

A retrospective chart review was conducted after the approval by the Institutional Health Research Ethics Board of Anzhen Hospital and Capital Institute of Pediatrics in Beijing, China. From 2007 to 2014, 46 children with ALCAPA underwent cardiac surgery for complete repair. Among them, 36 patients had the anomalous origin of LCA arising from the main pulmonary artery (14 cases from the right posterior sinus with 2 deaths, 20 from the left posterior sinus with 3 deaths, 2 from the left anterior sinus with 1 death). The remaining 10 patients had the aortic intramural

LCA arising from the RPA. Charts of 10 children (7 boys, 3 girls) of the latter anomaly group undergoing surgical repair at Anzhen Hospital were reviewed.

The patient characteristics are listed in Table 1. The age of 10 patients ranged from 3 months to 11 years (median 21 months). The initial presentations included poor feeding, respiratory distress, and fussiness. Two of them (patients 2 and 3) were misdiagnosed as endocardial fibroelastosis. Electrocardiograms showed ischemic manifestations with Q-wave or/and T-wave inversion in I, AVL, and V5-6 leads.

The diagnosis was made predominantly from 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 (patients 2, 5, 6, and 10). The left ventricular aneurysm at apex was found in 5 patients (patients 2, 5, 7, 8, and 9) with the diameter ranging from 15 to 60 mm. The intramural aortic course of LCA was not identified preoperatively in any of the patients.

RESULTS

Surgical Techniques

Cardiopulmonary bypass was established through median sternotomy with cannulation of aorta and the 2 caval veins. Hypothermia was maintained at 25°C. Cardioplegic arrest was performed with antegrade cardioplegia to the aorta and main pulmonary artery with occlusion of the left and right pulmonary arteries to maintain sufficient pressure in the LCA and right coronary artery. The main pulmonary artery was incised, and the location of the orifice of ALCAPA was identified according to which 2 types were

TABLE 1. Clinical data in 10 patients

Patient no.	Gender	Age (mo)	Weight (kg)	Height (cm)	EF (%)	FS (%)	Mitral regurgitation	LV aneurysm (mm ²)	CPB time (min)	ACC time (min)	Surgical techniques (in addition to button transfer)	Death or follow-up period (mo)
1	Male	3	5.5	56	45	22	Mild	No	326	118		Death
2	Female	21	8	78	56	29	Severe	30 × 50	192	115	MV annuloplasty, aneurysm patch repair	72
3	Female	8	5.9	62	45	22	No	No	178	95		2
4	Male	132	42	150	48	24	No	No	226	58	Unroofing	18
5	Male	14	9	80	36	17	Severe	50 × 60	196	129	Unroofing, MV annuloplasty aneurysm patch repair	Death
6	Female	28	11	89	45	22	Severe	No	123	59	Unroofing, MV annuloplasty	12
7	Male	62	16.3	104	39	10	No	20 × 20	164	71	Unroofing, aneurysmal plication	13
8	Male	19	9.7	81	29	15	Mild	15 × 15	105	62		5
9	Male	21	12	94	47	23	No	20 × 20	148	68	Aneurysmal plication	72
10	Female	6	5	61	64	32	Severe	No	111	62	MV annuloplasty	4
Mean ± SD		31 ± 39	12 ± 11	85 ± 27	45 ± 10	22 ± 6			177 ± 66	84 ± 27		25 ± 30

EF, Left ventricular ejection fraction; FS, left ventricular fractional shortening; LV, left ventricular; CPB, cardiopulmonary bypass; ACC, aortic cross clamp; MV, mitral valve; SD, standard deviation.

classified. In the first type in 3 patients, the orifice of AL-CAPA was at the bifurcation of the main pulmonary artery and RPA; in the second type in 7 patients, it was more distal along the RPA from the bifurcation. In both types, it gave rise to the bridging portion of the LCA between the RPA and aorta. The LCA then entered the aortic wall, forming the intramural segment of 3 to 5 mm. It descended vertically in the aorta before emerging from the aortic wall over the left of the sinus of Valsalva to branch in the usual manner to supply the left ventricle. In the first type, the bridging portion of the LCA was relatively longer to allow sufficient mobilization. Reimplantation using a direct button transfer was performed. A horizontal incision was made at the left side of the ascending aorta at the same level of the LCA orifice. The collar of the button around the LCA orifice was made approximately 5 mm to allow sufficient tissue for anastomosis to the aorta incision and to form the “fish eye” shape to avoid tension or distortion of the LCA (Figure 1). In the second type, the bridging portion of the LCA was fairly short. The button transfer was combined with the unroofing technique, whereby the bridging portion of the LCA from the orifice and the aortic intima of the intramural segment were incised throughout and anastomosed with the horizontal incision of the ascending aorta (Figure 2). The incision of the aorta was directly closed. The incision of the main pulmonary artery was closed with a pericardial patch.

In 4 patients with severe mitral regurgitation, annuloplasty was performed with sutures at both commissures of the mitral valve. No repair was made in 2 patients with mild to moderate mitral regurgitation. Among the 5 patients with ventricular aneurysm, the first 2 patients with a large aneurysm ($30 \times 50 \text{ mm}^2$ in patient 2, and $50 \times 60 \text{ mm}^2$ in patient 5; Figure 3) had aneurysmal exclusion by closing the aneurysmal collar with a Dacron patch. One of them died of cardiac failure (patient 5). In the subsequent 2 patients (patients 7 and 9) with a small aneurysm

($20 \times 20 \text{ mm}^2$ for both), the aneurysm was longitudinally plicated without any incision and sutured using 3-0 Prolene with 2 small Dacron patches. No repair was made to the smallest ventricular aneurysm ($15 \times 15 \text{ mm}^2$) in patient 8.

Cardiopulmonary bypass was maintained for 177 ± 66 minutes and aortic cross clamping for 84 ± 28 minutes.

Patient Outcomes

In the early postoperative period, 2 patients died of cardiac failure (patients 1 and 5). Patient 1 was our first patient. Patient 5 had the largest left ventricular aneurysm with a Dacron patch repair. Others had an uneventful recovery.

The follow-up period in the 8 survivors ranged from 2 months to 6 years (median 1 year). Echocardiography showed the mean left ventricular ejection fraction $67\% \pm 6\%$ and mean fractional shortening $32\% \pm 3\%$, significantly increased as compared with preoperative values ($P = .01$ for both). Among the 4 patients who underwent mitral valve repair, mitral regurgitation became mild. Others did not show mitral regurgitation.

DISCUSSION

The present study reviewed our diagnostic and surgical experience in 10 children with an extremely rare association of congenital heart anomalies of ALCAPA arising from the RPA with an intramural course of the coronary artery. Our series, although a small cohort, is the largest series of children to date.

The incidence of ALCAPA arising from the RPA is substantial in our institute, accounting for 22% of all the ALCAPA cases during the study period. Importantly, all the 10 cases with such an anomaly were associated with an intramural aortic course but without other congenital heart defects. Reviewing the literature shows that the association has been described in many of the case reports of ALCAPA arising from RPA without other congenital heart defects,¹³⁻¹⁵ except a few^{16,17} that were reported from

FIGURE 1. A, Anatomy of the anomalous origin of the LCA from the bifurcation of the right and MPA with the intramural aortic segment. B, Operative illustration of the button transfer showing a 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, which is detached and switched anteriorly. C, Operative illustration of anastomosis of the button to the aorta incision. LCA, Left coronary artery; Ao, aorta; MPA, main pulmonary artery; RPA, right pulmonary artery.

FIGURE 2. A, Anatomy of the anomalous origin of the LCA from the RPA more distal to the bifurcation with the intramural aortic segment. B, Operative illustrations showing that the short bridging portion of the LCA and the aortic intima of the intramural segment were incised. C, Operative illustrations of anastomosis of the button to the aorta incision. LCA, Left coronary artery; Ao, aorta; MPA, main pulmonary artery; RPA, right pulmonary artery.

earlier years, and the aortic intramural course might have been unnoticed. Of note, preoperative diagnosis of the intramural aortic course of the anomalous LCA is difficult. Similar to the report by Atik et al,¹⁵ all of our patients were diagnosed during the operation. As such, careful attention should be paid to the potential association of an aortic intramural course before the operation as well as during the operation when dealing with ALCAPA from the RPA.

Most ALCAPA cases present during infancy in the early months after birth, and approximately 20% of patients present in childhood or even as adults.^{5,7,8} It is generally considered that early diagnosis and surgical treatment are vital; if untreated, a mortality rate of up to 90% results within the first year of life.^{5,6} This is not exactly the case in our series. Most of our patients (7 of 10) presented after infancy from 14 months to 11 years old, and were

complicated with congestive heart failure, mitral valve regurgitation, and left ventricular aneurysm. Their clinical manifestations, such as mitral regurgitation and left ventricular aneurysm, were not related to age, likely attributable to the varied degree of development of coronary collaterals.^{18,19} Should the collateral circulation be well developed and distributed, the left ventricle may remain well perfused, the heart retaining its essentially normal form and function. This may be the case in the 11-year-old patient (patient 4). In situations in which the collateral circulation is poorly developed, the left ventricle becomes ischemic, infarcted, and fibrosed with mitral valvar incompetence, as happened to other patients.

Reimplantation to establish a 2-coronary arterial system is the standard technique for ALCAPA.²⁰ A few cases of ALCAPA arising from the RPA have been reported to have undergone successful direct reimplantation.^{9,11,12}

FIGURE 3. Two computed tomograms of apical view (A) and lateral view (B) showing the large LV aneurysm of $50 \times 60 \text{ mm}^2$ in Patient No. 5. LV, Left ventricular.

Direct reimplantation with button transfer of the LCA orifice was performed in all the patients in our series. In the 3 patients with the LCA arising at the bifurcation of the main pulmonary artery and RPA, the bridging portion was relatively longer and sufficient to allow extensive mobilization of the LCA. In these patients, only direct button transfer was performed, leaving the intramural segment untouched. However, the intramural coronary artery is known to pose potential risks of myocardial ischemia and even sudden death.^{21,22} In the 7 patients with the orifice of LCA located higher at the RPA, the limited length of bridging portion may pose the risks of tension or distortion of the LCA. Unroofing of the bridging portion of the LCA and intramural aortic segment was performed to extend the neo-orifice of the LCA.¹³ The combination of button transfer and unroofing procedure largely normalizes an entire course of the LCA without leaving the intramural segment and allows wider orifice of the LCA. Therefore, we consider the combination of button transfer and unroofing procedure is superior to button transfer alone, as well as to the previously advocated unroofing alone.¹³

The management of the regurgitant mitral valve at the time of ALCAPA repair has raised some controversy.^{23,24} Although some suggest that mitral regurgitation should be repaired at the time of coronary transfer,²⁵ most believe that mild to moderate regurgitation will improve with the improved left ventricular function after coronary transfer. We, like others,^{24,26-28} advocate mitral valve repair only for severe mitral regurgitation in the setting of ventricular dysfunction.

Little has been reported about the association of ALCAPA and left ventricular aneurysm resulting from infarction.²⁹ This is surprising to us, because 5 of 10 patients had a left ventricular aneurysm in our series. In the first 2 patients (patients 2 and 5) with large left ventricular aneurysm, aneurysm exclusion was performed by closing the aneurysmal collar with a Dacron patch, which severely affected left ventricular function. One patient died of cardiac failure early after the operation (patient 5). Since then, the “no-patch” technique was used with plication of the scar tissue.³⁰ Similar technique has been used for the Ebstein anomaly with repair with longitudinal plication of the atrialized chamber and the right atrium to reconstruct the right ventricle and reduce the size of the dilated right atrium.^{31,32} This technique used for the repair of left ventricular aneurysm in ALCAPA features several major advantages; namely, not having a ventricular incision, saving cardiopulmonary bypass time, and, more importantly, reestablishing a physiological-like ventricular function and morphology closest to the one before myocardial infarction.

In the 8 survivors, the left ventricular function improved remarkably at follow-up, consistent with previous reports.^{14,15,33-35}

Limitations

The follow-up period was fairly short in our patients, which is a general problem in current Chinese clinical practice in pediatric cardiology and surgery.

CONCLUSIONS

Careful attention should be paid to the extremely rare association of an aortic intramural course before and during an operation when dealing with the anomalous LCA from RPA. Surgical strategies for the associated anomalies include coronary button transfer or/and unroofing technique and provide encouraging outcomes. Coronary button transfer combined with unroofing of the intramural segment is recommended because it allows a more secured perfusion of the LCA without leaving risks of myocardial ischemia imposed by the intramural course.

Conflict of Interest Statement

Authors have nothing to disclose with regard to commercial support.

We are grateful to Professor Igor E. Konstantinov from the Royal Children's Hospital in Melbourne for providing precise surgical technique illustrations for this article.

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Key Words: surgery, anomalous origin of the left coronary artery, intramural aortic course