Medical Science and Discovery 2015;2(6):358-61

Doi: 10.17546/msd.07800

Anomalous origin of left coronary artery from pulmonary artery; Congenital anomaly presenting with dyspnea. A rare case study

Shahriar Anvari^{1*}, Sohrab Negargar², Ahmad Jamei Khosroshahi¹

Abstract

Anomalous origin of left coronary artery from pulmonary artery (ALCAPA) is rare congenital anomaly. Most of these patients die is infancy. Presentation in adulthood is very rare. Clinical manifestation in teenagers or young adult contains arrhythmia, myocardial perfusion likely causes significant chest pain and these symptoms of myocardial ischemia may be misinterpreted as routine infantile colic and sudden death.

Keywords: Anomalous origin of left coronary artery from pulmonary artery

Introduction

Anomalous origin of left coronary artery from pulmonary artery (ALCAPA) is a rare lesion with an estimated incidence of between 1 in 30000 and 1 in 300000. It is frequently lethal in early infancy with some reports suggesting a mortality rate as high as 90% in first year of life.

Case

Our case is an adult survivor of ALCAPA diagnosed at our hospital. The patient is a 55 years old female with history of effort dyspnoea (FC=II) from a few years ago. Patient has history of 12 time gestation deliveries without any problem. Physical 8 examination was normal only an II/VI systole murmur auscultate in left sternal border. Vital sign was normal. CXR was not remarkable. ECG was normal. Two dimensional echo revealed moderate mitral regurgitation and moderate left ventricular dilatation and mild LV dysfunction with EF about 45-50%, but it not seen any clue of ALCAPA. Then patient went for coronary angiography that revealed typical anatomy of ALCAPA. On cardiac CT angiography, typical anatomy of ALCAPA was detected.

Discussion and Conclusion

ALCAPA is rare lesion with an incidence of about 1 in 100000 accounting for 0.25% of congenital heart disease(1). The anomalous left main connects most often to the sinus of Valsalva immediately above the left of posterior cups of pulmonary trunk and rarely from that above the right cup. Collateral

between right & left coronary arteries always presents and grossly visible mainly is adults. Left ventricle is always hypertrophied and greatly dilated. Diffuse LV fibrosis is always present and patients dying in infancy usually leave evidence of anterolateral myocardial infarction. A considerable amount of LV dysfunction in infants must be ischemic in origin. There are some reasons for mitral regurgitation. There may be extensive fibrosis and sometime calcification in papillary muscles. Endocardial fibro-elactosis may involve mitral valve (2).

In patients who survive into adulthood, collateral circulation from right coronary artery is apparently adequate to prevent sever left ventricular failure (3). Presentation is often delayed beyond age 20 years. About half have effort dyspnoea. Occasionally a mitral regurgitation dominates clinical picture. Resting ECG is always abnormal with ST-T segment changes or evidence of old anterolateral infarction. Exercise ECT usually shows ischemic changes thallium is usually abnormal. CXR may be normal or shows cardiomegaly (4). Echocardiography (2-D) is the principle tools for diagnosis.

It may show enlarged RCA or dilated LV or abnormal regaining of LM from pulmonary trunks. Angiography shows more collateral in adults than infants and shows near normal or mildly decreased LV function. Coronary angiography is considered the gold standard technique for diagnosis (5). Most patients who survive infancy continue to be at risk of death from chronic heart failure and those who survive until the fourth decade occasionally die suddenly once diagnosis of ALCAPA is established.

Received: 17-06-2015, Accepted 15-07-2015, Available Online 01-10-2015

¹Cardiovascular Research Center, Tabriz University of Medical Sciences, Tabriz, Iran.

²Dept. of Anesthesiology, Cardiovascular Research Center, Tabriz University of Medical Sciences, Tabriz , Iran.

^{*}Corresponding Author: Shahriar Anvari E-mail: anvari_shahriar@yahoo.com

Early surgical correction including difference type of construction a two artery coronary system for prevention of complication and increase of survival is indicated (6). **Conflict of Interest:** The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Acknowledgments: Authors would like to thank the patient and all of our colleagues who helped us in this study.



Figure 1. Computed Tomographic angiography showing dilated right coronary artery with continuation with left coronary artery.



Figure 2. ECG is showing normal pattern view.



Figure 3. Chest – X – ray is showing mild cardiomegaly without pulmonary congestion.



Figure 4. Trans – Thorasic – Echocardiography shows dilated right coronary artery without evidence of left coronary artery.



Figure 5. Comptuded tomographic angiography shows much dilated right coronary artery with normal origin of aorta and abnormal origin of left coronary artery from pulmonary artery.

References

- Nicholas T. Kouchoukos, Eugene H. Blackstone, Frank L. Hanley, and James K. Kirklin, Cardiac surgery: kilkilin Barratt — boyes. Fourth edition. 2013. p. 1650-62.
- 2. Jonas RA. Comprehensive surgical management of congenital heart disease. 2014. p. 663-71.
- Parale GP, Pawar SS. Adult type anomalous left coronary artery from pulmonary artery, case report. 2006; 54:397-9.
- 4. Brooks H. two case of abnormal coronary artery of the heart, arising from pulmonary artery, with some remarks upon the effect of this anomaly in producing cricoid dilatation of vessel. Journal of anatomy and physiology. 1885; 20:26-9.
- Aykan AC, Yıldız M, Kahveci G, Ozkan M. Two adult cases of anomalous left coronary artery from the pulmonary artery. Turk Kardiyol Dern Ars. 2012;40(1):48-51.
- 6.
- Liu Y, Miller BW. ALCAPA Presents in an Adult with Exercise Intolerance but Preserved Cardiac Function. Case Reports in Cardiology. 2012;2012:3.

Copyright © 2014 The Author(s); This is an open-access article distributed under the terms of the Creative Commons Attribution License (http://creativecommons.org/licenses/by/4.0), which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited. All Rights reserved by international journal of Medical Science and Discovery.