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ORIGINAL ARTICLE

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Surgical treatment of anomalous left main coronary artery with an intraconal course

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Abstract

Objective: Anomalous left main coronary artery (LMCA) with an intraconal course is a relatively rare form of anomalous aortic origin of a coronary artery (AAOCA) from the wrong sinus of Valsalva. There is currently a paucity of information regarding this entity. The purpose of this article is to review our surgical experience with repair of anomalous LMCA with an intraconal course.

Methods: This was a retrospective review of 12 patients with an anomalous LMCA and an intraconal course who underwent surgical repair. The median age at surgery was 15 years (range 2-47). The seven oldest patients all had preoperative symptoms of exertional chest pain and one also had exertional syncope. The five youngest patients had no preoperative symptoms. One of these patients had a hemodynamically significant ventricular septal defect and one patient was the sibling of a patient who had undergone repair of AAOCA.

Results: The 12 patients underwent surgical repair including unroofing of the myocardial bridge overlying the intraconal LMCA and a LeCompte procedure. There was no early or late mortality and there were no significant complications. All 12 patients have resumed normal, unrestricted activities.

Conclusions: Twelve patients with an anomalous LMCA and intraconal course presented to our institution for treatment. Surgical repair was performed successfully in all 12, with resolution of symptoms in the 7 patients who were symptomatic preoperatively. These results suggest that the surgical treatment is safe and efficacious in patients with an anomalous LMCA and intraconal course.

KEYWORDS
congenital heart disease, congenital heart surgery, coronary artery anomaly

1 | INTRODUCTION

Anomalous aortic origin of a coronary artery (AAOCA) from the wrong sinus of Valsalva is a relatively uncommon form of congenital heart defect that has been associated with symptoms of myocardial ischemia and sudden death. There are multiple anatomic forms of AAOCA, including anomalous right vs left coronaries, intramural vs nonintramural, and single coronary vs two-coronary systems.[1-3] While there is a substantial amount of information available regarding the more common forms of AAOCA, there is still a paucity of information regarding many of the less common forms.

Anomalous left main coronary artery (LMCA) with an intraconal course is an example of a relatively rare form of AAOCA. In our experience, patients with an anomalous LMCA and intraconal course always have the same anatomy. Specifically, there is a single

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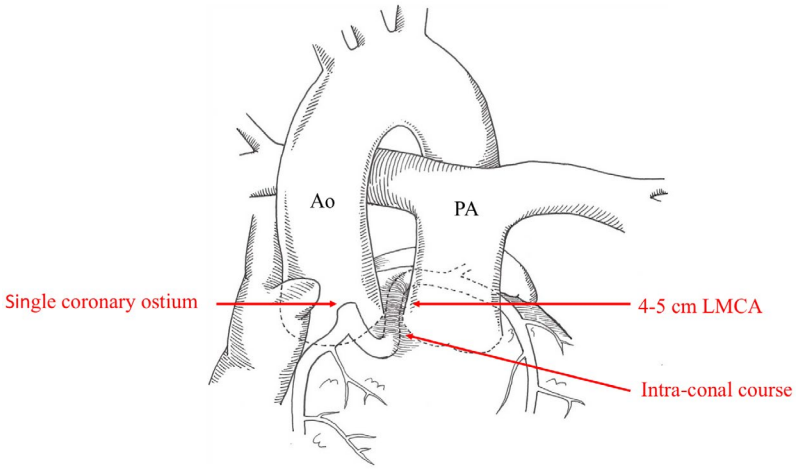

FIGURE 1 Artist's illustration demonstrating the anatomy of anomalous LMCA with an intra conal course. There is a common ostium from which the right and LMCA originate. The LMCA is approximately 4-5-cm long as it traverses between the aorta and pulmonary artery and there is a 3-4-cm-long myocardial bridge as the LMCA passes through the conus (Used with permission from Mainwaring et al[2])

coronary ostium from which both the right and left coronary arteries originate (Figure 1). By definition, the LMCA is approximately 4-5 cm in length as it passes between the aorta and pulmonary artery to reach a normal position behind the pulmonary artery (Figures 2 and 3). However, these images do not fully convey all of the anatomic features. The cartoon in Figure 4 demonstrates a cross-section of the aorta as seen from below. As depicted, there is a "bud" or "trunk" from which both coronaries originate, and this results in 2-3 mm of separation between the aortic wall and the coronary arteries. As a consequence, the origins of both coronary arteries are well away from the aorta and therefore are never intramural. In addition, because at end of the bud there is a smooth carina, both of the coronary origins have wide-open ostia and do not have any acute angulation at their take-off. The LMCA then drops down away from the aorta and passes through conal muscle before exiting in the normal position behind the pulmonary artery. This intraconal course also results in the LMCA being at the very bottom of the "V" created by the aorta and pulmonary artery. These anatomic features are unique compared to any other form of AAOCA.

Perhaps one of the biggest surprises that came out of the 4th Coronary Artery Anomaly Symposium held in Houston, TX, on 7 and 8 December 2018 was the change in sentiment regarding the treatment of an anomalous LMCA with an intraconal course. Prior to this conference, there was substantial skepticism regarding the importance of this entity, as most practioners in the congenital heart community believed that this was a benign form of AAOCA that never required surgical intervention. There is no better evidence of this preexisting sentiment than the title that was sent to the first author of this article, which was "Surgical Approach: Is it an Option?" However, by the end of the conference, when the audience was polled regarding the management of a patient with an intraconal LMCA, a history of exertional chest pain, and documentable evidence of myocardial ischemia, 94% of the audience recommended surgical treatment.

Given the dramatic change in sentiment that occurred at the symposium regarding management of intraconal LMCAs, it is imperative to summarize the data and discussions that lead to this conclusion. The purpose of this manuscript is to review the existing literature on the subject, summarize our institutional experience with the clinical presentation, surgical treatment, and surgical outcomes in patients with an anomalous LMCA and an intraconal course, and provide a synopsis of the relevant discussions that took place at the 4th Coronary Artery Anomaly Symposium.

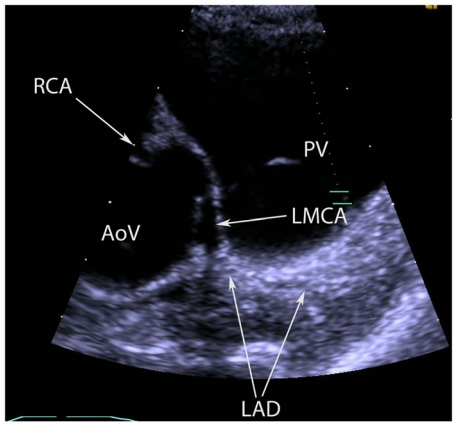

FIGURE 2 Echocardiogram demonstrating typical anatomy of an anomalous LMCA with an intraconal course

2 | MATERIALS AND METHODS

This was a retrospective review of 12 patients who underwent surgical repair of anomalous LMCA with an intraconal course at our institution from 2005 to 2018. There were 10 males and 2 females, and the median age at surgery was 15 years (range 2-47 years). The mode of presentation for these 12 patients is shown in Table 1.

The seven oldest patients all presented with symptoms of exertional chest pain. These patients (age 14-47 years) all had a solid history for chest pain during exercise. One of these seven patients had also experienced exertional syncope.

The five youngest patients were all asymptomatic. One of these patients (2 years old) had a hemodynamically significant ventricular septal defect that required surgical repair. The preoperative workup of this patient identified the presence of an anomalous LMCA. It was elected to repair the intraconal LMCA during repair of the

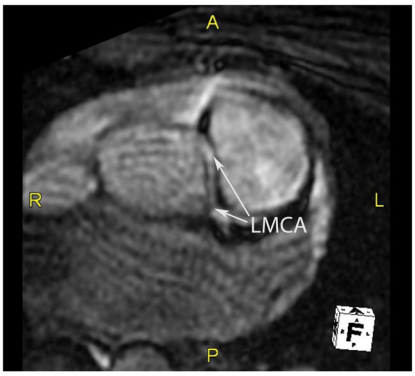

FIGURE 3 Computer tomographic angiogram demonstrating the typical anatomy of an anomalous LMCA passing between the aorta and pulmonary artery

TABLE 1 Patient characteristics

Age	Gender	Presentation
47	M	Exertional CP
30	F	Exertional CP
19	M	Exertional CP
18	M	Exertional CP
15	F	Exertional CP
15	M	Exertional CP
14	M	Exertional CP, syncope
7	M	Asymptomatic
5	M	Asymptomatic
5	M	Sibling with AAOCA
3	M	Asymptomatic
2	M	Ventricular Septal Defect, AAOCA detected

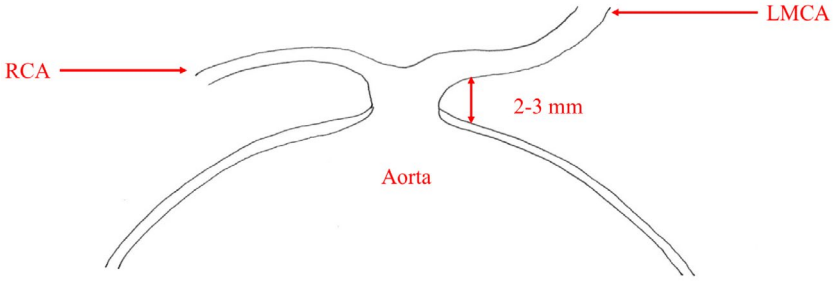

FIGURE 4 Cartoon demonstrating a cross-sectional view of the aorta from below. The single coronary ostium forms a "bud" or "trunk" from which both coronaries originate. There is a 2-3-mm distance between the aortic wall and origin of the coronaries, precluding the possibility of an intramural course. In addition, the smooth bifurcation means that the ostia will be wide open and the origins will have no acute angulation

ventricular septal defect. An additional patient was identified with anomalous LMCA during screening after his sibling underwent surgical repair of an AAOCA. The parents of this child elected to have the second child undergo surgical repair.

The three remaining children who were asymptomatic and underwent surgical repair all presented in the early part of the series (2005-2008). At that time, AAOCA was considered an extremely rare form of congenital heart defect. The imaging that was available was far less sophisticated than is currently available, and the identification of an AAOCA was considered sufficient information. Although today the imaging can readily discern the differences between intramural and nonintramural and can identify the hallmark findings of an intraconal LMCA, the imaging that was available in the mid-2000s was frequently not able to distinguish these findings. Furthermore, the understanding of AAOCA in that era was not what it is today, and the mandate to identify these anatomic patterns did not exist. The three young patients in this series were referred for surgery due to the presence of an anomalous left coronary artery, and it was only in the operating room that the anatomy was fully elucidated. Given what we currently know, these three patients today would not meet the criteria for surgical intervention at our institution.

The surgical technique that we have employed is illustrated in Figure 5. These procedures are performed through a median sternotomy. The basic anatomy is dissected out prior to instituting cardiopulmonary bypass including full mobilization of the branch pulmonary arteries to the hilum. Once bypass is implemented, the right pulmonary artery is divided to facilitate exposure of the intraconal course of the LMCA (Figure 5A). The aorta is then cross-clamped in order to achieve a motionless field. The initial phase of this dissection is quite similar to that used in harvesting the pulmonary root for a Ross procedure, as there is a plane where the muscle fibers change orientation and this is the natural plane through which the intraconal coronary courses. Sharp dissection is utilized to divide the muscle fibers overlying the LMCA (Figure 5B). This dissection is continued until the entire length of the intraconal course is unroofed (Figure 5C). The aorta is then be unclamped and the area of dissection inspected for bleeding. The right pulmonary artery is brought anterior to the aorta (LeCompte), and the posterior aspect reanastomosed to the main pulmonary artery. A homograft patch is used to augment the anterior surface (Figure 5D).

3 | RESULTS

The 12 patients underwent surgical repair of anomalous LMCA with an intraconal course without any significant complications. The

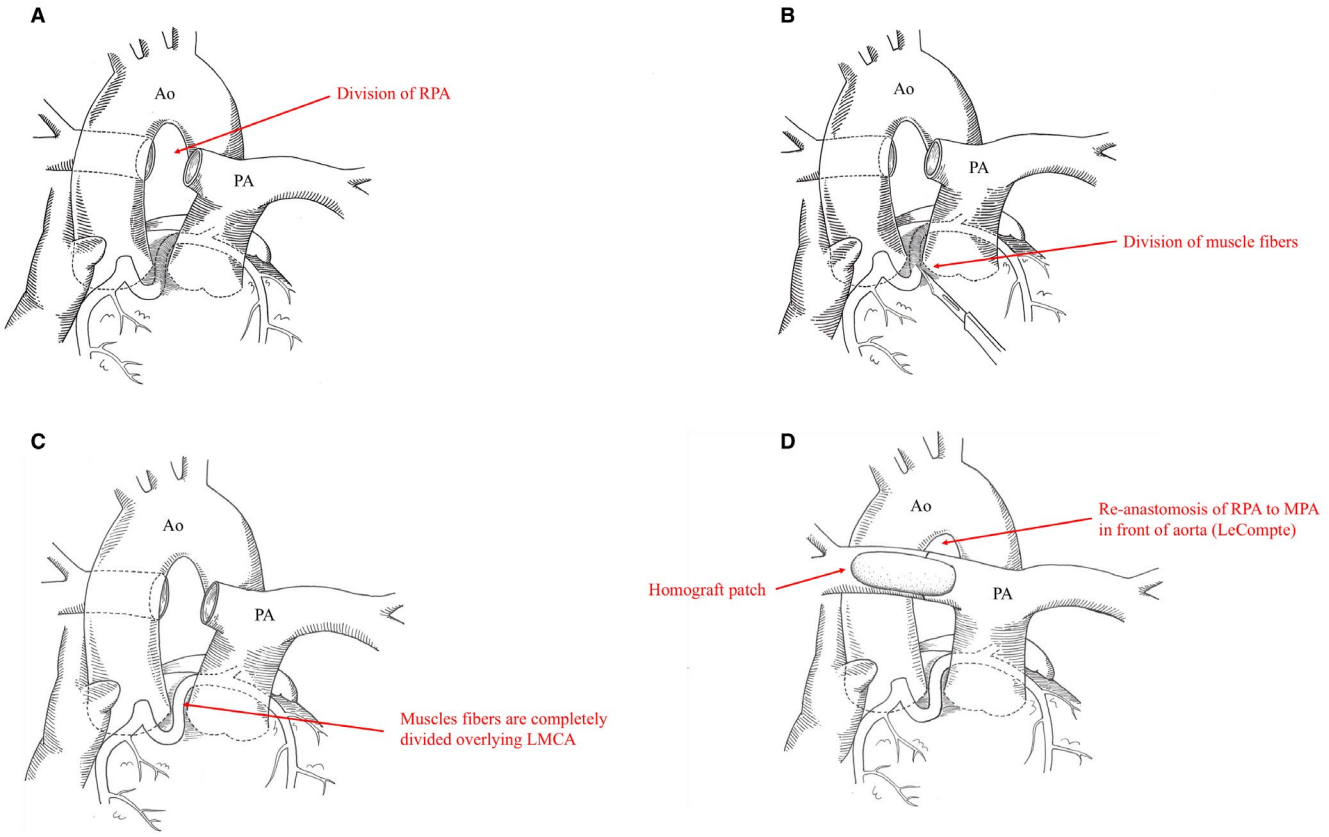

FIGURE 5 A, Artist's illustration demonstrating surgical division of the right pulmonary artery. This is performed first in order to provide access to the intraconal LMCA. (Used with permission from Mainwaring et al[2]). B, Artist's illustration demonstrating initial steps in dividing the muscle fibers overlying the LMCA. The aorta is cross-clamped during this part of the procedure in order to achieve a motionless field and facilitate precise division of the conal muscle fibers. C, Artist's illustration demonstrating completion of the division of the muscle fibers overlying the LMCA. D, Artist's illustration demonstrating reanastomosis of the right pulmonary artery to the main pulmonary artery in front of the aorta (LeCompte). It is usually our practice to have a tissue-to-tissue connection posteriorly and place a homograft patch anteriorly to reduce the possibility of any tension of this anastomosis (Used with permission from Mainwaring et al[2])

median length of stay was 5 days (range 3-8 days). There has been no early or late mortality.

The patients have been followed for a median of 5 years (range 1-11 years). All patients have returned to normal, unrestricted activities. The seven patients who presented with symptoms of exertional chest pain have not had a recurrence of symptoms, and no patient has had any other cardiovascular symptoms.

4 | DISCUSSION

This study was performed to evaluate the surgical results in patients undergoing repair of anomalous LMCA with an intraconal course. In this series of 12 patients, there were no significant complications and no mortality. All of the patients have resumed normal activities including the seven who had symptoms of exertional chest pain. Thus, the surgical treatment described in this article appears to be both safe and efficacious.

One of the interesting observations from our data is that the symptomatic patients were all age 14 years or greater and there was a greater preponderance of males in this cohort than is typically seen in AAOCA. It is our speculation that these two observations are both related to the unique anatomy of this entity. As previously described, the anatomy of an anomalous LMCA with an intraconal course is never going to be intramural and will always have a wide-open ostium without any angulation. Thus, the mechanisms that have been attributed for creating myocardial ischemia for most forms of AAOCA are not applicable in this situation. The two remaining possibilities for creating myocardial ischemia would be the long myocardial bridge or the coronary coursing at the very bottom of the "V" between the aorta and pulmonary artery and being pinched between these structures. It would seem plausible that age and gender predispose to changes in wall tension and compliance and thus account for relationship with these two factors.

In a recent study by Martens et al, the group at LA Children's Hospital described their surgical experience with 10 patients who presented with an anomalous LMCA with an intraconal course.[4] All 10 of their patients had symptoms of myocardial ischemia, including one patient who had experienced a ventricular fibrillation arrest. The

median age at surgery was 13 years. The surgical approach that this group has developed is similar in principle to our approach, although it differs in some of the finer details. Their approach includes harvesting the pulmonary root (as one would do for a Ross procedure), and this results in dividing the majority of the muscle fibers overlying the LMCA. Any remaining muscle fibers are divided at this juncture. The pulmonary root is then reimplanted approximately 1 cm more proximally in the right ventricular outflow tract, thus moving it away from the LMCA. The distal anastomosis is facilitated by fully mobilizing the branch pulmonary arteries and dividing the ligamentum arteriosum. This pulmonary root "translocation" requires slightly more dissection than the approach that we utilize, but has the potential advantage of making it very straightforward to divide the muscle fibers around the back side of the main pulmonary artery. All 10 of these patients underwent successful surgical repair and have returned to unrestricted activities.

Brothers et al published a manuscript summarizing an experience with 13 patients who had an anomalous LMCA and intraconal course in which they described the work-up of this entity.[5] MRI was effective in all 13 patients in identifying wide-open ostia and absence of any angulation in the origin of the coronary arteries. These observations are identical to those that we have made regarding the anatomy of anomalous LMCA. However, despite making the same observations, their group came to a diametrically opposite conclusion than we did, as they stated that these findings were … "the basis for understanding the benign clinical course and showing that surgery is unnecessary for this coronary anomaly."

There are several recent case reports that shed some additional light on the clinical presentation of anomalous LMCA with an intraconal course. One such case report summarized an experience with an 11-year-old male who was asymptomatic but had an echocardiogram as part of a sports screening program.[6] This study revealed the presence of an anomalous LMCA. Based on their understanding of the literature on this topic, they believed that this was a benign entity. However, a stress perfusion study revealed the presence of an anteroseptal region of reversible ischemia. The patient underwent surgical repair, had an uneventful recovery, and 6 months later had complete normalization of the stress perfusion study.

Another recently published case report is that of a 19-year-old male who was playing basketball and fainted onto the floor.[7] He awoke several minutes later and was taken to a local hospital where he proceeded to evolve a myocardial infarction (Troponin leak and EKG changes). This patient underwent surgical repair of his anomalous LMCA and had an uneventful recovery.

A third case report summarizes an experience with a 16-year-old female who presented with neurocardiogenic syncope (including one that required resuscitation), the patient underwent a work-up that revealed the presence of a single coronary artery giving rise to an intraseptal LMCA.[8] This was surgically treated with resolution of her syncope and a return to athletic activities.

There are at least three case reports in older patients with an anomalous LMCA originating from a single coronary ostium who presented with either unstable angina or ventricular tachycardia. The two patients with unstable angina were a 51-year-old male[9] and a 62-year-old female.[10] Both of these patients had angiographic evidence of clean coronary arteries. The patient who developed recurrent episodes of ventricular tachycardia was 42-years old, otherwise had normal coronary arteries, and had complete resolution of the tachycardia following coronary bypass grafting.[11]

At the 4th Coronary Artery Anomaly Symposium in Houston, there were seven patients who were presented who had an anomalous LMCA with an intraconal course and had documented evidence of myocardial ischemia. Five of these patients were presented by Dr Silvana Molossi from Texas Children's Hospital.[12] These five patients were all being treated medically at the time of the conference. However, given the systematic approach that this group has undertaken to evaluate and manage patients with AAOCA, this presentation had a major impact in convincing most of the participants that this entity had real clinical importance. There were two additional case reports presented of patient with an anomalous LMCA with an intraconal course and documented evidence of myocardial ischemia. These two patients added weight to the premise that this entity is being seen by many groups around the country. Following these presentations, the audience was polled regarding their opinion of the treatment of an intraconal LMCA with documented ischemia, and 94% responded that they would now recommend surgery.

Based on our own experience at Stanford, a review of the literature, and a summary of the Houston conference, there are now 29 patients with an anomalous LMCA and intraconal course who have presented with symptoms of exertional chest pain or syncope. The majority of these cases also had documented evidence of myocardial ischemia. This collection of cases provides the best evidence that an intraconal course is not *always* a benign entity.

Several large catheterization series provide some data on the overall prevalence of coronary artery anomalies.[13-15] These studies suggest a prevalence of anomalous left coronary arising from the right sinus of Valsalva ranging between 0.017% and 0.15%. These data generally did not distinguish between single vs dual coronary ostia and did not delineate those cases with a single ostium and an intraconal course. Anomalous LMCA originating from a single coronary ostium with an intraconal course would represent a fraction of all anomalous left coronaries (we would estimate perhaps a quarter), and so this would result in a prevalence of approximately 0.004%-0.04%.

Dr Jane Newburger presented an analysis of the autopsy data on anomalous LMCA with an intraconal course at the 4th Coronary Artery Anomaly symposium.[16] This analysis revealed not a single case of an anomalous LMCA with an intraconal course attributed as the cause of death. From this data, one must conclude that the anatomy and physiology of an anomalous LMCA with an intraconal course may cause enough ischemia in some patients to result in exertional chest pain or syncope but somehow does not predispose to sudden death. This difference clearly makes it a distinct entity from the standard AAOCA population.[17,18]

Our overall surgical series for AAOCA at Stanford currently numbers 152 cases. This includes 108 males (67%) vs 44 females (33%).

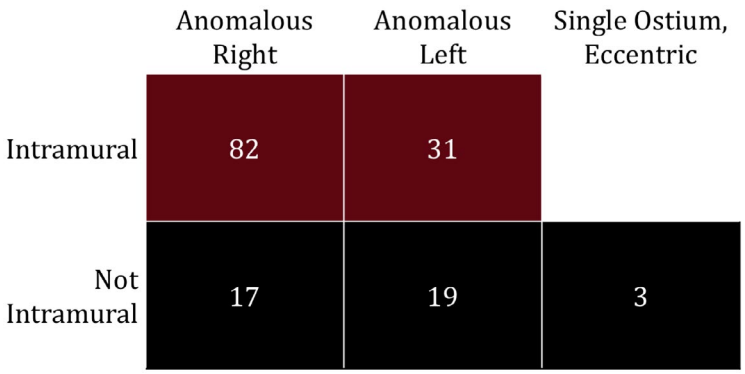

FIGURE 6 2 × 2 box demonstrating the prevalence of anatomy for AAOCA based on right vs left and intramural vs nonintramural

Seventy-eight patients (51%) had symptoms of exertional chest pain, syncope, or were survivors of sudden death. Twenty-seven patients (18%) had various forms of congenital heart defects. The anatomy of these surgical cases can be described by a 2 × 2 box, as shown in Figure 6. By far and away, the most common type of AAOCA was an intramural right, accounting for 52% of all cases. The 12 cases of anomalous LMCA originating from a single coronary ostium with an intraconal course would account for 8% the patients in this series.

The 152 patients have undergone surgical repair without any early or late mortality. The median length of stay was 5 days. Following recovery from surgery, the patients are allowed to resume unrestricted activities. These patients have been followed for an average of 5 years (760 patient years). Three patients have required a reoperation for recurrent symptoms, all of which had an anatomic basis. These data fulfill Koch's postulate that symptomatic patients have resolution of their symptoms following surgery (98.5%) and patients with recurrent symptoms (1.5%) have a residual anatomic problem.

Mery et al have recently reported their experience with 44 patients who underwent surgical intervention for AAOCA over a 4½-year time period.[19] These 44 patients were part of a large, prospective cohort study in which all of the patients had extensive and comprehensive testing. The majority of the patients who had surgery were male, had anomalous right coronaries (80%), and were intramural (80%). There has been no early or late mortality, and 90% of the patients have returned to normal activities. These results are representative of what can be accomplished at larger centers that see a significant volume of patients with AAOCA.

It should be noted that the anatomy of a single coronary ostium originating from the left sinus of Valsalva is substantively different from its counterpart from the right. In our experience, single coronary ostium from the left will also have a bud or trunk from which the LMCA and right coronary arise. This means that an anomalous right from a single coronary ostium will rarely be intramural. However, the course of the anomalous right coronary usually wraps around the lowermost aorta and is well away from the bottom of the "V." As a consequence, an anomalous right coronary artery originating from a single coronary ostium is never intraconal. These patients typically are asymptomatic, and it is our conjecture that this probably represents a relatively low-risk subset of patients with AAOCA. Ultimately, it is going to be important to evaluate each of the many subsets of AAOCA to determine the natural history risk of each separate entity.[20]

In conclusion, the 4th Coronary Artery Anomaly symposium held in Houston revealed some important and exciting new information regarding anomalous LMCA with an intraconal course. At the end of the conference, there was a strong consensus that patients with this anatomy can develop symptoms of exertional chest pain or syncope in association with documentable evidence of myocardial ischemia. Thus, the view that this is a benign entity is no longer tenable. Another observation that came out of this conference was that these patients develop symptoms of chest pain or syncope but seem to be at low risk for sudden death. Finally, there was a strong consensus that patients who have an intraconal LMCA and evidence of myocardial ischemia should undergo surgical repair. Hopefully, these viewpoints can be disseminated and eventually incorporated into the guidelines for management.

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Figure 5B and C were drawn by Erin Anne Mainwaring.
Figures 1, 5B and C were also drawn by Erin. These figures were previously published in Mainwaring et al.[2]

CONFLICT OF INTEREST

The authors declared that they have no conflicts of interest with the contents of this article.

AUTHOR CONTRIBUTIONS

Dr Richard Mainwaring is the first author of this manuscript and presented this article at the 4th Coronary Artery Anomaly symposium in Houston, Texas. Dr Mainwaring was the principle author of the manuscript, and his contributions included data collection, data analysis/interpretation, and drafting the article. Dr Mainwaring has been a congenital heart surgeon for 28 years and has authored numerous articles on the subject of AAOCA.

Dr Frank Hanley is the senior author on this manuscript. Dr Hanley contributed to this manuscript in data analysis/interpretation

and approval or article. Dr Hanley has been a congenital heart surgeon for 31 years and was the surgeon for the majority of the patients in this study.

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