IHJ Cardiovascular Case Reports (CVCR) xxx (2017) xxx–xxx

Contents lists available at ScienceDirect

IHJ Cardiovascular Case Reports (CVCR)

journal homepage: www.elsevier.com/locate/ihjccr

A rare case with Supraventricular Tachycardia at presentation in an adult—a case report

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ARTICLE INFO

Article history:
Received 19 July 2017
Received in revised form 19 October 2017
Accepted 23 November 2017
Available online xxx

Keywords:
Anomalous Left coronary artery origin from Pulmonary artery
Supraventricular Tachycardia
Congestive Heart Failure
Adenosine

ABSTRACT

Anomalous left coronary artery origin from the pulmonary artery (ALCAPA), usually manifests in infancy. Its survival into adult age and manifesting as supraventricular tachycardia is rare and hence being presented.

1. Introduction

Anomalous Left Coronary Artery origin from the Pulmonary Artery (ALCAPA) is a rare congenital cardiac malformation requiring surgical treatment in infancy. Survival until adulthood and into the adult age group is rare. Presentation in adults is asymptomatic or with angina pectoris, dyspnea, congestive heart failure, sudden cardiac death after exertion. Its association with an supraventricular tachycardia in an adult at presentation is rare. It is the first case report to our knowledge in the medical literature.

2. Case Presentation

A 42 year old Male, presented with fever, shortness of breath and palpitations. He was referred to our hospital and was found to be having supraventricular tachycardia(SVT) and congestive heart failure (CHF). The tachycardia reverted to sinus rhythm by Inj. Adenosine 12 mg IV.

Initial Electrocardiogram(ECG) showed SVT with rate of 190/min, after reversion to sinus rhythm had heart rate of 90/min, axis of +90°,QS complexes in lead V1,V2,V3 and 1 st degree atrioventricular block with PR interval of 240 msec (Fig. 1A and 1B). His serial cardiac enzyme profile were negative and there was no progression of the ECG changes. Two dimensional echocardiography (2D Echo) showed scarred interventricular septum and global hypokinesia of the left ventricle (LV) with an ejection fraction of 40% (Fig. 2A). Chest X ray showed cardiomegaly with features of pulmonary edema (Fig. 2B).

Laboratory profile showed increased total leucocyte counts with predominant neutrophils. Blood urea was 28 mg/dl, serum creatinine was 0.8 mg/dl. Hemglobin was 13.0gm/dl. Ultrasonography of the abdomen showed mild hepatomegaly with congestive changes. Serum bilirubin was normal.

He was stabilized with oxygen supplementation, decongestive therapy and after four days a coronary angiography was performed to rule out the coronary artery disease etiology. His left main coronary artery could not be selectively engaged and could not be localized after sinus injection of the aortic root. Selective right coronary injection revealed dilated and prominent right coronary artery (RCA) with large collaterals of it filling the left coronary artery(LCA) retrogradely (Fig. 3A). Left coronary artery draining into the pulmonary artery could be demonstrated with long cine phase of right injection. The origin of the left coronary artery was tortuous (Fig. 3A). Post coronary angiography the renal parameters were normal.

An computed tomography (CT) coronary angiography and pulmonary angiography was done for correct delineation of the origin

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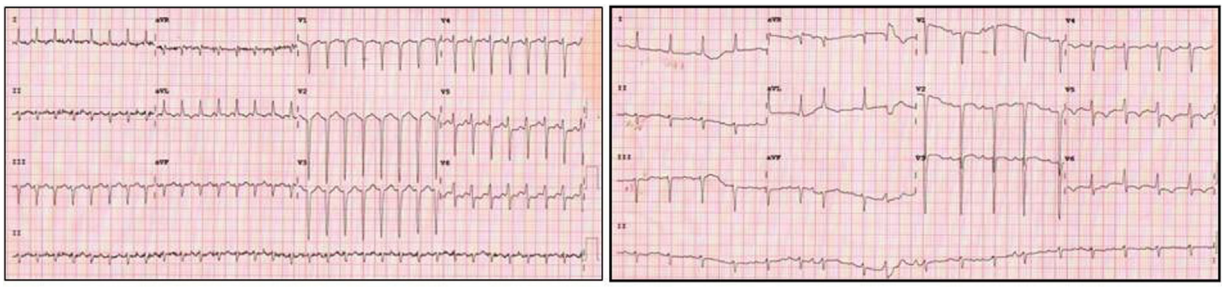
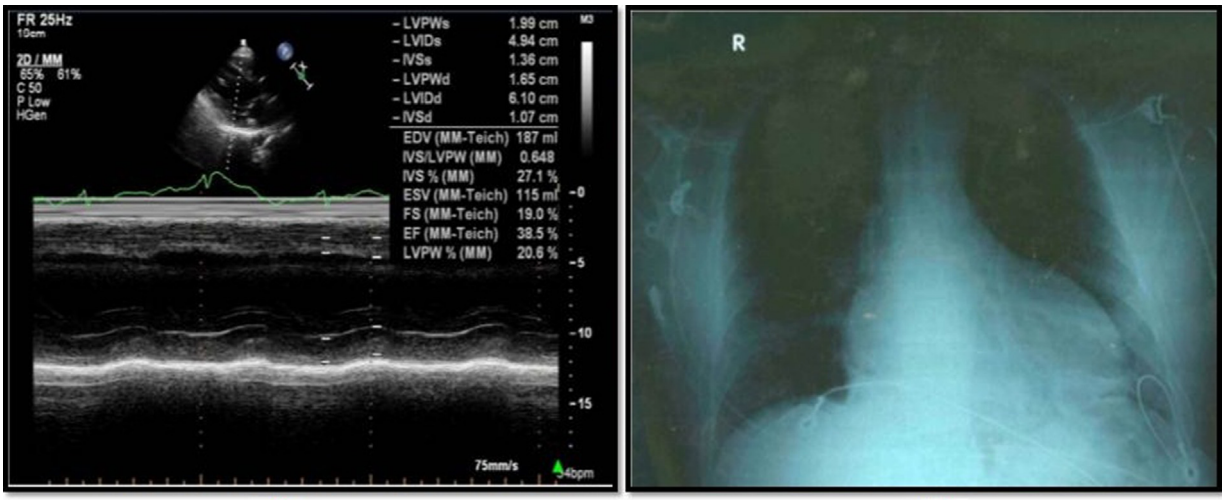
https://doi.org/10.1016/j.ihjccr.2017.11.004



1A 1B

Fig. 1. A. Supraventricular tachycardia with ventricular rate of 190/min. B – Post injection Adenosine 12 mg IV.



2A 2B

Fig. 2. A –M mode of LV showing moderate LV systolic dysfunction, EF = 40%. B – Chest X ray showing cardiomegaly, with dilated LV.

of the LCA and its collaterals from RCA. Left main coronary artery showed anamolous origin from posterior medial aspect of main pulmonary artery trunk just before its bifurcation. Short segment aneurismal dilation at the root of left main pulmonary artery noted (Fig. 3B). Left main coronary artery was coursing between pulmonary artery and aorta to short distance before its division into Left anterior descending artery and circumflex arteries (Fig. 3C). Right coronary artery was normal in course arising from right sinus and was dilated.

Patient has been referred to cardiothoracic surgeon for bypass or reimplantation of the left coronary artery into the aortic sinus. Electrophysiology study has been planned after the surgery and subsequent radiofrequency ablation. Patient is asymptomatic and stable at follow up.His recent echo revealed moderate left ventricular dysfunction.

3. Discussion

ALCAPA is a rare congenital malformation of the coronary circulation,with reported incidence of 1 in 300,000 live births.[1] It accounts for 0.25–0.5% of all congenital heart diseases.[1]The first case reported was by St.John Brooks in 1886.[2]It is also known as Bland –White – Garland syndrome.[3] Fontana and Edwards *et al* reported a series of 58 necropsies which revealed that most of the patients died at 13-15 months of age.[4] ALCAPA is not considered an inheritable congenital cardiac defect. No risk factors for the occurrence of ALCAPA in any individual family are known. ALCAPA is not assosciated with any syndromes or non cardiac conditions,but has been asssosciated with patent ductus arteriosus,[5]ventricular septal defect,coarctation of aorta.[6]

Natural history of the ALCAPA is that 80-90% die within the first year because of intractable congestive heart failure which presents after 2-3 months of age.[7] About 10-15% survive to adulthood.[8] The disorder can be asymptomatic until the adult age because of extensively developed collaterals from the right coronary artery.Though asymptomatic there is high risk of sudden cardiac death,which usually occurs during exertion.[9] Presentation can be with dyspnea on exertion,angina pectoris,atrial fibrillation,chronic mitral insufficiency and congestive heart failure.[9]

Presentation of ALCAPA in the neonate is unsual because of the elevated pulmonary arterial pressure which results in the antegrade flow in the anamolous left coronary artery.[10] With a gradual decrease in the pulmonary vascular resistance, usually seen at 2-3 months of age,there can be myocardial infarction due to coronary steal phenomenon secondary to decrease in the antegrade flow in the left coronary artery.[11] The symptoms can be irritability,dyspnea,wheezing,cough and diaphoresis. Sudden cardiac death can result in absence of extensive collateral blood flow from the right coronary artery. Children who survive this critical phase have failure to thrive features as a result of the chronic congestive heart failure. About one third of patients who survive the childhood present with sudden cardiac death.[12]

The anomalous left coronary artery is thin walled resembling a venous channel.[13]The right coronary artery originates from its

Please cite this article in press as: Nagula P, et al, A rare case with Supraventricular Tachycardia at presentation in an adult—a case report, *IHJ Cardiovasc Case Rep*. (2017), https://doi.org/10.1016/j.ihjccr.2017.11.004