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Right coronary artery anomalous origin from left coronary sinus as a cause of sudden cardiac arrest in athletes

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Abstract

Coronary anomalies constitute an entity that occurs in rates below 1% of the general population, but which could potentially lead to sudden cardiac arrest as the first symptom, especially, in young people and athletes. We have developed this issue presenting the case of an athlete who suffered a sudden cardiac arrest during a race and explaining how we managed the case.

Keywords

Sudden death in athletes; Coronary vessel anomalies; Case report.

Introduction

We present the case of a 32-year-old male who suffered a sudden cardiac arrest during a half-marathon race. He was reanimated in situ, presenting complete recovery; and, in the diagnostic study, a coronary anomaly was discovered as the cause of the event.

Case Report

This was a 32-year-old male, regular runner, due to the cardiac arrest of ventricular fibrillation of uknown etiology without a known medical and familiar personal history and who had never suffered any syncope, chest pain or palpitation. Running in the Behobia-San Sebastian Race —a popular half-marathon race in Donostia-San Sebastian - he presented a witnessed cardiorespiratory arrest at the 20Km. Initial defibrillable rhythm - ventricular fibrillation - was observed and with just one defibrillation he recovered spontaneous breathing and circulation. After orotracheal intubation due to decreased level of consciousness, he was transferred to Donostia University Hospital. A 12-lead electrocardiogram displayed a sinus rhythm with a previously an unknown right bundle branch block and QT interval was normal. An urgent coronary angiography was performed (Figures 1 & 2), showing anomalous origin of the right coronary artery, very

hypoplastic, from the left coronary sinus. Within few hours, the patient was extubated and showed complete neurological recovery. During the study, a coronary computed tomography was performed, which verified the anomalous origin of the right coronary artery from the left coronary sinus and specifying an interarterial course between the aorta and the right ventricular outflow tract —not being able to rule out an intramural path - (Figures 3,4 & 5). An echocardiography and magnetic resonance imaging were performed, excluding other cardiac arrest causes - cardiomyopathy, dysplasia, or necrotic scare.

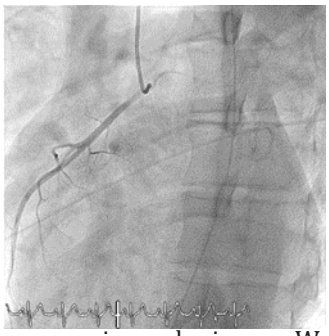

Figure 1: Coronary angiography image. We can see an hypoplastic right coronary artery, showing a clear left dominance.

Figure 2: This also is an image of a coronary angiography but the one that corresponds to the left coronary trunk. If we compare with figure 1, we clearly can see that artery size is much bigger. 1) Descendent anterior artery. 2) Circumflex coronary artery.

Figure 3: Anomalous origin of the right coronary artery in cardiac computed tomography. We can identify the right coronary artery origin in left coronary sinus simultaneously with left coronary trunk. The size difference between both arteries is very striking. We can see how the circumflex artery takes the right coronary arteries irrigation area coming underneath the aorta in the image. 1) Aorta. 2) Left coronary sinus. 3) Right coronary sinus. 4) Non coronary sinus. 5) Left coronary trunk. 6) Right coronary artery. 7) Circumflex coronary artery. 8) Continuation of circumflex coronary artery. 9) Descendent anterior artery.

The diagnosis of an anomalous origin of a hypoplastic right coronary artery (RCA) from the left coronary sinus with an interarterial course between the aorta and the right ventricle exit tract was made and, consequently, an assessment for surgical treatment was carried out. Evaluated by the Cardiac Surgical Team, the manipulation of the right coronary artery was rejected due to its hypoplastic nature and the clear left dominance. Surgical options were discarded and an implantable cardioverter defibrillator (ICD) was implanted as the most appropriate treatment. In the 18 months of follow-up, there were not clinical nor electrical events.

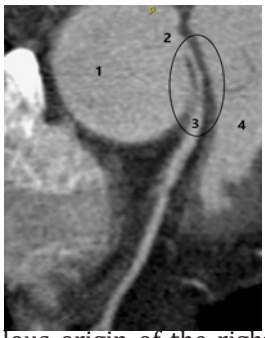

Figure 4: Anomalous origin of the right coronary artery with interarterial course in cardiac computed tomography. The right coronary artery origin is in the left coronary sinus and there we can also observe its interarterial course between aorta and pulmonary artery. 1) Aorta. 2) left coronary sinus. 3) interarterial course of right coronary artery. 4) pulmonary artery.

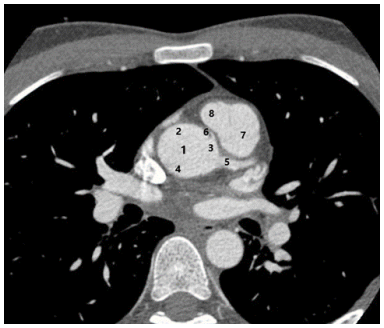

Figure 5: Anomalous origin of the right coronary artery in the left coronary sinus and its interarterial course in cardiac computed tomography. 1) Aorta. 2) Right coronary sinus. 3) Left coronary sinus. 4) Left coronary trunk. 5) Origin of right coronary artery in left coronary sinus. 6) Right ventricle. 7) Pulmonary artery.

Discussion

We present the case of a young athlete with no relevant personal medical history who suffered a sudden cardiac arrest during intense physical exercise at the end of a half-marathon. The patient had no neurological sequelae, including memory, language, and other superior functions; and he was diagnosed with anomalous origin of a hypoplastic RCA with an interarterial course. There was no option for surgical revascularization due to anatomical reasons and, therefore, ICD implantation was decided upon.

Among general population, the estimated incidence of coronary anomalies is below 1% [1], although critical literature shows ranges between 0.1 and 8.4% [2]. They are mainly diagnosed during coronary angiographies performed due to ischemic heart disease or in autopsies, which poses further difficulty when attempting a more accurate estimation of the real incidence. Among coronary anomalies, 90% are origin anomalies and their distribution rates vary across literature. Nonetheless, it is generally considered that two thirds are comprised of left anomalies - anterior descending artery and circumflex artery—, whereas RCA constitutes the other remaining third. The clinical expression of the disease is variable, and only a range between 20% to 45% of coronary anomalies cause clinical symptoms such as chest pain, arrhythmias, or cardiorespiratory arrest [2,3].

Sudden cardiac arrest related to coronary anomalies usually occurs in young athletes. According to different studies, among people under 35 years, it is positioned as a cause of cardiac arrest in a range of 7% to 17% [4-7]. In the analysis of two registries - one with 2,304 patients in the UK, and another with 1,435 athletes in the US -, coronary anomalies are positioned as the second cause of the entity, after obstructive hypertrophic cardiomyopathy [4,7].

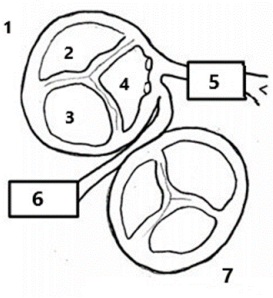

Figure 6: Schematic representation of the interarterial trajectory of a right coronary artery originating in the contralateral sinus. 1) Aorta. 2) Non-coronary sinus. 3) Right coronary sinus. 4) Left coronary sinus. 5) Left coronary trunk. 6) Right coronary artery. 7) Pulmonary artery.

There are several types of coronary anomalies which imply different risk and probability levels to suffer from sudden cardiac arrest. The most important coronary anomalies associated with sudden cardiac arrest are two. On one hand, 70% of the cases are related to an anomalous left coronary artery which originates in the contralateral sinus, and it is cause of cardiac arrest due to the extensive cardiac territory it supplies. On the other hand, the remaining 30% is due to right coronary artery coming from the left coronary sinus, a rate most which is related to an interarterial course, as in our case (Figure 6), in relationship with the high pressure obtained in the interarterial course and the subsequent compression of the RCA [8].

If the aforementioned interarterial course is not present, the risk is much lower. Diagnosis may be difficult in some cases considering most usual cardiovascular tests like 12-lead electrocardiogram (ECG), stress ECG with maximal exercise, and echocardiography may be within normal limits [2]. If the diagnosis of the anomalous origin is made incidentally by performing a coronary angiography, cardiac computed tomography (Cardiac-CT) or an echocardiogram; the study should be completed with an ischemia detection test to quantify the risk. Should the tests be positive or display clinical signs as angina, arrhythmia, or sudden cardiac arrest; an anatomical study must be completed and the treatment decided. Classically, coronary angiography constituted the gold-standard although, however, it is currently controversial once Cardiac-CT made its appearance, alongside the high resolution of echocardiograms and the cardiac magnetic resonance. Currently, several studies show the superiority of the Cardiac-CT with its ability to detect 100% of the lesions, whereas coronary angiography displays detection rates of only 53% [9]. The echocardiogram is postulated as a screening method as it allows the detection of the anomalous origin, though it does not show characterization. Thus, Cardiac-CT or cardiac magnetic resonance are required for a complete study [5,6,8-10].

Options for treatment are different depending upon the risk and age of the athlete. Patients under 35 and in high risk, require surgical or invasive treatment. In those over 35 and performing high-intensity or competitive sports aggressive treatment is also recommended. Should the patient be over 35 and not perform high intensity sports, individualized treatment for each case must be called for [3].

Management is mainly surgical, which show two technical options. The first, which is more physiological but, technically, more difficult; is to perform an 'un-roofing', that is, the affected artery is removed and implanted in the corresponding coronary sinus, which displays low morbidity and mortality in young

patients [11]. The alternative option would consist of the performance of a bypass to avoid secondary ischemia. Such indication is controversial, on grounds of this entity affecting young patients and the duration of the bypass being limited [12]. Should the patient feature great surgical comorbidity, an implantation of a stent in the affected area may constitute a suboptimal option. If it were not an anatomical possibility to treat the artery, such as in the case we present, an ICD ought to be implanted as a secondary prevention strategy to prevent sudden cardiac arrest. In this case, and due to clear left dominance alongside the very high difficulty to surgically manipulate this hypoplastic right coronary artery, the implantation of an ICD was decided upon in a medical-surgical session. Different possibilities were considered, such as subcutaneous ICD, but this possibility was discarded since it fails to meet the appropriate electric detection criteria. Finally, a conventional VVI-ICD was implanted with no complication whatsoever. Intense sport ought to cease until surgical repair is performed. Sport may be resumed once the cause has been treated, the athlete is asymptomatic and in those cases of incidental finding without evidence of ischemia [13]. In our case, and in light of the impossibility for a surgical solution, the recommendation of not doing competitive sport is retained, although moderate physical activity may be performed, which is recommended.

Currently, there is still controversy regarding what the most appropriate strategy for screening tests to be carried out in competition athletes to avoid sudden cardiac arrest may be. There are two main models worldwide: on one hand, the Italian model, which is based upon a basal electrocardiogram and - should there be a positive result or signs of risk - further additional testing. And, on the other hand, what is a more conservative model, i.e. the use of a screening tool - mainly a questionnaire - and the study of only positive cases. In Spain, the Federated Athletes' Council performs a medical examination, including a basal ECG, but not for lay athletes. In the Behobia-San Sebastian race a questionnaire is recommended for runners, but it is not mandatory [6,14,15].

Given the difficulty to perform an appropriate screening on all competition athletes, the allocation of resources is a needed option to treat sudden cardiac arrest when it occurs. Immediate availability of automatic external defibrillators (AED) could be life-saving in those cases. Thus, the supply of AED performed by trained people in sport facilities or competition proves to be mandatory [2].

Conclusion

The anomalous origin of the coronary arteries is usually involved in sudden cardiac death of young athletes and should be analyzed in those cases.

Declarations

- There has been no significant financial support for this work that could have influenced its outcome.

- We know of no conflicts of interest associated with this publication. As Corresponding Author, I confirm that the manuscript has been read and approved for submission by all the named authors.

- All authors contributed to the study conception and design. Compliance with Ethical Standards: none of the authors has any conflict of interest.

- Informed consent was obtained from participant included in the study.

- We declare that this manuscript is original, has not been published before and is not currently being considered for publication elsewhere.

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