Case Report

Single coronary artery arising from the left sinus of valsalva

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ABSTRACT

A single coronary artery is a rare congenital anomaly of the coronary arteries where only one coronary artery arises from the aortic trunk by a single coronary ostium, supplying the entire heart. We report a case of a 57-year-old woman with atypical chest pain, in whom coronary angiography showed a L-I subtype single coronary artery (arising from a single ostium in the left sinus of valsalva) without associated cardiovascular disease. The clinical significance and subtype of the single coronary artery are discussed.

Saudi Med J 2002; Vol. 23 (12): 1537-1540

Single coronary artery is a rare congenital anomaly of the coronary arteries in which only one coronary artery arises from the aortic trunk with a single coronary ostium. In 1716, Thebesius[1] reported the first case of single coronary artery. In 1967, the first antemortem diagnosis was made by means of coronary angiography.[2] The prognosis of individuals with this finding is unclear. Some authors report that the presence of a single coronary artery has no impact on life span,[3] but there is evidence that these patients have a high incidence of sudden death.[4]

Case Report. A 57-year-old female with palpitations and chest pain was admitted to Sani Konukoglu Medical Center, Gaziantep, Turkey. Physical examination revealed that blood pressure was 150/100 mm Hg. Heart rate was noted as mean 110/min (maximal: 135/min, minimal: 95/min) with a regular rhythm on 24 Holter electrocardiographic monitoring. The routine laboratory findings were normal. Electrocardiogram showed sinus tachycardia, the non-specific changes of time of eletrical activity stopped - wave of ventricular repolarization (ST-T) segments and left ventricular hypertrophy. Coronary angiography was then performed using L Amplatz 2 catheter in standard projections. In coronary angiography, it was observed that the single coronary artery originated from the left sinus of valsalva **(Figure 1a)**. Its angle take off was usual. It was no observed the odd angulation and slit-like orifice of the single coronary artery. The single coronary artery gave off the anterior descending branch in the usual fashion and then continues on in the atrioventricular groove as the left circumflex branch. It traveled beyond the crux into the right atrioventricular groove where it provides branches to the right ventricle and atrium **(Figures 1b & 1c)**. The markedly dominant left coronary artery perfuses the entire myocardium. The right coronary artery ostium was congenitally absent. No significant coronary artery lumen narrowing was found. The patient had no evidence of cardiovascular diseases or any congenital anomaly. When the sinus

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Received 25th March 2002. Accepted for publication in final form 15th June 2002.

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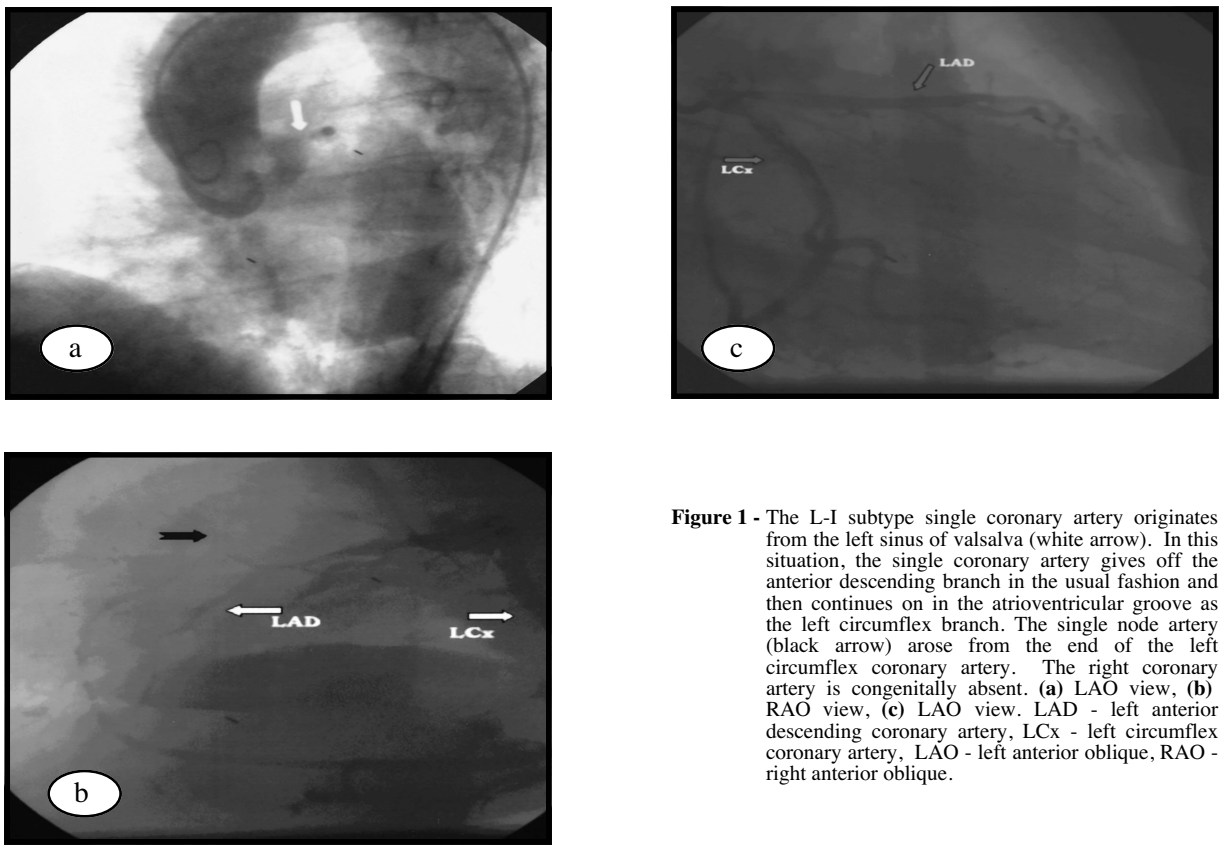

Figure 1 - The L-I subtype single coronary artery originates from the left sinus of valsalva (white arrow). In this situation, the single coronary artery gives off the anterior descending branch in the usual fashion and then continues on in the atrioventricular groove as the left circumflex branch. The single node artery (black arrow) arose from the end of the left circumflex coronary artery. The right coronary artery is congenitally absent. **(a)** LAO view, **(b)** RAO view, **(c)** LAO view. LAD - left anterior descending coronary artery, LCx - left circumflex coronary artery, LAO - left anterior oblique, RAO - right anterior oblique.

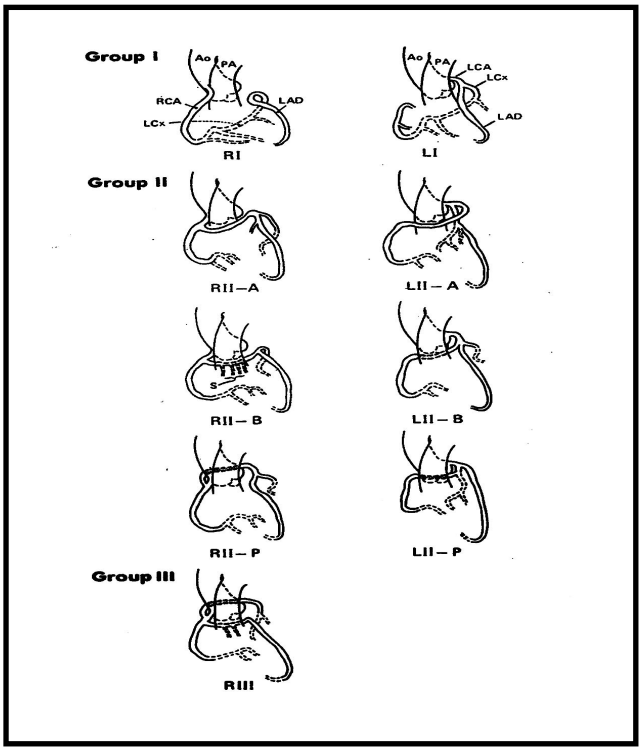

Figure 2 - The angiographic types of isolated single coronary artery patterns were classified by Lipton et al.[5] R - right, L - left, RCA - right coronary artery, LCA - left coronary artery, LAD - left anterior descending coronary artery, LCx - left circumflex coronary artery, Ao - aorta, PA - pulmonary artery, A - anterior, B - between, P - posterior to the great vessels. Shading (drawing small line) of the transverse trunk indicates this artery courses posterior to the aorta. According to Lipton's classification, our case is the same L-I subtype single coronary artery.

node artery was examined, it was observed that it arose from the end of the left circumflex coronary artery and it was very slight **(Figure 1c)**. In the present case, the single coronary artery was classified as type L-I according to previous studies.[5,6] This coronary artery anomaly was thought to be the cause of the atypical chest pain and the patient was maintained on oral medication.

Discussion. A single coronary artery ostium, in the absence of other cardiac disease, is a rare coronary artery anomaly, as an isolated single coronary artery occurs in approximately 0-0.7%[5,7] of the population. In association with certain other congenital anomalies, however, it is found considerably more frequently; Butto et al[8] reported that the incidence in a series of 54 cases of persistent truncus arteriosus was 18.5%, and in another paper it was reported to be 18% by De la Cruz et al,[9] Shrivastava et al[10] examined coronary angiograms of 296 patients with tetralogy of Fallot and they found a single coronary artery in 7 cases and Calder et al[11] found that the incidence of the single coronary artery in pulmonary atresia was approximately 17%. According to the site of origin and anatomical distribution of the branches, isolated single coronary arteries were classified in 3 groups in 1979 by Lipton et al,[5] **(Figure 2)**, further modified in 1990 by Yamanaka and Hobbs.[6] The anomalous coronary artery is first designated with "R" or "L" depending upon whether the ostium is located in the right or left sinus of valsalva. It is then designated as group I, II, III. Group one have an anatomical course of either a right or left coronary artery. In the R-I variant a very large right coronary artery continues in the atrioventricular groove extending to the anterior base of the heart where it gives rise to an anterior descending branch. The L-I pattern occurs when the right coronary artery is congenitally absent. In this situation, the single coronary artery gives off the anterior descending branch in the usual fashion and then continues on in the atrioventricular groove as the left circumflex branch. Group 2 anomalies arise from the proximal part of the normal right or left coronary artery, and cross the base of the heart before assuming the normal position of the inherent coronary artery. Group 3 describes the anomaly where the left anterior descending artery and left circumflex coronary artery arise separately from the proximal part of the normal right coronary artery. The final designation refers to the relationship between the anomalous coronary artery and the aorta and pulmonary artery. The letters "A", "B", and "P" refer to "anterior", "between", and "posterior" patterns. In 1990, Yamanaka and Hobbs[6] modified the Lipton et al[5] classification by adding "septal" (S) and "combined" (C) types, in order to describe the anatomical variations more precisely. The present case was classified L-I subtype single coronary artery according to previous studies.[5,6] Prognosis for patients with a single coronary artery varies from excellent with no decrease in life expectancy[3] to sudden death.[4] Taylor et al[4] studied sudden cardiac death associated with isolated congenital coronary artery anomalies in 242 patients. They found that sudden death occurred in 6 (14%) patients with single coronary artery of 44 patients with congenital coronary artery anomalies. Yamanaka and Hobbs[6] reported in their paper that a retired professional basketball player (Pete Maravich) with an unsuspected R-I subtype single coronary artery died suddenly during exercise. Halperin et al[2] reported a case of the patient with single coronary artery in congestive heart failure. Moodie et al[12] suggested an associated between single coronary artery of type R-II-B subtype single coronary artery (in which the anomalous left main trunk runs between great vessels) and sudden death during exercise. For the other types of single coronary artery, rare reports attribute myocardial ischemia to the coronary anomaly itself.[13-15] During cardiac surgery unexpected complications may also arise in patients with single coronary artery. Desmet et al[7] reported that an anomalous main stem in a patient with type R-II-P subtype single coronary artery in their series was accidentally ligated during elective replacement of a biodegenerated artificial valve in the aortic position necessitating immediate bypass grafting.

In the literature, Lipton et al[5] and Desmet et al[7] reported that the patients with L-I subtype single coronary artery had atypical chest pain although with no atherosclerotic heart disease. Surprisingly in the present study, the patient also had atypical chest pain without atherosclerotic heart disease. In 1974, Gould and Lipscomb[16] reported that single coronary artery may contribute to angina as a relatively small proximal vessel may either become diseased or may make distal coronary arterial lesions more hemodynamically significant by reducing coronary blood flow like a resistance in series.

In summary, the presence of a single coronary artery is an uncommon finding that needs to be considered during cardiac catheterization when both ostia cannot be identified and as a rare cause of atypical chest pain, myocardial ischemia, congestive heart failure and sudden death.

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