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Sudden Cardiac Death in a Young Athlete: Imaging Techniques to Evaluate the Etiology

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A B S T R A C T

Anomalous aortic origin of the coronary artery (AAOCA) is a rare form of congenital heart disease with varying implications. Although most of the population with this congenital heart defect may remain completely asymptomatic and never know of the diagnosis, the risk of sudden cardiac death among healthy children and young adults during or just after exercise is significant. A previously healthy adolescent female athlete presented to the pediatric intensive care unit after sudden cardiac arrest on the playing field. She was successfully resuscitated, and further imaging revealed a diagnosis of AAOCA. She underwent surgical correction of her defect without complications. AAOCA remains a rare and often undetected form of congenital heart disease based on the asymptomatic nature of the defect; however, with proper imaging, a diagnosis can be made to allow for correct treatment.

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Introduction

Anomalous aortic origin of the coronary artery (AAOCA) is defined as a form of congenital heart disease in which both the right and left main coronary arteries arise from the same side of the aorta, or aortic sinus, either with one common origin or two separate origins (Brothers, Gaynor, Jacobs, Poynter, & Jacobs, 2015). The coronary artery that is arising from the incorrect aortic sinus will then typically have an intramural course within the aortic wall and descend to its proper location. The anomalous coronary artery can also take an interarterial course and lie between the pulmonary artery and aorta, or an intraconal course, and descend through the myocardium. Different variations of this congenital heart defect include anomalous aortic origin of the right coronary artery (AAORCA) arising from the left aortic sinus and anomalous aortic origin of the left coronary artery (AAOLCA) arising from the right aortic sinus (Figure 1).

The true prevalence of this congenital heart defect is unknown, simply based on lack of symptomatology among affected patients until they experience sudden cardiac death (SCD); however, studies have shown that approximately 0.1% to 0.7% of the population is affected (Brothers et al., 2015). In addition, studies have shown that the interarterial AAORCA is approximately three to six times more common than interarterial AAOLCA (Brothers et al., 2015). Of note, AAOCA is a completely different anomaly from anomalous left coronary arising from the pulmonary artery (ALCAPA), which is a form of congenital heart disease that typically presents at 3 to 6 months of life. Symptomatology of ALCAPA typically includes congestive heart failure and left ventricular failure.

The risk of SCD in those with AAOCA is greater during or just after exercise when cardiac output and myocardial oxygen demand are at its greatest. Myocardial ischemia because of impaired coronary blood flow through the anomalous coronary artery leads to ventricular arrhythmias and subsequently SCD. Impaired coronary blood flow derives from multiple factors including compression of the anomalous coronary artery itself because of its abnormal course, the typically small ostium of the anomalous coronary artery, and often the acute angle at which it arises from its anomalous origin. Active children and young adults are the most of those who experience SCD (Poynter et al., 2014). In young athletes in the United States who were previously healthy, AAOCA is the second leading cause of SCD because of undiagnosed congenital heart disease (Poynter et al., 2014). The purpose of this article is to present a case report of AAOCA and describe the current recommended imaging for definitive diagnosis to promote optimal management and outcomes.

Case report

A 14-year-old female with no significant medical history presented to the pediatric intensive care unit (PICU) after cardiac arrest while playing field hockey. According to witnesses and her parents,

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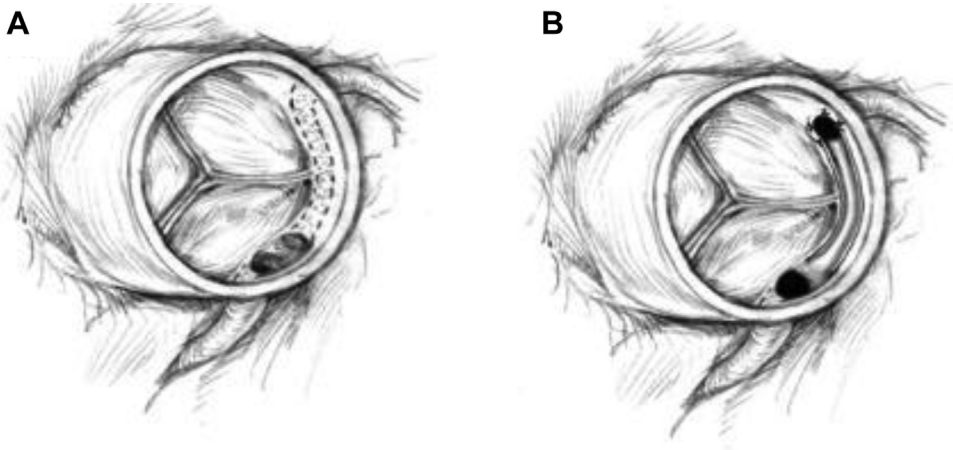

Figure 1. Portrait depicting the (A) anatomy of anomalous aortic origin of the left coronary artery and (B) the unroofing procedure. Reprinted from The Annals of Thoracic Surgery, 92/3, Kaushal, S., Backer, C.L., Popescu, A.R., Walker, B.L., Russell, H.M., Koenig, P.R., et al., Intramural coronary length correlates with symptoms in patients with anomalous aortic origin of the coronary artery, 986-992, Copyright (2011), with permission from Elsevier.

she had been playing field hockey and suddenly requested to leave the game, complaining of chest pain. She then collapsed and was noted to be pulseless, apneic, and with epistaxis. Cardiopulmonary resuscitation was immediately performed, and on arrival of the emergency medical technicians (EMTs) 8 min later, she was diagnosed with torsades de pointes, a form of ventricular tachycardia. She was successfully converted to a normal sinus rhythm with defibrillation and administration of intravenous lidocaine. She was transported to a hospital emergency department where she was intubated because of respiratory distress and subsequently transferred to the PICU.

Initial diagnostic testing for the patient on admission included chest radiograph (Figure 2), the electrocardiogram (ECG) to evaluate her cardiac rhythm, and the echocardiogram. Chest radiograph showed diffuse bilateral lung opacities consistent with pulmonary edema. The initial ECG was consistent with normal sinus rhythm, and the echocardiogram revealed a structurally normal heart with good function although the coronary artery originations were difficult to visualize. On day 2 of her hospitalization, the patient underwent cardiac magnetic resonance imaging (MRI), both with and without contrast to further evaluate cardiac function and revealed an ejection fraction of 67% (normal, 55–70%), normal myocardial wall motion, and no abnormal myocardial perfusion or delayed myocardial enhancement. Unfortunately, artifact from the patient's orthodontic hardware prevented accurate assessment of the coronary anatomy. In an effort to assess the patient's coronary artery anatomy, a computed tomography angiography (CTA) of the chest was performed on the third hospital day. The study revealed a limited evaluation of the coronary arteries because of tachycardia. Despite the limitations with the CTA, the right coronary artery was found to arise normally from the right coronary cusp and medial origin, or arising from the middle of the aorta as opposed to the left side, of the left main coronary artery with early branching into the left anterior descending and left circumflex artery.

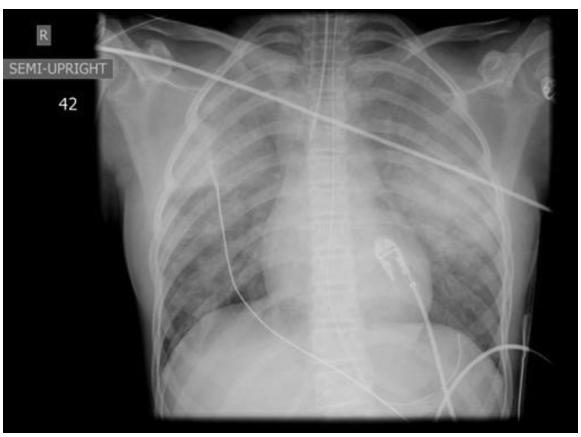

Figure 2. Chest radiograph on admission revealing diffuse bilateral lung opacities consistent with diffuse pulmonary edema.

Given the initial cardiac rhythm noted by the EMTs, the diagnosis of prolonged QT syndrome was hypothesized as the etiology of her aborted SCD; however, initial and subsequent ECGs obtained in the PICU showed a normal sinus rhythm with a normal corrected QT interval. QT interval is a measurement within the heart's electrical cycle that represents electrical depolarization and repolarization of the ventricles. Prolonged QT interval represents an increased risk for dysrhythmia, specifically torsades de pointes. She was scheduled for placement of an automatic implantable cardioverter defibrillator in the event of a repeat SCD. Before this procedure, further diagnostic testing was completed to accurately assess the coronary artery anatomy. These subsequent diagnostic studies included a repeat echocardiogram and CTA. The echocardiogram showed the right coronary artery arising normally from the aorta and the left coronary artery arising from the right coronary cusp with an intramural course. This was further confirmed with a gated CTA, which showed AAOLCA arising from the right aortic cusp with an approximate 6 mm intramural course and narrowing of the vessel to approximately 1 mm (Figure 3). Based on these findings, the patient underwent unroofing of the intramural portion of her AAOLCA without complications. This procedure removes the aortic wall tissue overlying the coronary artery allowing the coronary artery to fully expand and provide myocardial oxygenation during times of high demand (Kaushal et al., 2011). There was no reimplantation of the anomalous coronary artery. Before discharge to home on postoperative day 5, the patient underwent a follow-up echocardiogram, which showed prograde (forward) blood flow through the AAOLCA and normal biventricular function.

At subsequent pediatric cardiology follow-up appointments, the patient had repeat imaging including CTA and nuclear medicine myocardial perfusion single-photon emission computed tomography (SPECT). The CTA showed patent coronary arteries with the origin of the left main measuring approximately 3 mm by 2 mm in diameter. The exercise myocardial perfusion SPECT

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