

Surgical unroofing of intramural anomalous aortic origin of a coronary artery in pediatric patients: Single-center perspective

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ABSTRACT

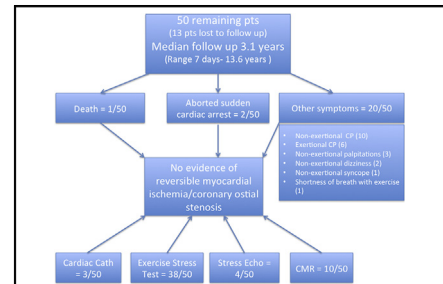
Background: Intramural anomalous aortic origin of a coronary artery (AAOCA) is associated with an increased risk of sudden cardiac death. This is amenable to surgical coronary unroofing, but outcomes studies are lacking.

Objective: To perform a comprehensive review of our institutional experience with pediatric patients with AAOCA who underwent surgical repair with unroofing of the intramural segment, focusing on preoperative and postoperative course and testing as well as intraoperative findings.

Methods: A retrospective cohort study was conducted to evaluate patients with AAOCA status post coronary unroofing at Children's Hospital of Wisconsin. Data extraction included symptoms, preoperative and postoperative imaging and testing, surgical findings, and postoperative clinical course.

Results: From January 1999 to December 12, 2015, 63 patients underwent unroofing at a median age of 13 years (0.5-18 years). The majority underwent unroofing of an intramural right coronary (79%); 21% had an intramural left AAOCA. Symptoms suggestive of possible ischemia were present in about 50%. Additional structural cardiac anomalies were present in 33%. Transthoracic echocardiography was diagnostic in 60 of 63 (95%) and correlated with surgical findings in all cases. There was no surgical mortality associated with the unroofing, and no additional coronary reinterventions were performed. The median duration of postoperative follow-up was 3.1 years (7 days to 13.6 years). Symptoms either persisted or developed in 46% postoperatively. Postoperative exercise stress testing, stress echocardiography, and cardiac magnetic resonance imaging were performed in 76%, 8%, and 20%, respectively, of the cohort. None identified findings consistent with reversible coronary ischemia. Three patients had sudden cardiac arrest (1 death) after surgery without an identified residual coronary abnormality.

Conclusions: Transthoracic echocardiography, with carefully designed coronary imaging protocols, can be diagnostic in accurately identifying intramural AAOCA in pediatric patients. Unroofing can be performed safely with no early morbidity, but symptoms can persist (including rare life-threatening events) without evidence of ischemia by postoperative provocative testing. (J Thorac Cardiovasc Surg 2017; ■:1-9)



Outcomes after unroofing of intramural anomalous aortic origin of a coronary artery (AAOCA). Shown are the postoperative outcomes in patients with AAOCA, status post unroofing surgery in terms of symptoms and investigation findings.

Central Message

Coronary unroofing can be performed safely, with minimal early morbidity, but symptoms can persist without evidence of ischemia by postoperative provocative testing.

Perspective

This study questions that unroofing is a curative procedure that relieves the risk of coronary ischemia in patients with intramural anomalous aortic origin of a coronary artery. This study highlights that, although surgery can be safely performed when the preoperative risk of myocardial ischemia is frequently unknown, risk stratification for subsequent events after unroofing is difficult to assess in the absence of standardized protocols.

See Editorial Commentary page XXX.

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Isolated anomalous aortic origin of a coronary artery (AAOCA) is a rare congenital heart defect (~0.2% of the population), but it is the second-leading cause of sudden



Scanning this QR code will take you to a supplemental video for the article.

Abbreviations and Acronyms

AAOCA	= anomalous aortic origin of a coronary artery
AAOLCA	= anomalous aortic origin of the left coronary artery
AAORCA	= anomalous aortic origin of the right coronary artery
AICD	= automatic implantable cardioverter-defibrillator
AR	= aortic regurgitation
CMR	= cardiac magnetic resonance imaging
CT	= computed tomography
ECG	= electrocardiogram
LV	= left ventricular
PVC	= premature ventricular contraction
SCA	= sudden cardiac arrest
TTE	= transthoracic echocardiogram

cardiac death in young athletes, after hypertrophic cardiomyopathy.¹ This anomaly appears to be of greater risk when the anomalous coronary courses intramural within the aortic wall before exiting the aorta from the contralateral sinus.² Children and adolescents can present with symptoms of myocardial ischemia, including exertional chest pain, syncope, or cardiac arrest.²⁻¹¹ Once identified, there is an ongoing controversy regarding exercise restriction, as the risk of acute myocardial ischemia is difficult to predict in the individual patient.¹²⁻¹⁴ Over the past 20 years, a surgical technique to address this pathology has been developed that involves unroofing of the intramural segment of the anomalous coronary artery (Video 1).^{7,15-21} Studies have shown improvement in symptoms postoperatively at experienced centers^{18,22-29}; however, there have been recent reports of late mortality after the unroofing procedure.³⁰ There is an ongoing controversy regarding exercise limitation and frequency and nature of follow-up studies in this population after surgery. Single-center studies have found conflicting results in assessing myocardial perfusion after coronary unroofing using stress echocardiography, exercise treadmill testing, and nuclear imaging, with discrepant findings between tests in the same patient.²³⁻²⁶

In this review, we provide a comprehensive description of our institutional experience in pediatric patients with intramural AAOCA who underwent surgical unroofing of the intramural segment, focusing on their preoperative, intraoperative, and postoperative course and testing.

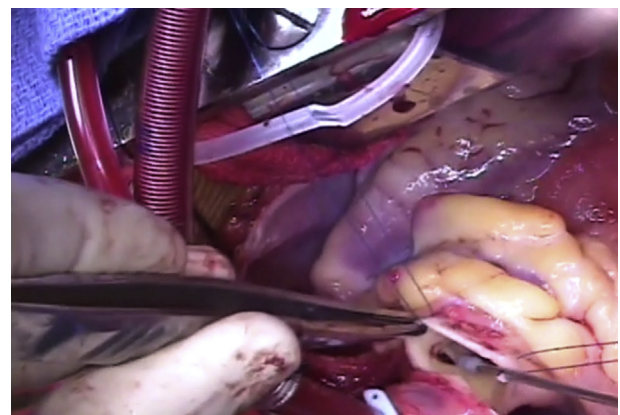
METHODS

This was a retrospective cohort study of patients who underwent coronary unroofing surgery for an intramural AAOCA at Children's

Hospital of Wisconsin. This was approved by the Institutional Review Board (date and number of IRB approval: 8/25/2015, 782223-5). We identified all patients for this study by searching our institutional database from January 1999 (when coronary artery unroofing was first performed here) to December 2015. Medical record data were reviewed for demographic data, symptoms, preoperative investigations, surgical details, postoperative clinical course, and postoperative testing for the cohort. We reviewed the preoperative imaging to describe the anomalous coronary origin and course of the coronary artery as well as to identify other structural anomalies and assess ventricular function. All postoperative imaging was reviewed for coronary anatomy and course of the anomalous coronary, coronary flow patterns, ventricular wall motion abnormalities, and perfusion defects at rest and under pharmacologic stress if performed.

RESULTS

A total of 63 patients were identified who met the inclusion criteria. The patient characteristics are summarized in Table 1. The median age at diagnosis was 12.2 years (range, 1 day to 18 years). The median age at the time of surgery was 13 years (range, 0.5-18 years). Of the patients, 68% were male. The majority of our cohort (79%) had anomalous aortic origin of the right coronary artery (AAORCA), and 21% had an anomalous aortic origin of the left coronary artery (AAOLCA). Symptoms were present on presentation in 54% of the patients, with 44% having either an aborted sudden cardiac arrest (SCA) or exertional symptoms. The most common presenting symptom in both the groups was exertional syncope. There were 4 patients presenting after an aborted SCA event, of whom 3 had AAOLCA. Three of the patients (all with AAOLCA) presenting with an aborted SCA event had documented ventricular fibrillation during the event, and 1 patient had pulseless electrical activity; an additional



VIDEO 1. Unroofing procedure. This intraoperative video shows the unroofing procedure performed for intramural anomalous aortic origin of the right coronary artery from the left sinus with interspersed images detailing the procedure. The unroofing extended beyond the wall of the aorta, which required placement of sutures to reestablish continuity of the aortic wall to coronary artery. Video available at: <http://www.jtcvsonline.org>.

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