

Review Article

Anomalous Coronary Artery Origins: A Practical Overview for the General Pediatrician

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Abstract

Anomalous aortic origin of a coronary artery (AAOCA) is a rare but potentially life-threatening congenital heart defect, occurring in approximately 0.4–0.8% of the population. While many patients remain asymptomatic, certain variants—particularly anomalous origin of the left coronary artery (AAOLCA) with interarterial or intramural courses—carry an elevated risk of sudden cardiac arrest (SCA), especially during exertion. Primary care providers play a critical role in the early identification and coordination of care for patients with AAOCA. Although routine electrocardiograms are typically normal, exertional chest pain or syncope should prompt referral to pediatric cardiology. Echocardiography is often the first-line diagnostic tool, but further imaging with cardiac computed tomography (CCT) and/or magnetic resonance imaging (MRI) is essential for confirming the diagnosis and risk-stratification. Identifiable high-risk anatomical features include a slit-like ostium, intramural course, and acute angle of take-off. Exercise stress testing is used for risk stratification but has low sensitivity and is recommended to be used with adjunctive testing such as stress perfusion imaging and occasionally cardiac catheterization. Surgical repair is indicated in patients with AAOLCA, symptomatic individuals, or those with demonstrable ischemia. Asymptomatic patients with AAORCA and low-risk features may be followed conservatively. Regardless of surgical

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status, all patients require lifelong cardiology follow-up, periodic imaging, and individualized sports clearance. Additionally, first-degree relatives may warrant screening due to potential familial clustering. AAOCA impacts quality of life, with patients and families often experiencing emotional distress due to lifestyle restrictions. Primary care providers should monitor for psychosocial concerns and coordinate mental health support as needed. This review aims to equip primary care clinicians with practical knowledge to ensure timely referral, appropriate counseling, and long-term support for children with AAOCA.

Keywords

Pediatrics, Anomalous Coronary Artery, Congenital Heart Disease

1. Introduction

Anomalous aortic origin of the coronary artery (AAOCA) occurs when the right or left coronary arteries do not arise from the usual origins from their respective aortic sinuses (Figure 1) [1]. Estimates of AAOCA prevalence range from 0.4%–0.8% [2]. While the specific mechanism of development of AAOCA during embryologic development remains unknown, an error in outgrowth of the arterial stems from appropriate sinuses has been postulated as the cause. Anomalous aortic origin of the right coronary artery (AAORCA) occurs three to eight times more frequently than anomalous aortic origin of the left coronary artery

(AAOLCA), with estimated incidences of 0.28%–0.92% and 0.03%–0.15%, respectively [3]. Although this is a rare occurrence among the general population, AAOCA, especially AAOLCA, carries a higher risk of sudden cardiac arrest/death (SCA/SCD). While it has been challenging to quantify the exact risk due to the infrequency of the event of interest and the uncertainty about the denominator (i.e. the prevalence of AAOCA), data from autopsy reports and SCA registries report a cumulative risk of death for those aged 15 to 35 years in sports to be 6.3% for AAOLCA and 0.2% for AAORCA [4, 5].

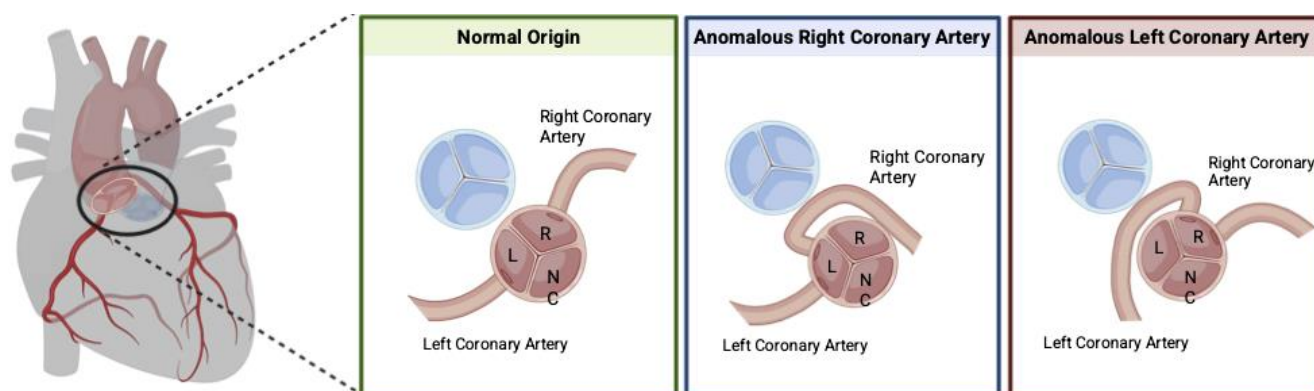


Figure 1. Diagrammatic Illustration of the Types of Anomalous Aortic Origins of Coronary Arteries. [Own work].

Within AAOCA, there are multiple variations defined by the anatomic courses of the anomalous artery which alter the overall risk profile (Figures 2 and 3). Agrawal et al pictorially described the six main courses are: interarterial, intramural, intramyocardial (inclusive of intraseptal), prepulmonic, subpulmonic, and retroaortic [6]. Of these, the interarterial and intramural variants are considered the highest risk for symptoms and risk of SCD, especially when accompanied by other high-risk features such as slit-like ostia, stenotic ostia, or acute-angled takeoff. Interarterial AAOLCA has a prevalence of ~0.03%, compared to ~0.23% for interarterial AAORCA [7]. The remaining subtypes are less common, with a prevalence estimate of 0.07% for subpulmonic, 0.28%

for retroaortic, and 0.04% for prepulmonic [7].

The diagnosis and management of AAOCA remains a challenging yet crucial congenital heart defect that can challenge the most experienced pediatric cardiologist on the best treatment approach. Recognizing this it is essential that primary cardiologist partners with their primary care counterparts to care for these patients. The aim of this document is not to provide a systematic review of all the literature related to AAOCA, nor to provide medical or surgical recommendations, but to act as a high-yield tool for primary pediatric clinicians who play a pivotal role in ensuring timely diagnosis and appropriate care, ultimately safeguarding the health and well-being of affected children.

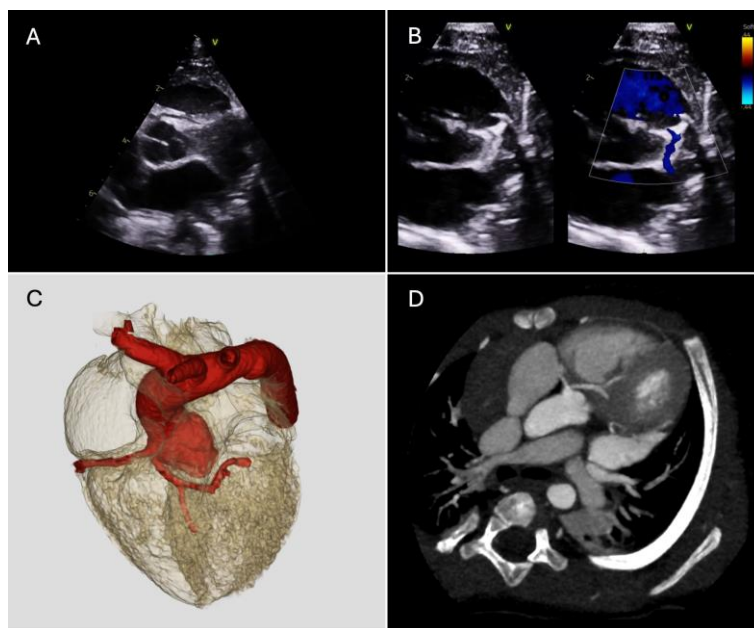


Figure 2. Anomalous aortic origin of the left coronary artery (AAOLCA). This 6-month-old female underwent evaluation to rule out vascular ring due to suggestion of a posterior esophageal indentation on an upper gastrointestinal study. Echocardiogram raised concern for a single coronary artery origin from the right sinus of Valsalva (A), with Doppler flow in the left coronary artery continuing past the left sinus of Valsalva (B). CCT angiography confirmed AAOLCA (C) and additionally demonstrated an intramyocardial course of the left coronary artery (D) [Own work].

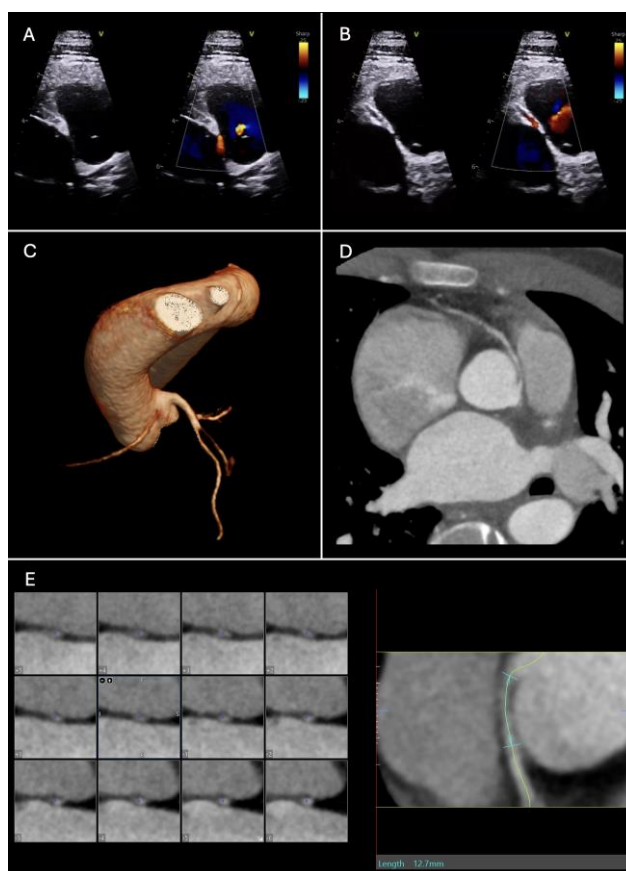


Figure 3. Anomalous aortic origin of the right coronary artery (AAORCA). This 16-year-old male was referred for evaluation of heart murmur, found to have innocent murmur on exam but left ventricular hypertrophy by voltages on electrocardiogram. Echocardiogram raised concern for AAORCA, with true origin seen by color Doppler (A) and false origin suggested adjacent to the right sinus of Valsalva (B). CCT angiography confirmed AAORCA (C, D) with a 12.7 mm intramural course (E). [Own work].

2. Clinical Presentation

Patients with AAOCA have a wide variation in clinical presentation. Most patients are asymptomatic, and the diagnosis is made as an incidental finding during an evaluation for a non-contributing complaint like a benign murmur or as a part of an evaluation for family history of cardiac disease [8]. Symptoms may also occur both at rest (24-31% of patients) or with exertion (36-38% patients) [9]. Symptoms can include exercise-induced syncope, dizziness, chest pain, or shortness of breath [8, 9]. Although extremely rare, some patients may present with aborted sudden cardiac death or have a fatal outcome as their initial presentation with diagnosis made on autopsy [10]. Clinical presentation is associated with the anatomic features of AAOCA including right vs left, ostial narrowing, course etc. that have also been described as “high-risk features” [8].

3. Diagnostic Testing

3.1. Electrocardiography (ECG)

It is important to understand that the resting ECG in an asymptomatic patient with anomalous aortic origin of coronary artery (AAOCA) is typically normal [9, 11]. In the rare instance that an ECG is obtained when the patient presents with exertional chest pain or syncope, it is conceivable that ST-T changes corresponding to the distribution of the anomalous coronary artery may be seen.

3.2. Imaging

3.2.1. Echocardiography

Transthoracic echocardiography usually establishes the diagnosis of AAOCA and delineates the origin and proximal course of the coronary arteries. The artery can arise above the sino-tubular junction or from the contralateral sinus, either via a separate or single ostium. A pre-pulmonary, retro-aortic or intra-septal course is thought to be benign. An intramural and/or interatrial course is considered higher risk. Transesophageal echocardiography is typically not performed for diagnosis but is important for intra-operative monitoring during surgical repair. It is important to note that one multi-institution study found poor correlation between institutional echocardiogram reports of AAOCA and an imaging core laboratory, as well as with surgical findings, and highlighted the need for a best practice protocol [12]. As such, the diagnosis may be missed in cardiology-resource poor areas or smaller centers where adequate visualization of coronary artery origins by 2-dimensional echocardiography and color Doppler is not part of the standard echocardiography protocol.

3.2.2. Cardiac Computed Tomography (CCT) and Cardiac Magnetic Resonance Imaging (MRI)

In the current era, cardiac CT (CCT) is routinely performed in individuals suspected to have AAOCA to confirm the diagnosis, determine high-risk characteristics, and aid in surgical planning. The advantages of CCT are that it is usually a quick non-invasive test with excellent spatial resolution that can provide the most accurate assessment of risk modifiers and surrounding structures amongst the available imaging modalities [13]. High-risk characteristics for sudden cardiac death (SCD) on CCT include a slit-like orifice, the presence and length of an intraarterial/intramural segment, an acute angle of take-off, and a high ostial location [11, 14, 15]. The limitations of this test include the administration of a contrast agent and radiation exposure; however, newer scanners have been successful in minimizing these exposures. The inability of the CCT to capture dynamic changes in ostial anatomy and hemodynamics during or shortly after peak exercise, which is when most SCD occurs, is a significant limitation [4]. In addition, for a coronary CCT, some centers also control heart rate with a beta blocker and may use nitroglycerin as a vasodilator in certain cases for better visualization of the coronary artery origins and course. Although there are no standard guidelines for determining the timing and frequency of testing, a CCT is usually performed to confirm the diagnosis when the child is older (typically 9 years or older) and able to follow breath-holding instructions unless there is concern for symptoms/signs of ischemia at a younger age, in which case sedation may be required. If there is an intervention, a follow-up CCT is often performed 3 months post-operatively with additional surveillance imaging considered 1 year post-operatively and then every 5 years.

By comparison to CCT, magnetic resonance angiography (MRA) provides coronary artery and functional imaging without radiation or iodinated contrast agents, but incurs lower spatial resolution, increased scan times, and higher cost [7, 16]. Over the past decade stress cardiac magnetic resonance imaging (CMR) has been increasingly utilized to evaluate for inducible myocardial ischemia in those with AAOCA. It has been demonstrated as safe and feasible, with some studies demonstrating a higher sensitivity when compared to nuclear stress perfusion imaging in the pediatric population [16]. Stress CMR can be performed using pharmacological agents such as dobutamine or adenosine and is recommended to be used as an adjunct to exercise stress testing (EST). [9] Alternatively, nuclear stress perfusion imaging with single photon emission computed tomography (SPECT) can also be performed; however this again involves additional radiation exposure with a higher degree of false positives [17].

3.3. Exercise Stress Testing

Exercise stress testing (EST) plays a central role in the

evaluation of children with anomalous aortic origin of a coronary artery (AAOCA), aiding in risk stratification and surgical decision-making. Exercise testing, particularly when combined with echocardiography and stress CMR, can be adjunctive for detection of inducible ischemia, arrhythmias, or abnormal hemodynamic responses. However, it's important to note that a normal EST does not exclude the risk. Data suggest limited sensitivity of ECG stress testing alone and improved sensitivity/specificity with additional use of cardiopulmonary exercise testing (CPET) [9]. Interpretation requires pediatric-specific expertise and individualized consideration of anatomy and symptoms.

Stress echocardiography remains a safe, accessible, and radiation-free option, although extensive pediatric studies are lacking [18]. Outcomes data are mixed; most published pediatric AAOCA stress echocardiograms, including those postoperatively, have been negative even in symptomatic patients [1]. Thus, adjunctive imaging is essential for comprehensive risk stratification.

3.4. Cardiac Catheterization

With advances in imaging modalities, cardiac catheterization is rarely required for AAOCA diagnosis. However, cardiac catheterization may be helpful in risk stratification when other testing is equivocal. Utilizing medications to mimic physiologic stress, such as dobutamine, can identify changes in coronary caliber when myocardial demand increases. Similarly, fractional flow reserve and instantaneous free-wave ratio testing can confirm the presence of coronary compression and flow impairment in hemodynamically significant lesions [2, 19].

4. Indications for Surgical Repair

In the past decade, there have been important guidelines published for which individuals with AAOCA require surgical repair [1, 14]. There are certain types of AAOCA in which surgery is indicated based on the known increased risk of SCD, such as AAOLCA with an inter-arterial course, or any type of AAOCA with ischemic chest pain, evidence of ventricular arrhythmias, or inducible ischemia on cardiac testing (Table 1). However, there is still ongoing controversy on whether surgery is recommended for asymptomatic children with AAORCA who have no evidence of inducible ischemia. Additionally, it is unclear if this same cohort of

children will benefit from surgery if they have high risk anatomic features on cardiac imaging.

Surgical decision-making involves a shared decision-making approach and utilizes risk-stratification protocols [20, 21] that include clinical presentation, anatomic features, findings on provocative testing, patient activity (leaning more towards surgery in competitive athletes) and patient/family preference. There is variation between centers, but most consider the presence of high-risk anatomic features as a relative indication to proceed with surgery, while the asymptomatic individual with AAORCA with no inducible ischemia, and low-risk anatomic features is followed conservatively (Table 1). This is at least partly because the mechanism of sudden cardiac death in AAOCA patients is not definitively known, and so it is difficult to know which findings convey a higher risk of SCD. There is also limited data on the long-term outcomes of these AAOCA subtypes both with and without surgical intervention. Fortunately, there are ongoing studies that are attempting to shed light on these topics, and so it is possible that the recommendations for surgical intervention in these AAOCA subtypes will change in the future as we learn more.

5. Surgical Repair

Children who meet criteria for AAOCA surgery typically have the repair performed after about nine years of age (unless they present with SCA or ischemic symptoms at an earlier age, which would warrant repair at that time), before they engage in competitive sports and vigorous exercise that may elicit ischemic symptoms or provoke sudden cardiac arrest/sudden cardiac death.

Surgical repair of any AAOCA typically involves unroofing of the intramural segment or reimplantation to the proper sinus (see Figure 3, complex lesions like an intraconal LMCA or LAD from the RCA require repair that is outside the scope of this discussion). Some centers argue that simple unroofing is never adequate while others favor a tailored approach [22]. The type of surgical repair varies for specific patient anatomy and data on outcomes based on repair strategies is not available at this time. Regardless of the surgical technique employed, the goals of repair are to eliminate any high-risk anatomic features (slit-like orifice, inter-arterial course, acute angulation from the aorta) and to place the coronary ostium into the correct anatomic sinus of Valsalva.

Table 1. Current Indications for Surgical Repair in Individuals with Anomalous Aortic Origin of a Coronary Artery.

Presentation	Is Surgery Indicated?
Individuals with anomalous aortic origin of the <i>left coronary artery</i> from the right sinus of Valsalva with an inter-arterial course.	Indicated
Individuals with anomalous aortic origin of <i>any coronary artery</i> and any one of the following:	Indicated

Presentation	Is Surgery Indicated?
1) Symptoms of ischemia 2) Suspected ventricular arrhythmia 3) Evidence of inducible cardiac ischemia on cardiac testing 4) History of sudden cardiac arrest or aborted sudden cardiac death Individuals with anomalous aortic origin of the <i>right coronary artery</i> from the left sinus of Valsalva with an inter-arterial course who:	
1) Are asymptomatic	May be Indicated
2) No evidence of inducible ischemia or arrhythmias	
3) Have anatomic high-risk features on cardiac imaging.	
Individuals with anomalous aortic origin of the right coronary artery from the left sinus of Valsalva with an inter-arterial course without above listed features	Likely Not Indicated

6. Follow Up

6.1. Follow-Up of Post-op Patients

Life-long follow-up with cardiology is indicated, and should include preventive care (healthy lifestyle, heart-healthy diet, lipid screening and management) [20, 21]. Patients are restricted from competitive and recreational athletics until at least 3 months post-op. At the post-operative follow up within the first few months (no current consensus recommendations available and variable based on institu-

tional practices, at 1-6 months post-op), an ECG and echocardiogram are performed, and most centers will obtain exercise stress testing, CCT and a CMR before clearing patients to return to sports (see Figure 3 for pre- and post-operative CCT scans after coronary unroofing). If those studies are reassuring, patients are typically cleared for competitive sports at that time. The current recommendations suggest long term follow-up with cardiology every 1-2 years with an ECG and echocardiogram. A Holter monitor is performed if there are clinical concerns. Functional testing (with EST or CMR) is typically performed every 5 years or so and may be performed sooner if there are clinical concerns.

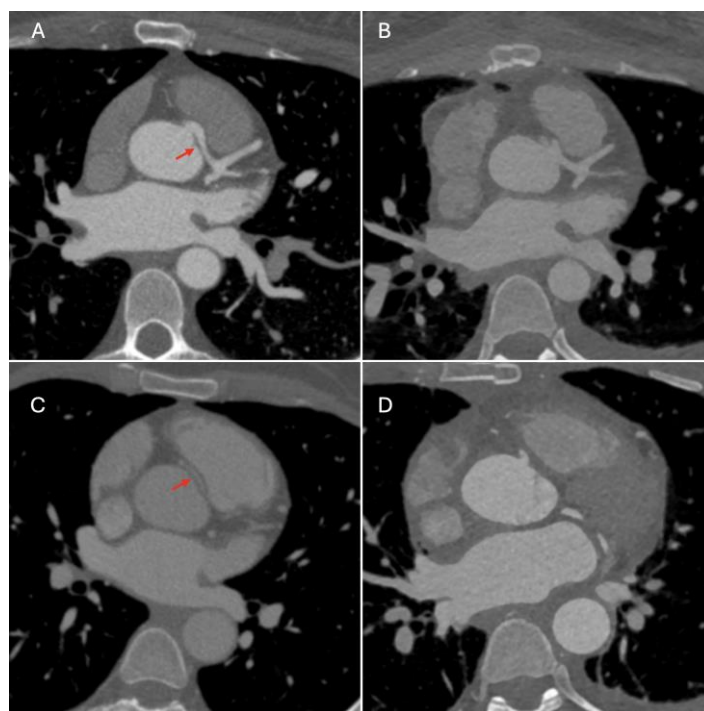


Figure 4. Coronary artery unroofing for anomalous aortic origins of the coronary arteries. The intramural segment is “unroofed” by cutting from the coronary’s true ostium to where it exits the wall of the aortic sinus, as can be seen by comparing the preoperative (A, C) and post-operative (B, D) CCT angiograms in patients with anomalous left (A, B) and right (C, D) coronary arteries.

6.2. Follow Up of Non-operated Patients

Identifying the asymptomatic individual with AAORCA at risk for ischemia remains a challenge [20]. With shared decision-making, many of these patients who do not have evidence of “high-risk” features on CT angiography may be permitted to participate in competitive sports. Families of patients with “malignant” forms of AAOCA may opt not to go for surgical intervention or the patient may be too young for surgery (e.g. diagnosis made in infancy for a murmur). These individuals should be restricted from competitive athletics but encouraged to participate in sports that require low activity levels. Sports counseling should include a discussion regarding awareness of having an automated external defibrillator available during training and the importance of staying well hydrated and resting as needed.

Summary of Follow-up:

Expert consensus guidelines exist that can guide long-term follow-up of these patients [1]. Both the post-op and non-operative patients should be followed annually by a pediatric cardiologist to ascertain a history of exertional symptoms and have an ECG. An echocardiogram should be performed every 1-2 years, while an EST or cardiac MRI is recommended every 3-5 years (starting after 6-8 years of age), depending on age, anatomy and participation in competitive sports. A Holter monitor is recommended only with symptoms. Follow-up encounters should review sports counseling as previously discussed.

7. Screening for Family Members

Although AAOCA is predominantly sporadic, familial cases have been documented with asymptomatic siblings being diagnosed using echocardiographic screening after a patient was found to have AAOCA [23, 24]. While definitive genetic mechanisms remain elusive, previously identified genes implicated in embryonic angiogenesis and development of coronary artery disease studies propose a polygenic contribution to anomalous coronary development [25]. Given the reported familial cases and familial clustering and the high stakes of sudden cardiac arrest/sudden cardiac death in these patients, some centers recommend screening of all first-degree family members of patients diagnosed with AAOCA, although this recommendation is not universal.

8. Quality of Life Implications

AAOCA is associated with decreased quality of life (QOL) as reported by both patients and caregivers, regardless of the patient’s risk classification or surgical status [26]. Patients frequently experience emotional distress—including sadness, anger, isolation, and a sense of being “different” when restricted from participating in sports [20], which may increase their risk for mental health disorders. Pediatricians can be

effective advocates for their patients by remaining vigilant, screening for these concerns, and referring for mental health services when appropriate.

9. Future Directions

There is ongoing research on optimizing imaging, cardiac catheterization techniques and surgical techniques for patients with AAOCA [22, 27]. Computational modeling is being used to understand the mechanisms of myocardial ischemia and sudden cardiac arrest in AAOCA [28, 29]. These models range from simplified 0D models predicting flow based on imaging data to high-fidelity 3D models that simulate blood flow, vessel wall deformation, and post-surgical changes. Advanced fluid-structure interaction models have shown promise in accurately predicting physiological parameters like cardiac catheterization-derived fractional flow reserve (FFR) and instantaneous wave free ratio (iFR) during rest and stress [30]. However, several challenges remain, including accurately modeling exercise-related ischemia, reconstructing detailed coronary anatomy from imaging, and incorporating interactions with aortic valve leaflets. There is also lack of long-term data on outcomes in these patients, including rates of re-stenosis, recurrent ischemia, need for re-operation or late complications following surgical repair. Future work aims to address these gaps through development of a registry that would along long term follow up, advancements in imaging resolution, development of physiologically accurate control models for exercise, and incorporation of patient-specific valve geometries into simulations [31].

10. Conclusions

AAOCA is associated with an increased risk of SCA, and this risk may be reduced through surgical repair. The primary goal is to identify those patients who are at genuine risk for sudden cardiac events. Multi-modality testing that integrates advanced cardiac MRI stress perfusion scans with anatomic features on CCT-scans to detect functional ischemia along with review of clinical symptoms help guide critical treatment decisions. Larger registries are needed to gather long term data and inform guidelines. Surgical decision-making should be individualized and guided by a shared decision-making model, incorporating standardized protocols to risk-stratify patients. Lifelong cardiology follow-up is essential for both surgically treated and non-operated individuals, and proactive preventive care is critical to ensure optimal long-term outcomes. Given the psychological impact of the condition and activity restrictions, pediatricians should routinely screen for mental health concerns and refer to appropriate mental health services when indicated.

Abbreviations

AAOCA	Anomalous Aortic Origin of a Coronary Artery
AAOLCA	Anomalous Aortic Origin of the Left Coronary Artery
SCA	Sudden Cardiac Arrest
ECG	Electrocardiography / Electrocardiogram
CT	Computed Tomography
MRI	Magnetic Resonance Imaging
AAORCA	Anomalous Aortic Origin of the Right Coronary Artery
SCD	Sudden Cardiac Death
GI	Gastrointestinal
CCT	Cardiac Computed Tomography
MRA	Magnetic Resonance Angiography
CMR	Cardiac Magnetic Resonance Imaging
SPECT	Single Photon Emission Computed Tomography
EST	Exercise Stress Testing
CPET	Cardiopulmonary Exercise Testing
QOL	Quality of Life
FFR	Fractional Flow Reserve
iFR	Instantaneous Wave-Free Ratio

Conflicts of Interest

The authors declare no conflicts of interest.

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