CASE REPORT

Cardiac arrest in an 18-year-old man caused by anomalous right coronary artery origin

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SUMMARY

An 18-year-old man presented to hospital following an out of hospital ventricular fibrillation cardiac arrest, with successful resuscitation at the scene. The incident happened during a competitive football match in which the man was playing. In the course of an extensive work up, a CT coronary angiogram demonstrated an anomalous right coronary artery (RCA) origin in the left coronary sinus. The RCA followed an initial intramural and subsequent interarterial course between the ascending aorta and pulmonary artery trunk. Cardiac MRI showed a small subendocardial area of delayed gadolinium enhancement in the inferoseptal wall, consistent with a small infarct. Treatment involved elective coronary artery bypass of the RCA with a right internal mammary artery graft. The patient made a full recovery. An implantable cardioverter-defibrillator was not inserted and he was advised not to return to competitive sports.

BACKGROUND

Cardiac arrest during sporting activity is a rare, but devastating event, and causes major public anxiety. The most recent study commissioned in Ireland to investigate the Epidemiology of Sudden Cardiac Death in the Young 2005–2007, retrospectively reviewed the incidence of sudden cardiac death in the 15–35-year-old population. The national sudden cardiac death (SCD) yearly incidence was found to be approximately 2.68 per 10 000 per year.¹ This is higher compared with other similar European studies, although the heterogeneity of different studies makes direct comparison more challenging. The incidence of sudden cardiac death in the young in England and Wales is reported at approximately 1.8 per 100 000 per year, which roughly corresponds to eight young lives per week.² A variety of congenital and acquired cardiac conditions can lead to an unexpected SCD. Coronary artery anomalies are an important cause of SCD and were the commonest (17%) cause of non-traumatic sudden deaths in a 25-year review of autopsies in US military recruits.³

Either a left coronary artery or right coronary artery (RCA) arising from the opposite coronary sinus is potentially fatal. The clinical importance of a coronary artery anomaly depends on the haemodynamic significance of the vessel/territory supplied and the dominance of either artery. When a coronary artery arises from the opposite sinus it may be forced to follow a number of different courses. These may include: interarterial (between the great vessels), intramural (embedded in the wall of the

aorta/pulmonary artery) and prearterial or retroaortic, among other possibilities. The latter courses may be less ominous compared to intramural and interarterial courses, due to the lower potential for extrinsic compression and occlusive symptoms.⁴ Congenital coronary artery anomalies are not uncommonly associated with sudden death in young athletes, however, they are amenable to surgical correction if identified. The issue of screening of young athletes with basic resting and stress ECG may not be useful based on data from a small retrospective postmortem series,⁵ and thus a thorough investigation of young athletes presenting with exertional symptoms of chest pain or syncope should be undertaken. This case also highlights the importance of the availability of cardiac defibrillators in sports clubs and public places, as well as bystander cardiopulmonary resuscitation (CPR) and basic life support training.

CASE PRESENTATION

An 18-year-old man was playing a competitive Gaelic football match when he suddenly began to feel dizzy and diaphoretic. He was taken off the field after approximately 20 min of play and went to the dressing room, where he collapsed and became unresponsive. A physiotherapist attended him and on feeling no pulse started CPR. A defibrillator present in the sports club was brought to the scene and two shocks were delivered for a rhythm of ventricular fibrillation. On arrival of the ambulance, return of spontaneous circulation had been achieved and the patient regained consciousness en route to the hospital. ECG on arrival in the emergency department confirmed normal sinus rhythm with sinus tachycardia 120 bpm and neither ST nor T wave changes.

The patient's complete history was taken and an examination performed, neither of which revealed any significant intercurrent illnesses; and there was no family history of sudden death. Direct questioning revealed that the patient had experienced one previous episode of dizziness and presyncope, requiring him to sit down, also during a warm-down phase of a match a few weeks previously.

The patient denied smoking and drank alcohol occasionally at social events. He was a very keen sportsman, engaging in high-intensity training with his local football team 3–4 nights per week and with competitive tournament matches at weekends.

INVESTIGATIONS

Following admission, a battery of investigations was performed. These included a full panel of bloods

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including: hs Troponin T, fasting lipids and glucose, toxicology/drug screen, chest X-ray, brain CT, transthoracic echocardiography, an exercise stress test to evaluate for exercise-inducible arrhythmias, and epinephrine and ajmaline provocation tests to uncover Brugada syndrome/other channelopathies. The results of all of these investigations were normal. The patient subsequently went on to have a CT coronary angiogram, which revealed an anomalous origin of his RCA from the left coronary sinus. The RCA arose high up in the left sinus above the pulmonary valve. The origin of the RCA was described as 'slit-like' with an initial intramural course in the wall of the pulmonary artery. Narrowing of the proximal segment of the artery occurred during its interarterial course between the ascending aorta and pulmonary trunk. The RCA was dominant and the remainder of the vessels were normal. Cardiac MRI revealed a focal region of subendocardial late gadolinium enhancement along the inferior wall/inferoseptum from the base to the mid-cavity level, consistent with a small infarct.

DIFFERENTIAL DIAGNOSIS

Differential diagnosis initially included drug misuse, electrolyte disturbances and cardiac causes such as channelopathies, for example, Long QT syndrome and Brugada syndrome, myocarditis, hypertrophic cardiomyopathy and anomalous coronary arteries. In view of the age of the patient, coronary artery disease was unlikely. Unfortunately, in a significant portion of such patients a cause is not found—up to 35% in the study by Eckart *et al.* In this case, CT coronary angiogram confirmed the presence of an anomalous RCA with a small myocardial infarct in the corresponding left ventricular wall territory as the cause of the cardiac arrest.

TREATMENT

The definitive treatment of the anomaly was decided on following detailed case conference/multidisciplinary team discussion involving the cardiologists as well as the cardiothoracic Surgeons. Transfer of the RCA to the correct sinus or 'unroofing' the artery from its intramural course were considered, but carried a high risk of injury to the aorta or pulmonary artery. Thus, the decision was made to perform a single vessel bypass of the RCA with a right internal mammary artery graft. This procedure was well tolerated by the patient and he returned to full physical activity, excluding competitive sport. A full discussion was had regarding the potential to return to competitive sport following the completion of another satisfactory exercise stress test, however, a consensus decision was reached that the area of scar formation in his left ventricle wall, albeit small, did potentially predispose him to further arrhythmias in the setting of maximal aerobic activity. An implantable cardioverter-defibrillator was not implanted in view of the fact that the anomalous artery had now been bypassed and a similar event should not recur in the context of everyday activity.

OUTCOME AND FOLLOW-UP

The patient was seen back in the cardiology outpatient clinic for follow-up approximately 8 weeks after his discharge from hospital. He had made a full recovery with complete return to activity including light jogging. He was again advised as to the importance of avoiding competitive sport/any activity of extreme exertion or rapid acceleration. He will be followed up routinely in the clinic for the foreseeable future.

DISCUSSION

Despite extensive literature review, there appears to be just one documented case of a survivor of aborted sudden death/cardiac arrest due to an anomalous RCA who proceeded to surgical correction. This was reported in the *European Heart Journal* in June 2012, and detailed the case of a 39-year-old man elite long-distance runner who suffered a ventricular fibrillation arrest and had successful resuscitation.⁶ The diagnosis was made using CT coronary angiogram and demonstrated an anomalous RCA arising in the left coronary sinus, following an intramural course between the aorta and pulmonary artery. The patient had surgical translocation of the RCA from the left sinus to the right-sided ascending aortic trunk and recovered well. Myocardial perfusion scintigraphy demonstrated no myocardial ischaemia.

The vast majority of other documented cases of the condition are from necropsy case series. One such case review was published in the *Journal of the American College of Cardiology* in 2000 entitled 'Clinical profile of congenital coronary artery anomalies with origin from the wrong aortic sinus leading to sudden death in young competitive athletes'.³ This was a review of combined registry data (from the USA and Italy) of young athletes who had suffered sudden cardiac death. The study detailed 27 cases of sudden deaths in young athletes, identified solely at autopsy and due to either left main coronary artery from the right aortic sinus (n=23) or RCA from the left sinus (n=4). Each athlete died either during (n=25) or immediately after (n=2) intense exertion on the athletic field. The study concluded that standard testing with ECG (resting or exercise) is unlikely to be reliable as a screening test in large athletic populations. Also, prodromal cardiac symptoms occurred shortly before sudden death in a number of cases, suggesting that a history of exertional syncope or chest pain requires exclusion of this anomaly.

Learning points

- ▶ Anomalous coronary arteries are a significant cause of sudden cardiac arrest/death in young people.
- ▶ Conventional resting and exercise ECG are usually normal. If a patient, particularly a young athlete, presents with syncope or exertional cardiac symptoms, then it should be among the list of differential diagnoses and cardiac CT should be considered.
- ▶ All sports clubs and public meeting places should have cardiac defibrillators as mandatory equipment. Bystander cardiopulmonary resuscitation/basic life support should be taught to all members of sports clubs and associations.
- ▶ While there are data available regarding the incidence of anomalous coronary arteries from angiogram databases, the true risk of sudden cardiac death from these anomalies is not clear because the data are retrospective.
- ▶ As with most conditions, the spectrum of clinical significance ranges from mild to severe/life threatening depending on the location of origin, nature of vessel involved and dominance of the supply system.

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