Anomalous Aortic Origin of the Coronary Arteries – State of the Art Management and Surgical Techniques

Carlos Bonilla-Ramirez, MD,[*,†] *Silvana Molossi, MD, PhD,*[*,‡] *Christopher A. Caldarone, MD,*[*,†] *and Ziyad M. Binsalamah, MD, FRCSC*[*,†]

Anomalous aortic origin of a coronary artery (AAOCA) can be associated with myocardial ischemia and sudden cardiac arrest. We describe and compare the management and surgical techniques for patients with AAOCA. Patients presenting to the Coronary Artery Anomalies Program are evaluated and managed following a standardized approach. Our approach and data were compared to other single-center and multi-institutional data and results. Patients with AAOCA present as an incidental finding approximately 50% of the time. Advanced axial imaging is essential to define the anatomic characteristics of this lesion. Preoperative and postoperative assessment of myocardial perfusion with provocative testing is feasible and contributes to risk stratification. The surgical techniques for AAOCA repair include coronary unroofing, transection and reimplantation, and neo-ostium creation, among others. In general, surgical repair of AAOCA can mitigate the risk of ischemia with low mortality. The specific morbidities and complications of each different technique should be considered during the surgical planning. Surgical repair of AAOCA can mitigate the risk of ischemia with a low associated mortality but with clinically relevant morbidities. Long-term follow-up is necessary to accurately balance the risks of repaired and unrepaired AAOCA.

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Surgical repair of anomalous aortic origin of the coronary artery.

Central Message

Surgical repair of AAOCA can mitigate the risk of ischemia with low mortality but significant morbidities. Long-term follow-up is essential to balance the risks of repaired and unrepaired AAOCA.

INTRODUCTION

Anomalous aortic origin of a coronary artery (AAOCA) is a congenital lesion where a coronary artery arises from an inappropriate location within the aorta (Fig. 1). Anomalous coronary arteries have been associated with myocardial ischemia and they are considered the second most common cause of sudden cardiac arrest (SCA) among young athletes.[1–4] The prevalence of AAOCA in the general population is unknown, although it has been estimated to range between 0.01% and 2%.[5] The mechanisms whereby AAOCA causes ischemia and SCA remain undefined. Furthermore, the clinical and anatomical features that increase the risk of ischemia and SCA are incompletely characterized.[4,6–8] Despite the unclear surgical indications and benefits, coronary artery surgical repair has been used to prevent or address SCA and myocardial ischemia.[9–15]

The surgical repair of AAOCA should yield an unobstructed coronary artery arising within the appropriate coronary sinus. Although unroofing of the intramural (IM) segment is more

Glossary: AAOCA, Anomalous aortic origin of a coronary artery; SCA, Sudden cardiac arrest; IM, Intramural; TAR, Transection and reimplantation; CAAP, Coronary artery anomalies program; CTA, Computed tomography angiography; sNPI, stress nuclear perfusion imaging; sCMR, Stress cardiac magnetic resonance imaging; AAORCA, Anomalous aortic origin of the right coronary artery; AAOLCA, Anomalous aortic origin of the left coronary artery

*Coronary Artery Anomalies Program, Texas Children's Hospital; Houston, Texas

†Division of Congenital Heart Surgery, Michael E. DeBakey Department of Surgery, Texas Children's Hospital, Baylor College of Medicine; Houston, Texas

‡The Lillie Frank Abercrombie Section of Cardiology, Department of Pediatrics, Texas Children's Hospital, Baylor College of Medicine; Houston, Texas

Address reprint requests to Ziyad M. Binsalamah, MD, Texas Children's Hospital, Baylor College of Medicine, 6651 Main Street, Legacy Tower, Houston, Texas 77030. E-mail: ziyadmss@gmail.com

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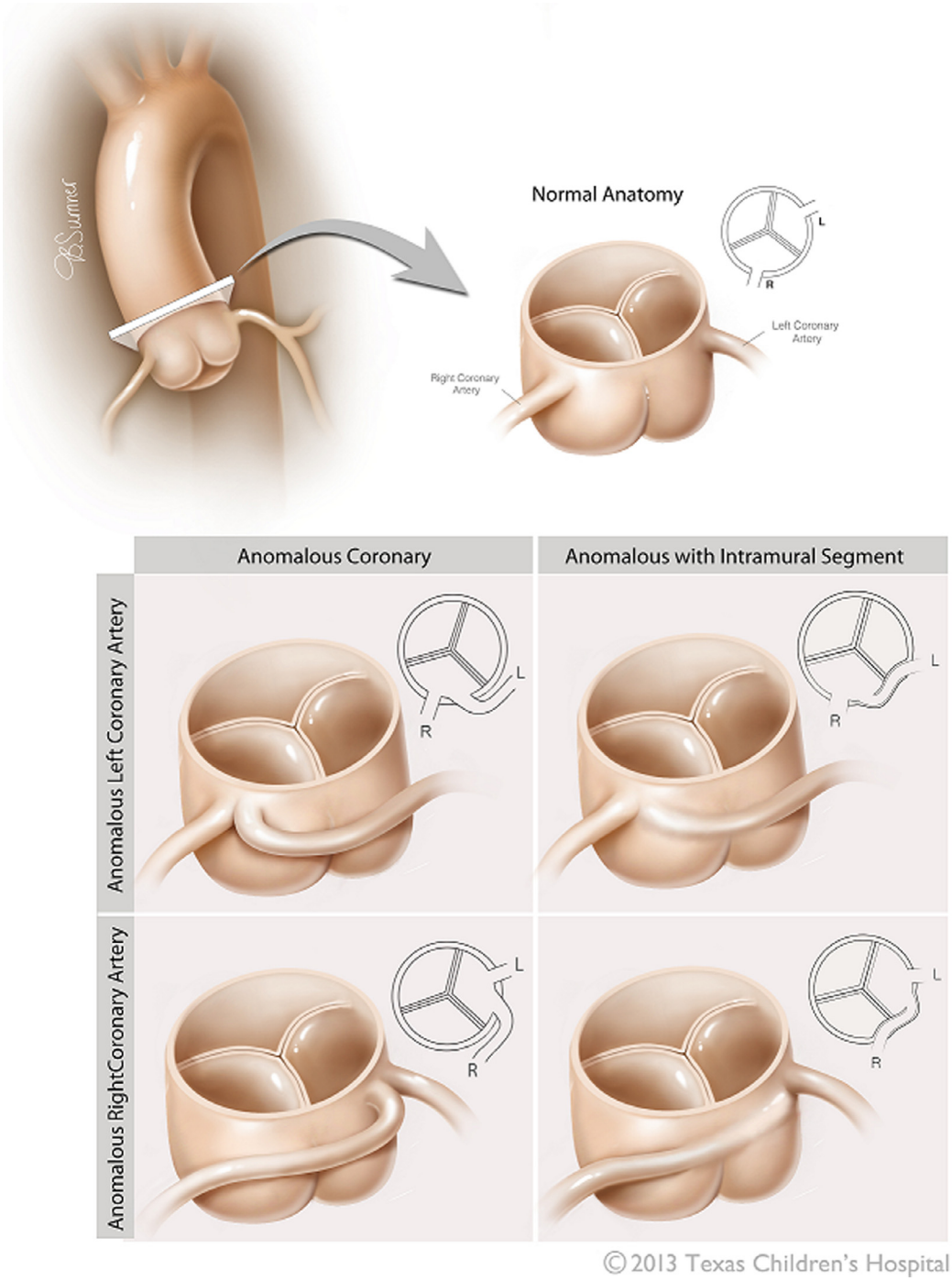


Figure 1 Anatomy of anomalous aortic origin of the coronary artery. Printed with permission from Texas Children's Hospital.

commonly performed, other techniques have been utilized, including transection and reimplantation (TAR) of the anomalous coronary and neo-ostium creation.[9,10,16–18] The benefits and drawbacks of each technique have been explored, although a comprehensive assessment has been hindered by the limited follow-up and the rare nature of the events they intend to prevent.[2,11,19,20]

The aim of this review article is to provide insight into the preoperative evaluation, surgical techniques, and postoperative outcomes of patients with AAOCA. We discuss the algorithm for the evaluation, surgical management, and follow-up of patients with AAOCA at Texas Children's. We compare our experience with other single-center and multi-institutional data.

THE CORONARY ARTERY ANOMALIES PROGRAM

The difficulties and questions surrounding the evaluation and management of AAOCA led to the creation of the

Coronary Artery Anomalies Program (CAAP) at Texas Children's Hospital in December 2012.[14] A consistent and multidisciplinary approach was a reasonable response to the limited data and uncertainties which will most likely remain until a longer follow-up time informs our field on the course of patients with repaired and unrepaired AAOCA. Additional to the clinical endeavors of the CAAP, all the patients presenting to the program have been approached for enrollment in Baylor College of Medicine's IRB-approved research studies (IRB H-30898, H-32955). The data of patients who have provided informed consent are prospectively collected for local and multi-institutional registries. Other centers like Lucile Packard Children's Hospital at Stanford, Children's Hospital of Philadelphia, and Boston Children's Hospital have opened similar programs to address this lesion in the pediatric population.

CLINICAL PRESENTATION

Approximately 50% of the anomalous coronaries are incidentally diagnosed while evaluating patients with a murmur, an electrocardiographic anomaly, or in preparticipation screening.[10,11,21–23] On clinical presentation, 40-60% of patients with AAOCA are symptomatic. The most common symptoms include chest pain, palpitations, syncope, and SCA.[3,10,11,23] An estimated 3%-5% of the diagnosed patients present with SCA and it has been reported that up to 45% of patients with SCA had symptoms prior to the event.[3,23,24] Although SCA tends to occur in athletes under exertion, the unpredictability of SCA and myocardial ischemia mandates a thorough evaluation of all patients diagnosed with AAOCA.[9,14,23]

PREOPERATIVE EVALUATION

At Texas Children's Hospital, all patients with AAOCA are evaluated with a standardized algorithm (Fig. 2).[23] Preoperative evaluation includes echocardiography, computed tomography angiography (CTA), and provocative ischemia testing. Although echocardiography can be used for diagnosis of AAOCA, it has limitations in the visualization of coronary anatomy features and poor inter-observer reliability has been reported.[25,26] Since the anatomical details can influence the surgical approach (described later), our CAAP team preoperatively evaluates CTA 3-dimensional reconstructions to topographically describe the coronary anatomy (Fig. 3), the coronary ostia, the IM course, the interarterial course, and the position of the ostium and IM course relative to the intercoronary pillar and commissure. Although cardiac magnetic resonance imaging has been used for axial imaging of the coronaries, we have favored the use of CTA because of its precise delineation of the IM course.[26–28] A thorough understanding of the anatomy allows for effective surgical planning.

At our institution, all patients undergo provocative ischemia testing with stress nuclear perfusion imaging (sNPI) and/or stress cardiac magnetic resonance imaging (sCMR). Patients presenting after SCA do not undergo provocative testing. Early in the program, sNPI was favored for ischemia testing. However, there were problems in the interpretation of sNPI, including decreased spatial resolution, artifacts caused by movement of the diaphragm, and perfusion defects that did not correlate with the anomalous coronary perfusion area. Based on the resources available at our center, we decided to transition to sCMR, which has been more reliable and has yielded more consistent results.[23] Nonetheless, sNPI or alternative provocative ischemia tests (e.g., positron emission tomography) could be performed at other institutions based on their resources and their experience.[3,10,11,23] At our institution, we do not perform fractional flow reserve nor intravascular ultrasound studies unless the patients have an intraseptal course.[29,30]

All patients in the CAAP are presented in our multidisciplinary meetings, where management is discussed. While patients with anomalous aortic origin of the left coronary artery (AAOLCA) represent 20%-25% of all patients with AAOCA, they account for up to 60% of AAOCA patients with evidence of ischemia and up to 85% of AAOCA patients with SCA, with some of them having normal exercise stress tests prior to the arrest.[3,21,23,24] In consequence, patients with AAOLCA are offered surgical repair at our center, regardless of whether we find perfusion abnormalities on provocative ischemia testing. In contrast, patients with anomalous aortic origin of the right coronary artery (AAORCA) represent 75%-80% of all patients with AAOCA and they account for approximately 40% of AAOCA patients with evidence of ischemia and 15% of AAOCA patients with SCA.[3,23,24] Therefore, patients with AAORCA are offered surgical repair only if they have clear symptoms suggestive of ischemia (SCA, significant arrhythmias, exertional syncope) or positive provocative ischemia testing. Surgical repair might be considered based on anatomical features such as the length of the IM segment and the presence of ostial stenosis. For asymptomatic patients younger than 10 years of age, we favor clinical follow-up alone. Assessment of myocardial perfusion is indicated for symptomatic patients at any age and in asymptomatic patients who are 10 years of age or older.

SURGICAL REPAIR

Surgical repair of AAOCA should address or mitigate the risk of SCA and myocardial ischemia while minimizing the complications that might arise from surgical intervention. Although the pathophysiology of AAOCA is undefined, a list of probable culprits has been proposed, including compression of the IM segment, compression of the interarterial segment, stenotic/angulated ostium, and compression by the intercoronary pillar.[3,8–11,15,18,21–23] At our institution, we have been mindful of the role of the intercoronary pillar, a thickening of the aortic wall that extends from the intercoronary commissure to the sinotubular junction, supporting the commissure.[2,8,9] It is reasonable to consider all the potential culprits when planning the surgical approach for AAOCA.

UNROOFING OF THE INTRAMURAL SEGMENT

Coronary unroofing is a widely used technique which consists of the incision of the IM segment from the ostium to the

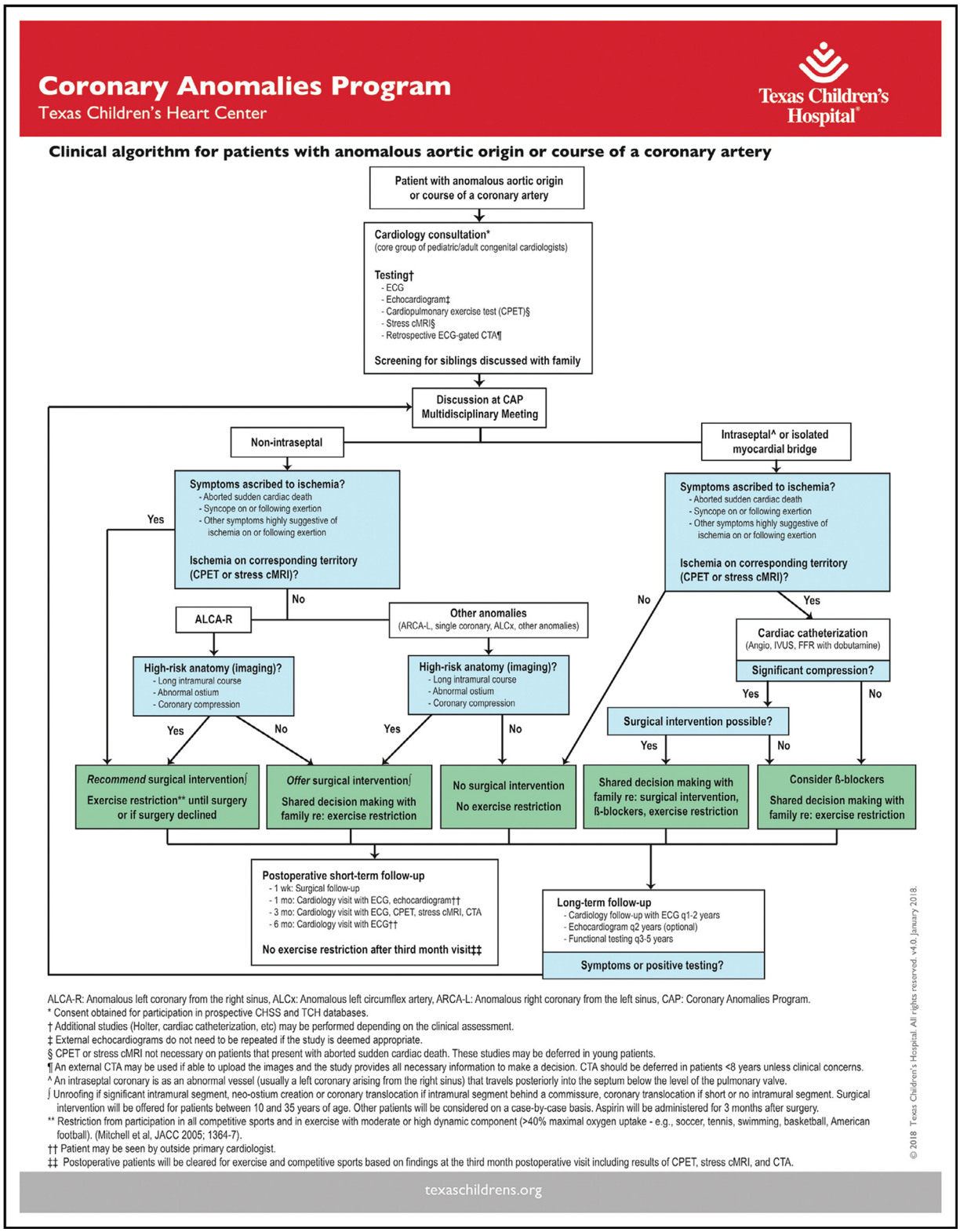


Figure 2 Algorithm for the management of patients with coronary anomalies at Texas Children's Hospital. Printed with permission from Texas Children's Hospital.

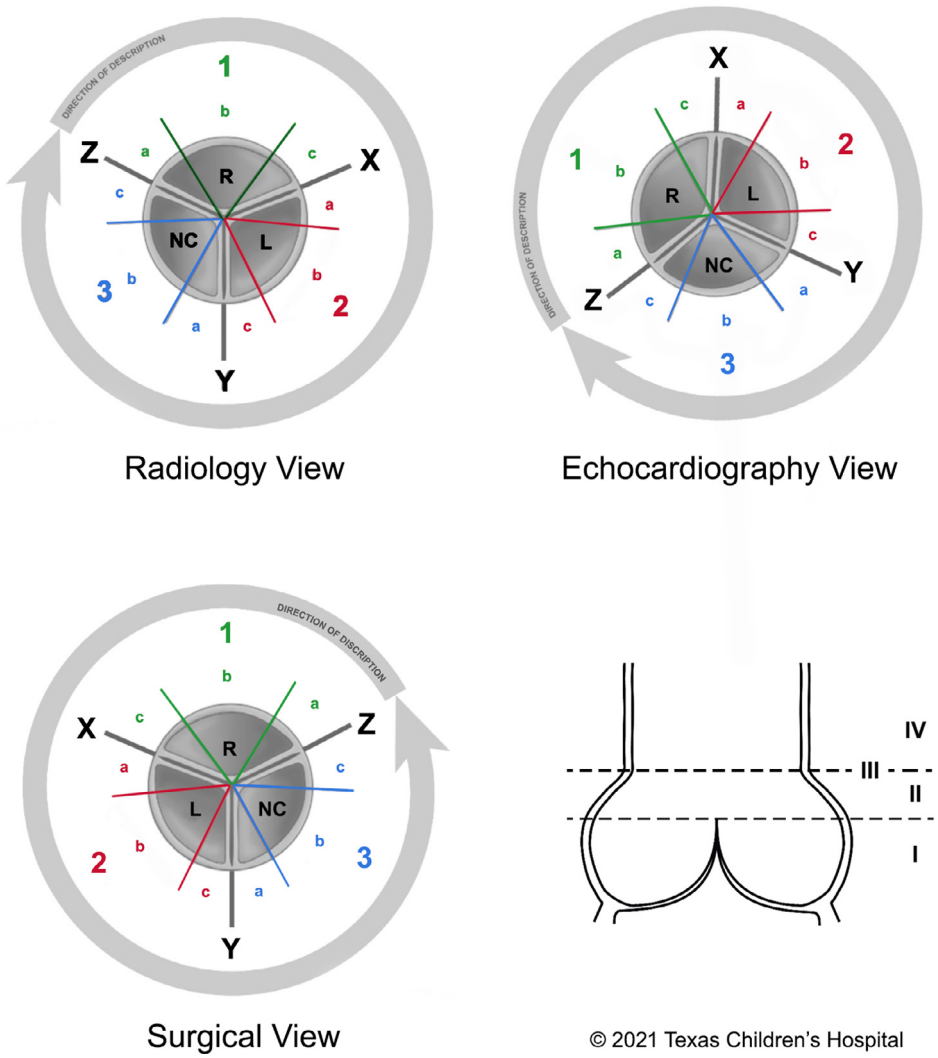


Figure 3 Topography map used for the description of the anomalous coronary arteries at Texas Children's Hospital. Printed with permission from Texas Children's Hospital.

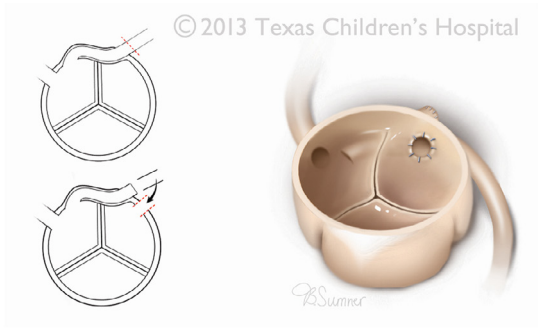
emergence of the anomalous coronary (Fig. 4). This technique avoids significant coronary manipulation, might relocate the coronary ostium to the correct sinus, and might mitigate the risk of ischemia in approximately 85%-95% of patients.[9,10,31–34] However, unroofing has its own drawbacks like partial compression by the pillar, coronary angulation, residual interarterial segment, residual IM portion, and postoperative aortic valve regurgitation.[9,11]

Postoperative aortic valve regurgitation has been reported to occur in 10%-17% of patients undergoing unroofing of an anomalous coronary artery. Although commissural manipulation does confer worse freedom from aortic regurgitation, patients without commissural manipulation can also develop postoperative aortic regurgitation, as unroofing without commissural takedown might still disrupt other structures supporting the aortic valve.[11,15,19,20] While other centers have successfully diminished their incidence of postoperative regurgitation by routinely resuspending the commissure, at our institution we favor TAR for coronary arteries with a course through or close to the commissure in order to avoiding manipulation of the commissure and aortic valve, with 100% freedom from aortic valve regurgitation at 4 years.[11,19,34]

Despite the reported safety and efficacy of coronary unroofing, it is worth noting that recurrent SCA has occurred in patients who have undergone this procedure, including 1 patient at our institution.[2,11,15] We have hypothesized that the recurrent SCA may be associated with the failure of unroofing to address all the potential culprits of SCA, including compression by the intercoronary pillar.[2,11,20,35] Therefore, we favor a coronary unroofing only when it can relocate the neo-ostium to the correct sinus without compression by the adjacent intercoronary pillar.

TRANSECTION AND REIMPLANTATION OF THE CORONARY

TAR is an alternative technique where the anomalous coronary artery is transected after its emergence and anastomosed to an adequate location within the correct coronary sinus without an aortic button (Fig. 4).[9,17,34,36] TAR avoids aortic valve



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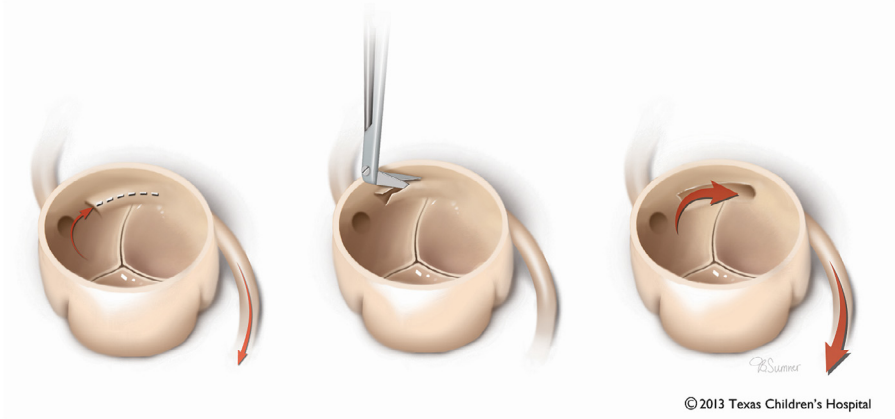

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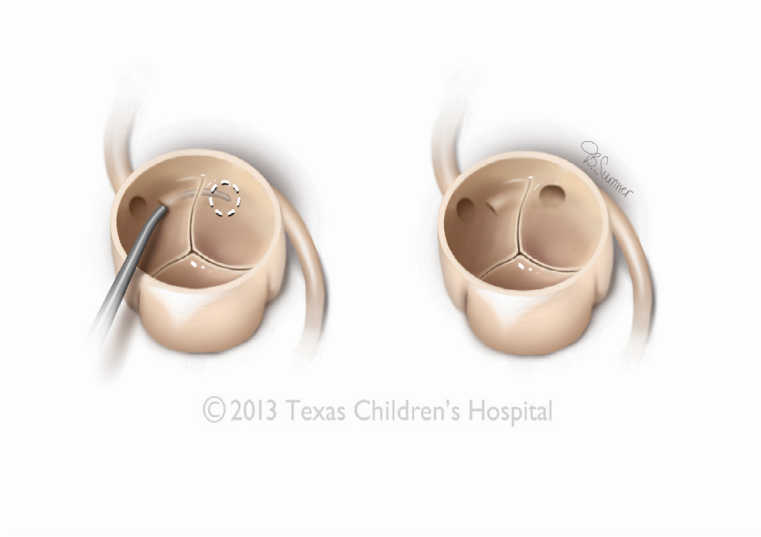

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Figure 4 Surgical technique for coronary artery transection and reimplantation (panel A), coronary unroofing (panel B) and neo-ostium creation (panel C). Printed with permission from Texas Children's Hospital.

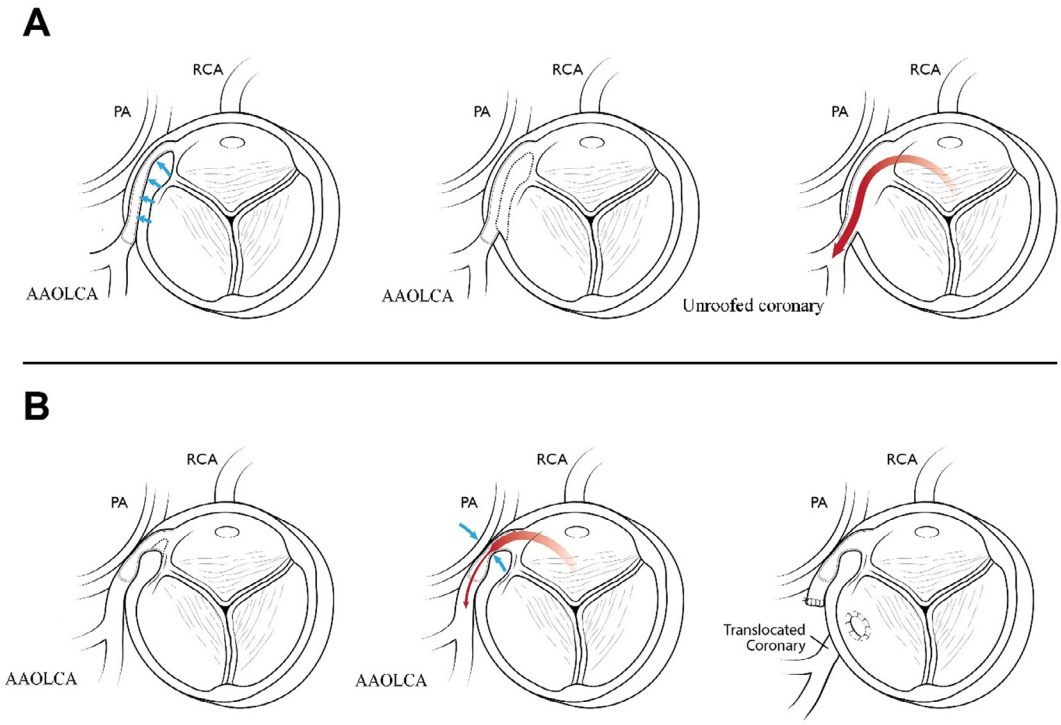

Figure 5 Effect of coronary unroofing according to position of the potential neo-ostium. In panel A (top), unroofing relocates the neo-ostium to the correct coronary sinus. In panel B (bottom), unroofing fails to relocate the neo-ostium and the coronary is compressed by the intercoronary pillar. Transection and reimplantation relocates the ostium to the correct coronary sinus. Printed with permission from Texas Children's Hospital. AAOLCA – Anomalous aortic origin of the left coronary artery; RCA- Right coronary artery; PA – Pulmonary artery.

disruption, eliminates residual IM segments and angulation, and relocates the ostium away from the intercoronary pillar. In exchange, there is significant coronary manipulation, potential for coronary twisting/kinking, and potential for scar-induced stenosis.[9,10,16,17,37]

At our institution, TAR is performed when unroofing does not relocate the ostium to the appropriate sinus or would result in compression by the intercoronary pillar. (Fig. 5) Among 16 patients who have undergone TAR at our center, 15 of 16 patients had negative postoperative testing and 1 patient underwent reoperation with a coronary artery bypass graft due to postoperative ischemia. Additionally, TAR was used for a reintervention on a patient who had recurrent SCA after a previous unroofing.[2,9,34] TAR has also been successfully used at other centers, with effective results and patency in all the translocated coronary arteries.[36,38] A variant of TAR uses a patch to augment the site of the anastomosis.[18] We avoid patches due to the risk of deformation and the implications of using patch material in a growing heart. Taking an aortic button as opposed to directly transecting the coronary may be acceptable if harvesting the button does not disrupt the aortic valve.

CREATION OF A NEO-OSTIUM

The creation of a neo-ostium is a technique that consists of unroofing the intramural portion located at the correct anatomic location of emergence of the anomalous coronary artery, without an incision of the entire intramural portion (Fig. 4).[9,39] At other centers, a variation has been described where an incision is made longitudinally in the proximal epicardial portion of the coronary and continued vertically in the correct coronary sinus. A patch is sutured into the aortocoronary incision to create the neo-ostium. With this technique, the intramurality, stenotic ostium, and angulated take-off are addressed without manipulation of the aortic valve.[18,40] However, the patch material has been reported to develop aneurysms or deformations which might cause stenosis or thrombosis.[18] Furthermore, depending on the site of emergence of the anomalous coronary, neo-ostium creation might yield a neo-ostium adjacent to the intercoronary pillar or arising from the incorrect coronary sinus.

OTHER TECHNIQUES

Although less commonly performed, a pulmonary artery translocation has been utilized to address interarterial compression by the pulmonary artery. Unroofing with patch plasty, coronary artery plasty, and combinations of these techniques have also been used.[31,41,42]

POSTOPERATIVE MANAGEMENT

At our institution, patients are postoperatively monitored with electrocardiography. Cardiac enzymes are measured only in presence of persistent electrocardiographic changes

suggestive of ischemia. Our recent review of our experience of ECG changes in our surgical cohort showed that ST segment changes occur in up to 75% of patients in the early postoperative period and do not correlate with ECG changes in a minority of patients seen at 3 months follow-up. Likewise, T wave abnormalities seen at discharge tend to resolve. Neither of these features correlate with need for reintervention.[43]

Acute pericarditis and pericardial effusions have been commonly reported (9%-46%) and we have managed this potential complication by leaving the right pleural space and pericardium open.[9,15,44] Patients are discharged on low dose aspirin (3-5 mg/kg/day) for 3 months postoperatively. They are reevaluated in clinic at 30 days after surgery.

OUTCOMES

In our center, all patients are re-evaluated at 3 months with CTA, exercise stress test, and stress perfusion imaging (sNPI or sCMR), regardless of the results of preoperative testing. The consistent use of postoperative ischemia testing has also been advocated for by other centers and organizations.[3,4,15,45] In our patient population, surgery has effectively mitigated the risk of ischemia in 57 of 64 (90%) of patients, with no associated mortality. Although surgical repair of AAOCA has also been reported to mitigate the short-term risk of ischemia in other studies (80%-95% resolution of ischemia after surgery), the long-term results of the surgical techniques are yet to be determined.[9,11,15,18,38,40] In our center, if the postoperative studies are reassuring, patients are released to exercise activities with no restrictions. To date, in our institution, 60/64 (94%) of patients have released to unrestricted exercise activities.

DISCUSSION

In this review, we discussed the clinical presentation, preoperative evaluation, surgical strategies, postoperative evaluation, and outcomes of patients with AAOCA. We compared our approach and strategies to data and results from other centers and multi-institutional registries. Despite the many questions that remain to be answered, there has been significant progress in the risk stratification, myocardial screening, and outcomes evaluation for patients with AAOCA.

The unknown pathophysiology of AAOCA, imperfect risk stratification, and rare but devastating nature of SCA are unique challenges that have required unique solutions and approaches. Surgical repair of AAOCA can mitigate the risk of ischemia with a low associated mortality but with clinically relevant morbidities.[9,11,15,18,20,23,38] At our institution, we have implemented the CAAP to consistently evaluate all patients with AAOCA. A consistent management is advisable and preoperative and postoperative ischemia testing should be undertaken for all patients.[3,21,45]

Although coronary unroofing is most commonly used, different surgical techniques have been proposed to address all the potential culprits in AAOCA and to solve the problems that can be encountered when performing a coronary unroofing, including a course through the commissure and potential compression by the intercoronary pillar.[8,9,15,18,19,31,34] Choosing the appropriate surgical technique depends on careful assessment of the anatomy, preoperatively with advance cardiac imaging and intraoperatively.

Although surgical repair of AAOCA can mitigate the risk of ischemia and provide excellent short-term outcomes, the potential surgical complications should be carefully considered. New postoperative aortic regurgitation has been identified in up to 10-17% of patients undergoing anomalous coronary repair and commissural manipulation has been associated with lower freedom from aortic regurgitation.[11,15,19,20] Every effort should be made to avoid disturbing the supporting mechanism of the aortic valve, which might occur even in absence of commissural manipulation.[15,19,20] In presence of a course through the commissure, alternative techniques like TAR or neo-ostium creation may represent better surgical options.[9,34,36,38]

Notably, recurrent SCA has been reported after coronary unroofing.[2,9,11] Therefore, every case should be carefully studied to choose a surgical technique that eliminates all possible mechanisms leading to coronary ischemia, such as the presence of IM segment, inter-arterial course, compression by the intercoronary pillar, and angulation. It is important to consider the significant technical challenges associated with the coronary artery manipulation in techniques like TAR or reimplantation with patch material. Although good clinical outcomes have been reported with these techniques, adverse events as coronary obstruction or patch material deformation which lead to coronary ischemia may occur.[18,34] Therefore, careful surgical planning and dedicated postoperative monitoring are essential.

Exercise activities are paramount for cardiovascular health. Surgical intervention in AAOCA aims at preventing myocardial ischemia and allowing patients to continue to lead a quality of life without restrictions. In our program, we seldom exercise-restrict patients. Following surgery and upon complete re-evaluation at 3 months post-operatively, patients are released to unrestricted exercise activities with progressive return to usual level of sports and/or exercise participation, unless remaining concerns. In our cohort, the great majority of patients are exercising after surgery with normal exercise capacity.[23]

The advancement of the knowledge and treatment of AAOCA depends on the continuity of the current efforts and on the use of novel and different approaches to the problem. Computational flow dynamics and advanced cardiovascular imaging might help to better understand the pathophysiology of the disease and the effects of the proposed culprits on the coronary flow.[46,47] Noninvasive imaging and 3-D modeling might help to have a better understanding of the anatomy.[27,48,49] Multi-institutional registries and collaboration among institutions will most definitely continue to improve the care of these patients.[3,15,45] Long-term follow-up will ultimately help to balance the risks of repaired and unrepaired AAOCA.

CONCLUSION

Surgical repair of AAOCA can mitigate the risk of ischemia with a low associated mortality but with clinically relevant morbidities. Long-term follow-up is essential to accurately balance the risks of repaired and unrepaired AAOCA.

SOURCES OF FUNDING

Internal

DECLARATION OF COMPETING INTEREST

None

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