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A RARE CASE OF ANOMALOUS SINGLE CORONARY ARTERY WITH ABSENT RIGHT CORONARY ARTERY

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ABSTRACT

The presence of a single coronary artery supplying the entire myocardium is an unusual anomaly. Anomalies of coronary arteries may be found incidentally in 0.3-1% of healthy individuals. Most coronary anomalies described in the literature are cases in which the origin of a given coronary artery is anomalous, but very few cases describe the complete agenesis of the ostium of one coronary artery. The congenital absence of a single coronary ostium is a rare finding with an incidence of 0.024%-0.066% in the general population. Most patients are asymptomatic, and prognoses vary. The presence of a single coronary artery may predispose an afflicted patient to angina, myocardial infarction, congestive heart failure or even sudden cardiac death, especially if this anomaly is compounded by atherosclerotic disease.

This case describes a single coronary artery originating from the left sinus of Valsalva with a dominant left circumflex artery continuing distally into the right side of the heart. Right coronary ostium and Right coronary artery was absent.

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INTRODUCTION

The human heart is a remarkably efficient, durable, and reliable pump, distributing more than 6000 litres of blood through the body each day, and beating 30 to 40 million times a year- providing tissues with vital nutrients and facilitating waste excretion. Consequently, cardiac dysfunction can have devastating physiologic consequences. Cardiovascular disease is the number one cause of worldwide mortality, with about 80% of the burden occurring in developing countries.

Disruption of any element of the heart- myocardium, valves, conduction system, and coronary vasculature can adversely affect pumping efficiency, thus leading to morbidity and mortality. Now because of change in lifestyle and habits, many cardiac pathologies are noticed in comparatively younger age group even in a developing country like India and some of these cardiac pathologies are inheritable.

Inside ascending aorta in proximal part the aortic valve has three semi lunar cusps- 2 anterior and 1 posterior with a circumference of 7.5cm (6-7.5cm). Behind the cusp the aortic wall bulges to form aortic sinuses (sinuses of Valsalva). The right and left coronary arteries arise from the upper part of right and left aortic sinuses. Right coronary artery (RCA) supplies right atrium, right ventricle except the area adjoining the anterior inter ventricular groove, a small part of left ventricle adjoining the posterior inter ventricular groove,

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posterior part of the inter ventricular septum, whole of the conduction system of the heart except a part of left branch of AV bundle. The left coronary artery (LCA) divides into the left anterior descending artery (LAD) and left circumflex artery (LCX). It supplies left atrium, left ventricle except the area adjoining the posterior inter ventricular groove, anterior part of the interventricular septum, a part of the left branch of AV bundle. By convention, the coronary artery (either right coronary artery or left circumflex artery) that gives rise to the posterior descending branch and thereby perfuses the posterior third of the septum is called "dominant". In a right dominant circulation, present in approximately 80% of individuals, the circumflex branch of the left coronary artery generally perfuses only the lateral wall of the left ventricle, and the right coronary artery supplies the entire right ventricular free wall and the posterobasal wall of the left ventricle and the posterior third of the ventricular septum. The right and left coronary arteries function as end arteries, although anatomically most hearts have collateral circulation.^{1,2}

Anomalies of coronary arteries may be found incidentally in 0.3-1% of healthy individuals. The presence of a single coronary artery supplying the entire myocardium is an unusual anomaly. Most coronary anomalies described in the literature are cases in which the origin of a given coronary artery is anomalous. Very few cases describe the complete agenesis of the ostium of one coronary artery. The congenital absence of a single coronary ostium is a rare finding with an incidence of 0.024%-0.066% in the general population. Most patients are asymptomatic, and prognoses vary. The presence of a single

coronary artery may predispose an afflicted patient to angina, myocardial infarction, congestive heart failure or even sudden cardiac death, especially if this anomaly is compounded by atherosclerotic disease.⁴

Case report

A 36 years old male committed suicide by hanging, due to personal reasons. Autopsy was conducted at the mortuary of Victoria hospital, Bangalore Medical College and Research Institute, Bangalore. On external examination the dead body measured about 160 cm in length, moderately built and nourished.Rigor mortis was present all over the body. Post mortem staining was seen over the back of the body. Lips and nail beds were bluish discolored. An oblique, incomplete ligature mark measuring 27 cm x 2 cm was present over the front and sides of neck. On internal examination asphyxial features were present in internal organs. Heart was elongated in shape, weighed 250 grams. Right side of heart was underdeveloped and showed slightly more epicardial fat. Right coronary ostium and Right coronary artery was absent. A single coronary artery originating from the left sinus of Valsalva with a dominant left circumflex artery was continuing distally into the right side of the heart. On histopathological examination, both Left main coronary artery and Left anterior descending artery showed Grade 4 to Grade 5 atherosclerosis with about 25% lumen narrowing. There was no previous history of chest pain or breathlessness. There was no family history of any congenital heart disease.



Fig 1 The photograph of anterior surface of heart showing slightly elongated heart with underdeveloped right side of heart having slightly more epicardial



Fig 2 The photograph of posterior view of heart showing elongation of heart



Fig 3 The photograph of heart showing presence of only left coronary ostium and absent right coronary ostium for right coronary artery.

DISCUSSION

The coronary artery anomalies can be classified into anomalies of origin, course or termination or as hemodynamically significant or insignificant.⁵ Hemodynamically significant anomalies are characterized by abnormalities of myocardial perfusion, leading to increased risk of myocardial ischemia or sudden death.⁶ These include an anomalous origin of either the Left coronary artery or Right coronary artery from the pulmonary artery, an anomalous course between the pulmonary artery and aorta (inter-arterial) of either the Right coronary artery arising from the left sinus of Valsalva or the Left coronary artery arising from the right sinus of Valsalva, and, occasionally myocardial bridging or congenital coronary artery fistula.

In the study conducted by Eckart RE *et al*, in America, over a period of 10 years, in 902 cases of adjudicated unanticipated sudden cardiac death, they found that out of total 298 cases of <35 years of age anomalous coronary artery was seen in 12 (4%) cases while out of 604 cases of age ≥35 years, 1 (0.2%) cases showed anomalous coronary artery.⁷

In the study conducted by Desmet W *et al*, a database consisting of the angiographic reports of 50,000 consecutive coronary angiographies performed in adult patients in the University Hospital of Leuven between March 1973 and August 1991 was searched for the diagnosis of single coronary artery. All films concerned were reviewed and classified according to their anatomical type. Thirty-three cases of single coronary artery were retrieved, yielding an incidence of 0.066%.

In a case reported by Tanawuttiwat T *et al*, a patient was evaluated by 64-slice multidetector computed tomography of the coronary arteries, which revealed an anomalous single coronary artery arising from the left sinus of Valsalva, together with an absence of the right coronary artery (RCA). The left circumflex coronary artery (LCx) was the dominant vessel; it appeared to continue, without significant stenosis, beyond the atrioventricular groove up to the level normally occupied by the RCA. The left main coronary artery bifurcated into the left anterior descending coronary artery (LAD) and the LCx. Only a single coronary ostium (that of the left main) was seen to arise from the aorta. Shammas RL *et al*, in 2001 reported two unusual cases of agenesis of right coronary ostium with

continuation of left circumflex artery as the right coronary. Kang WC *et al*, also reported a case of unusual dominant course of left circumflex coronary artery with absent right coronary artery during angiography in 2006. Gleeson T *et al*, in 2009, reported a case of a single coronary artery with origin from a single ostium in the right sinus of Valsalva with an anomalous course of the left coronary artery anterior to the pulmonary trunk.

Jo Y *et al*, in 2011 reported a case of sudden cardiac arrest associated with anomalous origin of the right coronary artery from the left main coronary artery in a 24 year old man.¹² Patel MP *et al*, in a study in 2012 in Gujrat determined the incidence of single coronary artery in unsuspected population was 0.48%.¹³

Satija B *et al*, in 2012 reported a case of an anomalous origin of a right coronary artery from the left coronary sinus with an inter-arterial course, between the aorta and the main pulmonary arterydetected by multidetector row computed tomography coronary angiography. This variant has been called malignant because of its association with sudden death, especially in young asymptomatic athletes.¹⁴

In our case we incidentally found single anomalous coronary artery originating from the left sinus of Valsalva and absence of right coronary ostium and right coronary artery in a case of hanging. But this young person was having this undiagnosed congenital coronary anomaly along with atherosclerosis in coronaries which might lead to sudden cardiac death later on if this person had not commit suicide by hanging.

CONCLUSION

In our case we describe a 36 years old male who committed suicide by hanging, due to personal reasons. During autopsy on dissection of heart, right coronary ostium and right coronary artery was absent. A single coronary artery was originating from the left sinus of Valsalva with a dominant left circumflex artery which was continuing distally into the right side of the heart. On histopathological examination both Left main coronary artery and Left anterior descending artery showed Grade 4 to Grade 5 atherosclerosis and about 25% lumen narrowing. This case emphasises the requirement of essential investigations and preventive measures to prevent sudden cardiac death in young individuals having rare congenital anomalies of coronary arteries and the role of meticulous autopsy to detect them.

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