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Case Report

Single coronary ostium in right aortic sinus without other major cardiovascular abnormalities presenting as angina pectoris in an adult- a rare anatomical variant

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ABSTRACT

Anomalous origin of coronary arteries represents a clinical challenge because of the anatomical variability, possible functional consequences and the lack of large published series. We report a 48 yr old diabetic female who presented with chest pain for 6 months. Her ECG, 2D Echo and TMT were normal. Coronary angiogram was done in view of persistent symptoms and diabetes which showed single coronary ostium in the right aortic sinus with anomalous origin of LMCA from proximal superdominant RCA. Epicardial coronaries were normal. CT coronary angiogram was done to evaluate the course of anomalous LMCA, which showed prepulmonic course excluding any extrinsic compression. What makes this case unusual is that; single coronary ostium without other major cardiovascular abnormalities in itself is a rare entity. To our knowledge, the anatomical subtype of our patient according to Shirani J et al classification of "solitary coronary ostium in aorta" is a rare subtype. In view of presentation at this age, prepulmonic course of LMCA, this anomaly was thought to be benign and she was advised medical management. Cause of angina in our patient could be multifactorial like microvascular angina, coronary spasm or exercise induced ischemia in the usual LCX territory as reported in patients with super dominant RCA. This case demonstrates the importance of evaluating young individuals with chest pain, since sudden death can occur with an anomalous coronary.

1. Introduction

Anomalous origin of coronary arteries represents a clinical challenge because of the anatomical variability and possible functional consequences, the pathophysiological mechanisms involved, and the lack of large published series that would provide evidence to guide the clinical and therapeutic approach. Representing one of the rarest forms of coronary anomalies is an origin of the entire left coronary artery (LCA) from the right coronary artery. Although these anomalies are present at birth, they are often not diagnosed until late adolescence or adulthood, due to the lack of symptoms or because the symptoms may not be recognized. When the LCA originates from the right sinus of Valsalva or right coronary artery, the anomalous artery pathway can present in four variants. The deadliest of pathways, with the worst prognosis, is the inter-arterial course. This is when the LCA makes its course between the aortic root and right ventricular outflow tract (RVOT). Myocardial ischemia, ventricular fibrillation, syncope, congestive heart failure and sudden cardiac death are associated with this anomaly. Most literature would agree that coronary angiography is the primary modality for discovering these anomalies. There are other modalities, including multi-slice computed tomographic angiography (CTA), magnetic resonance angiography (MRA), and transesophageal echocardiogram (TEE) that have also been used to diagnose anomalous pathways.

2. Case report

A 48-year-old female patient who was nonhypertensive and diabetic without other cardiovascular risk factors presented with chest discomfort on exertion, palpitations and fatigue for the last 6 months. The electrocardiogram was normal, and 2-dimensional

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echocardiography revealed normal left ventricular function, no left ventricular regional wall motion abnormalities. Hence, she was subjected to exercise ECG stress testing, which was negative for inducible ischemia. Coronary angiography was done in view of persistent symptoms and diabetes. While searching for the left coronary artery, to our surprise, the origin of the left coronary artery was found to be from the right coronary sinus. It was arising from the proximal part of right coronary artery. RCA was dominant supplying some part of usual LCX territory also. Epicardial coronaries were free of atherosclerotic disease. In view of abnormal origin of left coronary we proceeded with CT coronary angiography which showed normal prepulmonic course of LAD excluding any extrinsic obstruction. Presently patient is on betablockers, antianginals and doing well in our follow up.

3. Discussion

Congenital coronary artery anomalies are rare occurrences, with a reported incidence of 0.3% to 1.3%.,[1,2] The most frequently found anomalies include a circumflex artery (CX) originating with a separate ostium from the left sinus of Valsalva, or left coronary cusp; an origin of the CX taking off from the right coronary artery; or the CX arising separately from the right sinus of Valsalva, or right coronary cusp. However, anomalous origin of the LCA from the right sinus of Valsalva separately or from RCA is a rare anomaly reported in 0.09% to 0.15% of cases. [3–5] The anomalous LCA can take various courses. These various proposed courses are classified according to the pathway to left (contralateral) aspect of the heart:

Type A – Anterior: LCA turns anteriorly in front of RVOT;

Type B – Inter-arterial: LCA lies between the aorta and RVOT;

Type C – Septal: LCA courses through the crista supraventricularis portion of the septum;

Type D – Posterior: LCA turns posteriorly behind aorta.,[6,7]

The inter-arterial course has been known to have the worst prognosis and be associated with sudden cardiac death (>50%), particularly during or shortly after exercise.[8] Many have hypothesized as to why this phenomenon occurs. Myocardial ischemia and cardiac death can occur due to impaired coronary flow either by

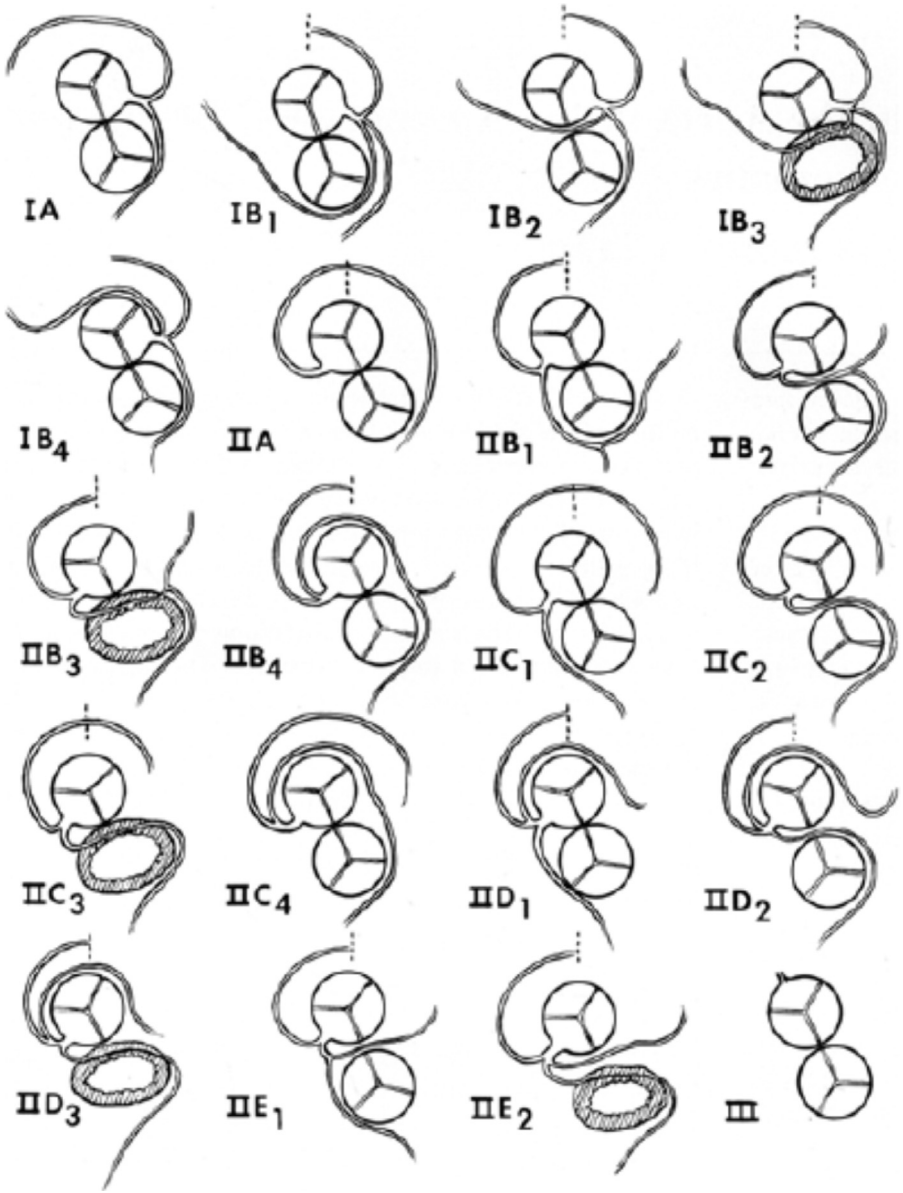

Fig. 1. Diagram showing the origin and pattern of distribution of various types of solitary coronary ostium in the aorta.