## Nees et al

## Congenital

## Patients with anomalous aortic origin of the coronary artery remain at risk after surgical repair

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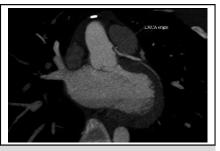
## ABSTRACT

**Objectives:** Anomalous aortic origin of a coronary artery (AAOCA) from the opposite sinus of Valsalva is a rare cardiac anomaly associated with sudden cardiac death (SCD). Single-center studies describe surgical repair as safe, although medium- and long-term effects on symptoms and risk of SCD remain unknown. We sought to describe outcomes of surgical repair of AAOCA.

**Methods:** We reviewed institutional records for patients who underwent AAOCA repair, from 2001 to 2016, at 2 affiliated institutions. Patients with associated heart disease were excluded.

**Results:** In total, 60 patients underwent AAOCA repair. Half of the patients (n = 30) had an anomalous left coronary artery arising from the right sinus of Valsalva and half had an anomalous right. Median age at surgery was 15.4 years (interquartile range, 11.9-17.9 years; range, 4 months to 68 years). The most common presenting symptoms were chest pain (n = 38; 63%) and shortness of breath (n = 17; 28%); aborted SCD was the presenting symptom in 4 patients (7%). Follow-up data were available for 54 patients (90%) over a median of 1.6 years. Of 53 patients with symptoms at presentation, 34 (64%) had complete resolution postoperatively. Postoperative mild or greater aortic insufficiency was present in 8 patients (17%) and moderate supravalvar aortic stenosis in 1 (2%). One patient required aortic valve replacement for aortic insufficiency. Two patients required reoperation for coronary stenosis at 3 months and 6 years postoperatively.

**Conclusions:** Surgical repair of AAOCA is generally safe and adverse events are rare. Restenosis, and even sudden cardiac events, can occur and long-term surveillance is critical. Multi-institutional collaboration is vital to identify at-risk subpopulations and refine current recommendations for long-term management. (J Thorac Cardiovasc Surg 2018; ■:1-11)



Cardiac CT angiogram in a patient with aborted sudden cardiac death 6 years after unroofing. Coronal view shows the takeoff of the anomalous left coronary artery.

#### Central Message

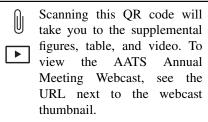
Repair of AAOCA can be performed safely; restenosis, and even sudden cardiac death can occur and long-term surveillance is critical.

### Perspective

Repair of AAOCA can be accomplished safely and can lead to improvement in symptoms and signs of ischemia. Restenosis, and even sudden cardiac events, can occur and long-term surveillance is critical. Multi-institutional collaboration is vital to identify at-risk subpopulations and refine current recommendations for longterm management.

See Editorial Commentary page XXX.

Anomalous aortic origin of a coronary artery (AAOCA) arising from the opposite sinus of Valsalva is a rare cardiac anomaly but is the second most common cause of sudden cardiac death (SCD) in young competitive athletes.<sup>1-5</sup> The incidence of AAOCA is reported to be



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## **ARTICLE IN PRESS**

Abbreviations and Acronyms	
	= anomalous aortic origin of a coronary
	artery
Abnl	= abnormal
AI	= aortic insufficiency
ALCA	
	from the right sinus of Valsalva
ARCA	= anomalous right coronary artery arising
	from the left sinus of Valsalva
AS	= aortic stenosis
aSCD	= aborted sudden cardiac death
AVR	= aortic valve replacement
СТ	= computed tomography
CUMC	= Columbia University Medical Center
ECG	= electrocardiogram
Echo	= echocardiogram
ECMO	= extracorporeal membrane oxygenation
EF	= ejection fraction
ICU	= intensive care unit
IQR	= interquartile range
LAD	= left anterior descending
LCx	= left circumflex artery
LV	= left ventricular
MPI	= myocardial perfusion imaging
MR	= magnetic resonance
MRI	= magnetic resonance imaging
NA	= not applicable
Nl	= normal
PDA	= patent ductus arteriosus
PPS	= postpericardiotomy syndrome
RCA	= right coronary artery
RV	= right ventricular
SCD	= sudden cardiac death
TR	= tricuspid regurgitation
WCMC	= Weill Cornell Medical Center

METHODS

### **Study Population**

We performed a retrospective cohort study of patients who underwent initial surgical repair of AAOCA from 2001 to 2016 at NewYork-Presbyterian/Columbia University Medical Center (CUMC) and New York-Presbyterian/Weill Cornell Medical Center (WCMC). Patients were identified through our institutional surgical database. Attending cardiothoracic surgeons at NewYork-Presbyterian operate at both campuses and report to the Society of Thoracic Surgeons database as a single center. All patients with isolated AAOCA who received surgery by one of our congenital heart surgeons were included, regardless of age. Patients with associated congenital heart defects were excluded.

## Perioperative Management and Long-Term Follow-up

The historical practice at CUMC and WCMC has been to refer all ALCA patients, regardless of symptoms, as well as ARCA patients who show symptoms and/or objective findings of ischemia for surgical repair. The decision to repair asymptomatic ARCA patients has been debated and has remained at the discretion of each cardiologist and cardiothoracic surgeon, in consultation with the patient and family. Repair is typically performed by unroofing of the septum between the aortic lumen and intramural portion of the coronary. In a small subset of patients with short intramural segments, coronary translocation was performed at the discretion of the surgeon. If the intramural portion of the coronary was at or below the level of the aortic commissure, takedown of the tip of the commissure was performed. In patients who required takedown of the aortic commissure, resuspension was attempted in all cases. During the study period, no standardized protocol existed for routine pre- or postoperative followup or testing, and testing was obtained at the discretion of the referring cardiologist and operating surgeon.

## **Data Collection**

We collected baseline data on patient demographic characteristics, indications for surgery, and type of repair. We examined perioperative and postoperative outcomes. Perioperative outcomes of interest included mechanical circulatory support, arrhythmias, infections, pneumothoraces, and pericardial effusions. Postoperative outcomes of interest included symptoms of ischemia including aborted SCD (aSCD), abnormal electrocardiogram (ECG), echocardiogram (echo), or exercise stress test, new aortic insufficiency (AI), or aortic stenosis (AS), arrhythmia, and reoperation. The ECG, echo, and exercise stress test results were obtained from the most recent available visit.

Most data were obtained via retrospective chart review. For patients who received surgery at CUMC, all patients were also contacted via phone. Verbal consent was obtained to use patients'/parents' subjective reporting of symptoms and vital status. For patients followed by a cardiologist at an outside institution, written consent was obtained to contact the primary cardiologist for follow-up records. Data on patients treated at WCMC were obtained via retrospective chart review with waiver of informed consent. Longitudinal follow-up data had recently been obtained and recorded in the medical records as part of an internal quality improvement effort. In patients for whom no longitudinal follow-up was available, the most recent vital status was obtained using the Social Security death index.

## **Statistical Methods**

Clinical and demographic variables were described using standard summary statistics. Change in pre- and postoperative symptoms and test data were described graphically. Standard univariable analyses were used to compare pre- and postoperative characteristics including  $\chi^2$ , Fisher exact, unpaired *t* test, and Kruskall–Wallis test, as appropriate. Analyses were performed using Stata software, version 13.1 (StataCorp, College Station,

between 0.1% and 0.7%, with an anomalous right coronary artery arising from the left sinus of Valsalva (ARCA) reported more commonly than an anomalous left coronary artery arising from the right sinus of Valsalva (ALCA).<sup>6-8</sup>

The goal of surgical AAOCA repair is to eliminate the risk of SCD. Single-center studies have shown that surgical repair is generally safe with short-term improvement in symptoms of ischemia after repair.<sup>9-15</sup> Although a few studies have shown persistent symptoms and/or signs of new ischemia postoperatively,<sup>11,15,16</sup> data on medium- and long-term outcomes for these patients are limited, and the residual risk of SCD remains unknown. The purpose of this analysis was to describe outcomes after surgical repair of isolated AAOCA at 2 cardiac centers where the same surgical team operates.

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