RESEARCH

Surgical treatment of anomalous aortic origin of the coronary artery in paediatric patients: a Chinese single-center experience

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Abstract

Purpose An anomalous aortic origin of the coronary artery (AAOCA) is a rare congenital heart disease. Some highrisk anatomical structures are at risk of inducing cardiogenic shock or even sudden death. This article summarizes our surgical experience with AAOCA in paediatric patients.

Methods We retrospectively analysed the clinical data of 27 paediatric AAOCA patients admitted to the Department of Cardiothoracic Surgery in our hospital from July 2015 to June 2023 and summarized the surgical treatment experience and follow-up results.

Results A total of 27 patients were included in this study, including 14 patients with an anomalous left coronary artery (ALCA) and 13 patients with an anomalous right coronary artery (ARCA). A comparison of clinical data between ALCA and ARCA patients revealed that the preoperative left ventricular ejection fraction (LVEF) in ALCA patients was significantly lower than that in ARCA patients (p < 0.05). There were significantly more patients with preoperative complications, such as major adverse cardiovascular events (MACEs) and mitral regurgitation (MR), in the ALCA group than in the ARCA group (p < 0.05). No postoperative adverse events such as severe bleeding, mediastinitis, central nervous system complications, the need for reoperation, pacemaker implantation, pleural effusion complications occurred after operation. The duration of follow-up was 58.5 (31.5, 77.3) months. During the follow-up period, none of the patients presented symptoms such as chest tightness or chest pain, and cardiac CTA revealed unobstructed coronary arteries. Compared with the preoperative LVEF, the LVEF significantly improved at the last follow-up (p < 0.05).

Conclusions Patients with an AAOCA should be taken seriously, and surgical treatment should be considered for these patients. Surgery should be considered for patients with ALCA as well as patients with ARCA with symptoms of myocardial ischaemia or a positive diagnosis of myocardial ischaemia or ventricular arrhythmia. For patients with other congenital heart defects that require surgical treatment, if the AAOCA is a high-risk anatomical structure,

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simultaneous surgery should be considered. The surgical method should be tailored to the coronary artery anatomy of the individual patient.

Keywords Anomalous aortic origin of the coronary artery, Major adverse cardiovascular events, Surgery, Paediatric

Introduction

An anomalous aortic origin of the coronary artery (AAOCA) is the second most common cause of sudden cardiac death after hypertrophic cardiomyopathy in children and adolescents [1, 2, 3, 4]. This is a rare congenital anomaly in which the left, right, or bilateral coronary arteries do not originate from the respective sinus of Valsalva but rather exhibit abnormal walking [5]. The prevalence of AAOCA ranges from 0.06 to 0.9% for abnormal right coronary arteries (ARCAs), 0.025-0.15% for abnormal left coronary arteries (ALCAs), and 0.02-0.67% for abnormal circumflex coronary arteries [5, 6, 7, 8]. Although most AAOCA are considered benign, an anomalous left coronary artery originating from the right sinus of Valsalva and an anomalous right coronary artery originating from the left sinus of Valsalva are associated with cardiac symptoms and sudden death [9]. The clinical manifestations of an AAOCA vary greatly, and most patients have no obvious symptoms during the early stages. Thus, the early diagnosis of an AAOCA can be challenging. Furthermore, there is controversy regarding the surgical indications for an AAOCA due to its diverse anatomical variations, especially among asymptomatic patients [10]. In this study, we summarized our surgical experience in treating patients with AAOCA.

Methods

We retrospectively analysed the clinical data of 27 paediatric AAOCA patients admitted to the Department of Cardiothoracic Surgery in our hospital from July 2015 to June 2023 and summarized the surgical treatment experience and follow-up results. All patients were diagnosed with an AAOCA by echocardiography and cardiac CTA.

The inclusion criterion for patients was surgical treatment for an AAOCA. The exclusion criteria for patients were as follows: (1) Severe heart failure before the operation; (2) Patients with multiple organ failure before the operation; (3) Patients who were lost to follow-up after discharge; (4) The parents of patients refused to participate in the study.

Surgical techniques

We performed median sternotomy. Cardiopulmonary bypass was established via the ascending aorta, superior vena cava, and inferior vena cava. After cardiac arrest, the aorta was cut open to explore the location and distribution of the abnormal coronary arteries. The appropriate surgical procedure was selected according to the different anatomical characteristics of the coronary artery.

Coronary unroofing

Unroofing of an intramural segment is the most common surgical procedure performed for AAOCA. This procedure is suitable for AAOCA patients with an anomalous coronary artery immediately above and to the side of the intercoronary commissure. Thus, the surgeon is able to incise the intramural segment without disrupting the support mechanism of the aortic valve. The operation steps are as follows [11].

- 1. A coronary probe was used to explore the direction and range of the intramural segment and determine the relationship between the intramural segment and the inner wall of the aorta.
- 2. This procedure consists of incising, and occasionally excising, the intramural segment of the anomalous coronary inside the aortic root and, by doing so, enlarging the ostium and virtually moving the ostium to the correct sinus. (Fig. 1)

Coronary transection and reimplantation

Transection and reimplantation of the coronary artery are performed when unroofing does not relocate the ostium to the appropriate sinus or would result in compression by the intercoronary pillar. The procedural steps are as follows [11].

- 1. The anomalous coronary artery ostium is excised from the aortic sinus, and a portion of the aortic wall tissue is preserved as a pedicle graft.
- 2. Complete release of the anomalous coronary artery ostium is performed to prevent stenosis caused by excessive tension during implantation.
- 3. An incision is made at an adequate location within the correct coronary sinus.
- 4. The anomalous coronary artery is anastomosed to an adequate location without tension. (Fig. 2)

For patients who obtain incomplete relief of coronary artery compression through conventional treatment, adjunctive procedures such as pulmonary artery transposition and aortic sinus enlargement surgery can be performed during the operation to further optimize the anatomical space available for coronary arteries and ensure adequate myocardial perfusion. Pulmonary artery translocation involves transecting the main pulmonary artery at the left and right pulmonary artery bifurcation, extending the incision to the pulmonary artery, suturing the right pulmonary artery end incision with a pericardial



Fig. 1 Process of coronary unroofing: A longitudinal section of the common wall of the aorta was cut along the direction of the coronary artery using a coronary blade or fine scissors to remove the limitation of the top of the coronary artery, extending the opening into the correct aortic sinus



Fig. 2 The process of coronary transection and reimplantation: The coronary artery of anomalous origin is excised from the aortic sinus. Then, an incision is made at the normal sinus position of the aortic root, followed by transplantation and suturing of the coronary graft

patch to increase the anteroposterior diameter of the junction, and reanastomosing the main pulmonary artery and pulmonary artery bifurcation junction to relocate the pulmonary artery trunk towards the left (Fig. 3). Special attention should be paid to avoid left pulmonary artery stenosis during pulmonary artery translocation. If there is a risk of left pulmonary artery stenosis, preventive widening of the left pulmonary artery should be performed. Aortic sinus enlargement surgery can be employed to augment the aortic sinus using an autologous pericardial patch, thereby enhancing blood supply to the coronary artery.

Follow-up management

Following discharge, all patients received standard antiplatelet therapy with aspirin at a dosage of 5 mg/kg/d for six months. Subsequent follow-up assessments were conducted at one month, three months, six months and one year after discharge, followed by annual evaluations thereafter. These assessments included electrocardiography and echocardiography. Cardiac CTA was reviewed six months after discharge. If normal, patients were then reviewed annually. The end point of follow-up of this study was June 2023 or death during this period.

Statistical analysis

SPSS 22.0 software was used for statistical analysis. Quantitative data are presented as medians (interquartile



Fig. 3 The process of pulmonary artery translocation involves transecting the main pulmonary artery at the left and right pulmonary artery bifurcation, extending the incision to the pulmonary artery, and reanastomosing the main pulmonary artery and pulmonary artery bifurcation junction to relocate the pulmonary artery trunk towards the left

ranges), and nonparametric tests were used for comparisons between groups. Count data were analysed using the chi-square test. P < 0.05 was considered to indicate statistical significance.

Results

A total of 27 patients were enrolled in this study, including 14 patients with ALCA and 13 patients with ARCA (Fig. 4). During the course of the disease, 18 patients exhibited varying degrees of myocardial ischaemia, presenting clinical symptoms such as postactivity chest pain, fatigue, feeding difficulties, and cardiogenic syncope. Among the 9 asymptomatic patients, 3 had other congenital heart diseases, whereas physical examination revealed abnormal electrocardiogram (ECG) results in 2 patients. Eighteen patients underwent coronary unroofing, and 9 patients underwent coronary transection and reimplantation. Among them, pulmonary artery transposition was performed in 6 patients, and aortic sinus patch enlargement was performed in 1 patient. The duration of cardiopulmonary bypass was 71 (51, 102) minutes, and the duration of aortic clamping was 48 (30, 68) minutes. (Table 1)

No postoperative adverse events such as severe bleeding, mediastinitis, central nervous system complications, the need for reoperation, pacemaker implantation, pleural effusion complications occurred after operation.

A comparison of clinical data between ALCA patients and ARCA patients revealed that the preoperative left ventricular ejection fraction (LVEF) in ALCA patients was significantly lower than that in ARCA patients (p < 0.05). The preoperative complications of major



Fig. 4 A: Left coronary artery originating from the right coronary sinus running through the aortic wall; B: Right coronary artery originating from the left coronary sinus running through the aortic wall

Table 1 Clinical features in AAOCA patients

	Number
Cases	27
Age (month)	105 (75, 152)
Weight (kg)	28.7 (19, 40.7)
Gender	
Male	19 (70.4%)
Female	8 (29.6%)
Types of AAOCA	
ALCA	14 (51.9%)
ARCA	13 (50.1%)
Associated malformation	
LVOTO	1 (3.7%)
ASD	1 (3.7%)
CSSD	1 (3.7%)
Symptomatic	18 (66.7%)
Surgical methods	
Coronary unroofing	18 (66.7%)
Coronary transection and reimplantation	9 (33.3%)
LVEF (%)	59.8 (38, 66.7)
LVEF<50%	11 (40.7%)
ECG abnormalities	16 (59.3%)

ALCA: anomalous left coronary artery, ARCA: anomalous right coronary artery, ASD: Atrial septal defect, LVOTO: left ventricular outflow tract obstruction, CSSD: coronary sinus septal defect, ECG: electrocardiograph, LVEF: left ventricular ejection fraction

adverse cardiovascular events (MACEs) and mitral regurgitation (MR) in ALCA patients were significantly greater than those in ARCA patients (p < 0.05). One patient with ALCA needed extracorporeal membrane oxygenation treatment after surgery, but none of the patients with ARLCA needed extracorporeal membrane oxygenation treatment. (Table 2)

 Table 2
 Comparison of clinical data between ALCA and ARCA patients

1			
	ALCA	ARCA	Р
Number	14	13	
Age (month)	121.5 (74.3, 178.5)	101 (62, 142.)	0.382
Weight (kg)	28.9 (17.5, 53)	28 (23, 37.8)	0.790
Preoperative LVEF (%)	47.8 (31.2, 61.6)	66 (46.5, 68.8)	0.022
MACE	7	1	0.016
Associated MR	4	0	0.037
Postoperative ECMO	1	0	-

LVEF: left ventricular ejection fraction, MACE: major adverse cardiovascular events, MR: mitral regurgitation, ECMO: Extracorporeal Membrane Oxygenation

There were no deaths, and no patients were lost to follow-up. The duration of postoperative intensive care unit stay was 3 (2, 5) days, whereas the length of postoperative hospital stay was 12 (9, 14) days. The duration of follow-up was 58.5 (31.5, 77.3) months. During the follow-up period, none of the patients exhibited symptoms such as chest tightness or chest pain, and cardiac CTA revealed unobstructed coronary arteries. Compared with the preoperative LVEF, the LVEF significantly improved at the last follow-up [59.8 (38, 66.7) vs. 65.5 (61.9, 69), p = 0.038]. At the last follow-up, only 3 patients presented with an LVEF less than 50, and only 3 patients presented with ECG abnormalities.

Discussion

Owing to the absence of obvious symptoms in many patients with AAOCAs and in some symptomatic patients, the symptoms occasionally resolve spontaneously, or the condition might be easy to ignore, which leads to challenges during diagnosis and an increased risk of missed or incorrect diagnoses. In this study, 33.3%

(9/27) of the patients presented no clinical symptoms. Among symptomatic patients, older individuals generally present with a prolonged disease course, with some experiencing myocardial ischaemia symptoms persisting for more than 2 years. Echocardiography has high accuracy in evaluating intracardiac structure and cardiac function. However, the assessment of coronary opening position and intramural running yields suboptimal results. In this cohort, 3 patients were initially misdiagnosed by echocardiography, resulting in a misdiagnosis rate of 13.7% (3/27). Among these patients, 1 patient was diagnosed with cardiomyopathy, 1 patient was diagnosed with Kawasaki disease complicated by coronary aneurysmal dilation, and 1 patient was diagnosed with an anomaly of the left coronary artery from the pulmonary artery. CTA is a Class I recommended examination method for the diagnosis of the AAOCA [10]. This method provides a complete image of the coronary artery contour, opening, valve sinus, intramural running range, degree of stenosis and other anatomical characteristics. However, CTA cannot be used as a routine screening tool because of the presence of radiation and its high cost.

The treatment of AAOCA has been controversial worldwide [12, 13]. A study conducted by Texas Children's Hospital revealed that AAOCA seriously affects the quality of life and health of children as well as the quality of life of their families [14]. The high-risk anatomical structures causing sudden cardiac death in AAOCA patients include the intramural coronary artery and the main coronary artery running between the aortic and pulmonary arteries [15, 16]. The guidelines from the American College of Cardiology/American Heart Association provided clear surgical indications for AAE-CAs [17]. Grade I indications include ALCA and ARCA patients exhibiting symptoms of myocardial ischaemia or positive diagnostic tests for myocardial ischaemia. Grade IIa indications include ALCA patients without symptoms of myocardial ischaemia and negative diagnostic tests, as well as ARCA patients without symptoms of myocardial ischaemia and negative diagnostic tests but with ventricular arrhythmias. Grade IIb indications are applicable to ARCA patients without symptoms of myocardial ischaemia and negative diagnostic tests but who lack ventricular arrhythmias. A multicentre clinical study proposed a classification of surgical indications based on the anatomical structure of the AAOCA [18]. (1) There are strong surgical indications for patients with myocardial ischaemia caused by compression of the abnormal left main coronary artery between the aortopulmonary artery or intramural aortic wall. (2) Beneficial surgical indications are noted for patients with myocardial ischaemia caused by obstruction of coronary blood flow due to vascular wall dysplasia. (3) Reasonable surgical indications are cited for patients with partial anterior descending branches running between the aortopulmonary artery. Our study revealed significant differences between the ALCA group and the ARCA group in terms of preoperative left ventricular function, incidence of cardiovascular adverse events, and incidence of MR (P < 0.05). These findings reflected the obvious effect of ALCA on cardiac function. We approached the determination of surgical indications in a similar manner. Surgical treatment should be considered for the following patients: patients with ALCA, patients with ARCA with symptoms of myocardial ischaemia, or patients with a positive diagnosis of myocardial ischaemia or ventricular arrhythmia. For patients with other congenital heart defects that require surgical treatment, if the AAOCA is a high-risk anatomical structure, simultaneous surgery should be considered.

The surgical techniques for treating the AAOCA primarily include coronary unroofing, coronary transection and reimplantation [19, 20, 21]. However, there has been no formal comparison of the various methods. Therefore, it is not possible to determine which of them is superior. The choice of surgical approach depends on the morphology of the abnormality of the AAOCA [22]. For example, the coronary artery running through the aortic wall is usually repaired by coronary unroofing. Other anomalies associated with AAOCA are usually not suitable for unroofing and are instead treated by coronary transection and reimplantation and pulmonary artery translocation.

Coronary unroofing is the most frequently employed surgical technique and is also extensively utilized within this cohort. Its primary advantage lies in its ability to relocate the ectopic opening to the normal valve sinus without compromising the coronary ostium tissue by eliminating intimal constraints of the common aortic wall, thereby effectively augmenting the inner diameter of the coronary artery. However, in cases where the intramural segment of the AAOCA is located below the aortic valve junction, an unroofing operation may not only transfer the ectopic opening of the coronary artery but also potentially damage the annulus and valve structure. Therefore, coronary transection and reimplantation are necessary. The key to this procedure is to fully free both the coronary nipple and main stem of origin while maintaining a natural graft opening and avoiding longterm restenosis caused by traction, torsion, contracture, or angle formation. For patients with limited space in the angle between the aorta and the pulmonary artery, especially those with long intramural passages, simple treatment involving coronary opening cannot completely relieve compression from the two major arteries. The combined application of aortic valve sinus enlargement, pulmonary artery translocation, and other methods can further expand the coronary origin and the gap between the aorta and the pulmonary artery so that the coronary artery can obtain sufficient space to fill and grow.

Postoperative aortic regurgitation and coronary artery occlusion are complications that require vigilance. (11, 23-24) Each case should be carefully studied before surgery to select an appropriate surgical technique to eliminate all mechanisms that cause coronary ischaemia. The roof of the coronary artery should be removed as much as possible during the coronary unroofing operation. The factors leading to coronary ischaemia, such as the angle of anastomosis, stretching and mesh material deformation, should be considered during coronary transection and reimplantation operations. Damage to the aortic valve and its supporting structures should be avoided as much as possible during the operation. For example, when the intramural segment of the aorta of the AAOCA extends below the aortic valve, coronary unroofing may risk damaging the aortic valve. In such cases, coronary transection and reimplantation operations are more appropriate. Kohlsaat K et al. reported that operation of side-by-side anastomosis/aortocoronary window can decrease/avoid development of postoperative aortic regurgitation. This strategy is completed by anastomosing the side of the coronary artery to a newly created ostia in the middle of the appropriate coronary sinus (depending on anatomy), and removes the need for commissural manipulation completely [9]. Therefore, if necessary, this technique can also be used to prevent aortic regurgitation. Finally, close postoperative monitoring, standardized anticoagulation and strict follow-up are also essential.

There were several limitations in this study. First, the study was a single-centre retrospective study. The sample size of this study was small. In addition, the follow-up time of this study was short.

Conclusions

Patients with an AAOCA should be taken seriously, and surgical treatment should be considered for these patients. Surgery should be considered for patients with ALCA. Patients with ARCA with symptoms of myocardial ischaemia or a positive diagnosis of myocardial ischaemia or ventricular arrhythmia should also be considered for surgery. For patients with other congenital heart defects that require surgical treatment, if the AAOCA is a high-risk anatomical structure, simultaneous surgery should be considered. The surgical method should be tailored to the individual patient's coronary artery anatomy.

Abbreviations

AAOCA	Anomalous aortic origin of the coronary artery
ALCA	Anomalous left coronary artery
ARCA	Anomalous right coronary artery
LVEF	Left ventricular ejection fraction
MACEs	Major adverse cardiovascular events
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MR Mitral regurgitation

ECG Electrocardiogram

Acknowledgements

We acknowledge all the staff from our unit who participated in this study.

Author contributions

Kai Luo, Bin Ji, Xiao-Min He, and Jing-Hao Zheng designed the study, performed the statistical analysis, participated in the operation, and drafted the manuscript. Xiao-Yang Zhang, Yan-Jun Pan, Qi-Liang Zhang, Qiang Chen and Zhong-Qun Zhu collected the clinical data and participated in the operation. All authors read and approved the final manuscript.

Funding

This paper was supported by the Shanghai Talent Development Fund and the National Natural Science Foundation of China (82470410).

Data availability

The datasets of the current study are available from the corresponding author upon reasonable request.

Declarations

Ethical approval and consent to participate

The present study was approved by the ethics committee of Shanghai Children's Medical Center (SCMCIRB-K2024028-1) and adhered to the tenets of the Declaration of Helsinki. In addition, the parents or guardians of the patients provided written informed consent for their respective minors to participate in the study.

Consent for publication

Not applicable.

Competing interests

The authors declare no competing interests.

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Received: 19 March 2024 / Accepted: 16 January 2025 Published online: 23 January 2025

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