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Recurrent syncope in an adolescent male: A case report of cor triatriatum

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Abstract

A 17-year-old male with no prior medical history, presented with recurrent syncopal episodes, the latest preceded by headache, nausea and abdominal pain. On physical examination, his blood pressure was 120/74 mmhg, pulse was 80 bpm, regular heart beat and clinical was no features of genetic abnormalities appearance. ECG showed sinus bradycardia, T-wave inversion over lead III, flat T wave on lead aVF. Holter found minmal heart rate was 36 bpm, average heart rate was 58 bpm, maximum heart rate was 132 bpm, one-time premature atrial contractions was found and no premature ventricular contractions. Transthoracic (Figures 1 and 2) echocardiography found a flap on left atrium with shunt to left ventricle, demonstrated Cor Triatriatum.

Keywords: Cor triatriatum; Syncope; Congenital heart disease.

Introduction

Cor Triatriatum is an uncommon congenital heart defect (prevalence: 0.1-0.4% of cardiac anomalies) characterized by a membrane dividing the left atrium or right atrium. Cor Triatriatum obstructs pulmonary venous return, leading to pulmonary hypertension, heart failure, or thromboembolism. The characterized by various clinical presentations, influenced by the extent of membrane blockage and concurrent heart complication, the prevalence of asymptomatic cases in children with Cor Triatriatum ranges widely, making the diagnosis challenging to determine an exact figure. In adults, the common symptoms including palpitation, exertional dyspnea and orthopnea, syncope is a rare presenting symptom, often misattributed to neurological causes. In the patient presented with obstructive Cor Triatriatum, usually associated with congestive heart failure and pulmonary hypertension. We report a case of Cor Triatriatum in an adolescent presenting with recurrent syncope and atypical prodromal symptoms.

Case presentation

A previously health 17-year-old male without special medical history presented with recurrent syncope, preceded by headache, dizziness and weakness. On examination, he was afebrile with a normal breathing rate. The pulse rate was 80 bpm, blood pressure 120/74 mmHg, and oxygen saturation 97% under room air, regular heart rate and normal heart sound without significant murmurs. ECG showed sinus bradycardia, T-wave inversion over lead III, flat T wave on lead aVF. Holter found minimal heart rate was 36 bpm, average heart rate was 58 bpm, maximum heart rate was 132 bpm, one-time premature atrial contractions was found and no premature ventricular contractions nor atrial fibrillation. A transthoracic Echocardiography revealed a Cor Triatriatum sinister, normal left ventricle size, no myocardial hypertrophy, no regional wall motion abnormalities, Left Ventricular Ejection Fraction (LVEF) 70%, right heart normal size with normal systolic function, Tricuspid Annular Plane Systolic Excursion (TAPSE) 2.0 cm, no pulmonary artery hypertension.

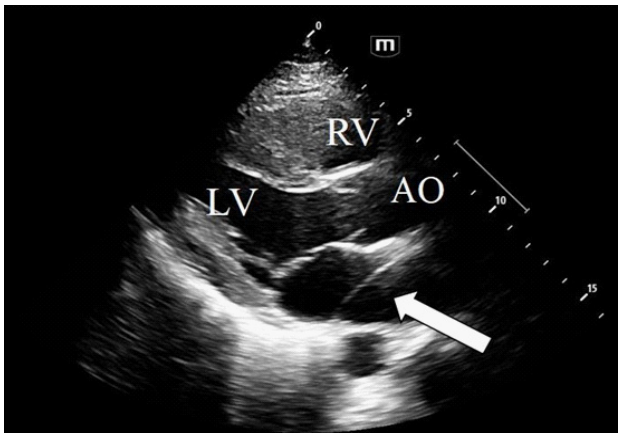

Figure 1: Parasternal long axis view showing a flap crossing the left atrial chamber (white arrow). LV, left ventricle, RV, right ventricle, AO, aorta.

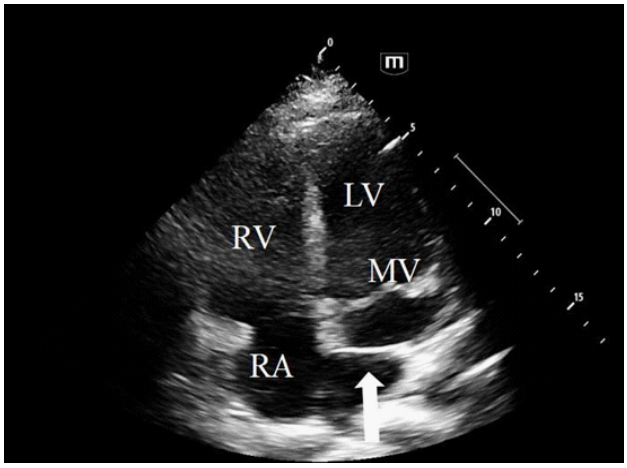

Figure 2: Apical 4 chamber view showing a horizontal flap crossing the left atrial chamber (white arrow). LV, left ventricle, RV, right ventricle, RA, right atrium, MV, mitral valve.

Discussion

Cor Triatriatum presents with a fibromuscular membrane dividing the left atrium into proximal part of the atrium with the pulmonary vein entry from a distal part communicating with the left ventricle through the mitral valve. The mechanism involved an abnormal incorporation of the common pulmonary vein in the left atrium. It can present in isolation or associated with other congenital heart disease [1]. Cor Triatriatum is classified according to various systems, such as the Loeffler classification, Lam classification, the modified Lucas classification, and the Mashadi-Narasimhan-Said classification. The Loeffler classified three presentations of Cor Triatriatum according to the amount of communication provided by the occluding membrane: Type 1 has no opening in the accessory membrane, with the proximal left atrium draining into the right atrium; type 2 has one or more small restrictive openings, resulting in significant left ventricular inflow obstruction; and type 3 has a large opening in the membrane [2].

The obstructive symptoms depending on the communication level within the two-chambered left atrium. Patient may present from asymptomatic to severe symptomatic. The symptoms in infants with Cor Triatriatum typically due to pulmonary hypertension, may presented with cough, tachypnea, tachycardia, difficult feeding, poor weight gain. In severe cases, the symptoms may present similar mitral stenosis or pulmonary vein stenosis and usually require early surgical repair for the pulmonary congestion and decrease cardiac output.

In adult, they are usually asymptomatic due to presence of a large foramen without a significant intra-atrial pressure gradient. Symptoms including palpitation, exertional dyspnea, orthopnea and syncope. The late symptoms may relate to the patient development of the pulmonary hypertension, mitral regurgitation or the membrane orifice obstruction cause by calcification. During physical examination, the patient may manifest as a diastolic murmur from mitral stenosis with absence of an opening snap and a distinct loud S1 with a pronounced second heart sound (P2) if presented pulmonary hypertension [3].

The management of Cor Triatriatum depending the patient symptoms, sign and institutional practices. A systematic review by Ullah et al. compiled 235 studies, the management of Cor Triatriatum included surgery (68.51%), medical management (12.23%), observation (12.79%), and balloon septoplasty (6.29%) [4]. Generally, the medical treatment has no role in the treatment of Cor Triatriatum, the best treatment is surgery. In a study by a Mayo clinic, reported Twenty-five patients underwent surgical correction of Cor Triatriatum between May 1960 and September 2012. All patients underwent excision of Cor Triatriatum membrane using cardiopulmonary bypass. Twenty patients (80%) required concomitant cardiac surgical procedures. There was no early mortality. None of the patients had any residual atrial obstruction. Two infants who had concomitant repair of complex congenital anomalies died at 2 and 5 months postoperatively after discharge from hospital. Kaplan-Meier survival at 10 years was 83%. All patients were in New York Heart Association class I or II at a mean follow-up of 12.8 years (maximum 44 years) [5].

The outcome of Cor Triatriatum, a retrospective review of patients with Cor Triatriatum followed at Mayo Clinic Rochester from 1990 to 2016. Fifty-seven patients (median age 34 years; men 32(56%)) were enrolled. In these 57 patients, initial diagnosis was made in adulthood in 35(61%) patients, and 33(58%) patients had additional Congenital Heart Disease (CHD) diagnosis. A total of 27(47%) patients required surgical resection of Cor Triatriatum membrane during median follow-up of 76 months. There was one perioperative mortality and no late mortality. There was no recurrence of Cor Triatriatum membrane obstruction in the patients that underwent surgery.

Similarly there was no significant increase in Cor Triatriatum membrane gradient in the patients without left atrial obstruction that were managed conservatively. The conclusions are the natural history of Cor Triatriatum is stability without progressive left atrial obstruction, especially in patients with isolated Cor Triatriatum and in those with initial Cor Triatriatum diagnosis made in adulthood. In patients requiring surgical membrane resection due to flow obstruction, surgery is safe and effective with very low risk of recurrence [6].

Conclusion

Cor Triatriatum is a rare congenital heart anomaly with challenges in diagnosis and management. This system review of 25 patients provides valuable insights into the evaluation, diagnostic, and treatment outcomes. The management of Cor Triatria-

tum continues to require a tailored approach, considering the anatomical diversity and associated cardiac anomalies. Moreover, the unique classification systems, such as the Loeffler and Lam classification, have been instrumental in enhancing our understanding of the anatomical variations within Cor Triatriatum. The case report emphasized the importance of early and accurate diagnosis, timely surgical intervention, and a personalized case-by-case management of different cases.

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