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Case Report

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Silent Messenger of Anomalous Origin of the Left Coronary Artery from the Pulmonary Artery (ALCAPA)

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Abstract Abnormal origin of the left coronary artery from the pulmonary artery syndrome (ALCAPA) is a rare congenital heart disease. While in is usually seen is an isolated anomaly, other heart anomalies may also be seen in 5% of cases. There are several case reports in the literature showing that mitral regurgitation in children may be a silent precursor of ALCAPA, especially in late presenting cases. Therefore, coronary arteries should be carefully evaluated, particularly in children with mitral regurgitation. A 4-year-old asymptomatic male who was followed up with the diagnosis of mild mitral regurgitation and mitral cleft was last checked 2 years ago. In the current evaluation, despite no problems being detected in the patient's physical development and exercise capacity, a mesocardia systolic 2/6 murmur was detected. In the ECG, the T-wave changes in D-I and aVL were remarkable. In the echocardiographic examination, evident views were obtained.Following the echocardiographic evaluation, the patient underwent catheterization and the was referred for correction surgery. Mitral regurgitation may be an early and innocent-looking messenger of the abnormal origin of the left coronary artery from the pulmonary artery syndrome. There are a few case reports in the literature indicating that mitral regurgitation in children is a silent precursor of ALCAPA. Our aim is to contribute to the literature to make this important condition more known. Therefore coronary arteries should be carefully evaluated, particularly in children with mitral regurgitation.

Keywords ALCAPA · coronary anomaly · mitral regurgitation · interarterial collaterals

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INTRODUCTION

Abnormal origin of the left coronary artery from the pulmonary artery (ALCAPA) syndrome, also known is Bland-White-Garland syndrome, is a rare congenital heart disease that affects approximately 1 in 300,000 live births and accounts for 0.25-0.5% of all congenital heart anomalies. [1]While in is usually seen is an isolated anomaly, other heart anomalies may also be seen in 5% of cases. Two forms are seen. In the infantile type, the disease has a severe course and 90% of the patients die within the first year of life. Reduced coronary perfusion causes ventricular dysfunction, dysrhythmias, mitral regurgitation, and sudden cardiac death due to myocardial infarction and congestive heart failure. [1,2] On the other hand, in the adult form, adequate collateral circulation development can provide a late-onset presentation with chronic myocardial ischemia and dysrhythmias. In may also lead to sudden cardiac death. The treatment of ALCAPA is surgery to reestablish a two-coronary system.

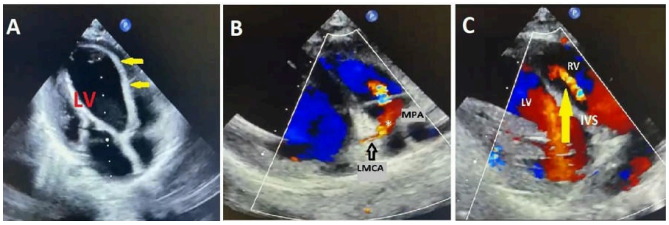
CASE PRESENTATION

We present a case of a 4-year-old asymptomatic male who was followed up with the diagnosis of mild mitral regurgitation and mitral cleft? The patient was not taken for regular follow-up. The was last checked 2 years ago. In the evaluation made in our center, no problems were detected in the patient's physical development and exercise capacity. On physical examination, a 2/6 systolic murmur on mesacardiac area was detected. In the ECG, the T-wave changes in D-I and aVL were remarkable. On echocardiographic examination, the left ventricle was slightly enlarged (LVEDD Z score: 2.3) and appeared spherical. Mitral leaflets are coarsened and coaptation is impaired with moderate regurgitation. Coronary circulation was prominent along the interventricular septum, and the left main coronary artery (LMCA) was arising from the main pulmonary artery. Reverse flow was also observed from the LMCA into the main pulmonary artery. (Figure-1) Following echocardiographic evaluation, the patient underwent catheterization with the preliminary diagnosis of ALCAPA. (Figure-2) After angiography, the patient's diagnosis was confirmed and the was referred for correction surgery.

DISCUSSION

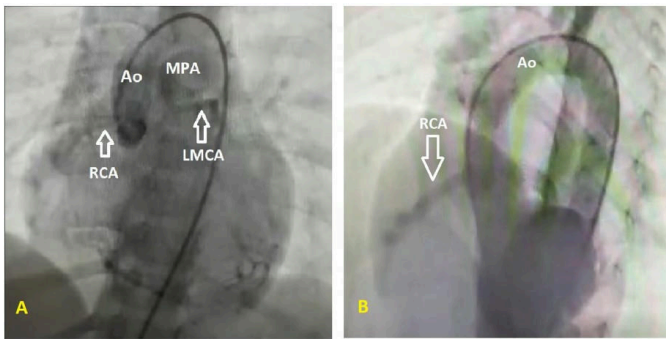
ALCAPA is an uncommon congenital heart anomaly with high mortality and morbidity. In may not be difficult to make a diagnosis, especially in the newborn period, by classic echo findings such is hyperechogenic papillary muscle, mitral insufficiency, and left ventricular dysfunction. [2,4,5] However, if the left ventricular structure and functions are not significantly affected in the early period, ALCAPA may not come to mind at first. Development of obvious left ventricular dysfunction due to incessant poor perfusion sometimes needs time due to interarterial collaterals.(3,4) In the literature, the diagnosis can be missed, particularly in late presenters.(6)

Figure 1. Echocardiographic evident views of ALCAPA



A: Arrows show enlarged and spherical left ventricle with coarsened mitral leaflets
B: Asterisk shows reverse flow from the LMCA into the main pulmonary artery
C: Arrow shows prominent coronary circulation along the interventricular septum
LV: Left ventricle, LMCA: Left main coronary artery, MPA: Main pulmonary artery, RV: Right ventricle, IVS: Interventriculer septum

Figure 2. Echocardiographic evident views of ALCAPA



Angiographic image of ALCAPA. While the right and left coronary arteries originate from seperate large vessels in Picture A, note the single right coronary artery arising from the aorta in Picture B.
*Ao: Aorta, MPA: Main pulmonary artery, RCA: Right coronary artery, LMCA: Left main coronary artery

In this case, the classical echocardiography findings of ALCAPA were not detected during infancy. During this period, we observed that a baby with ALCAPA who was not brought to the pediatric cardiology controls reached the childhood period without symptoms. On the other hand, a process that insidiously led to left ventricular enlargement and dilated cardiomyopathy occurred in the child who have only mitral insufficiency. Fortunately, the patient was diagnosed relatively early before cardiomyopathy was fully established and was saved from a possible heart transplant.Therefore, in patients with unexplained mitral regurgitation, the presence of ALCAPA must be considered.(6,7) This case report supports the literature that ALCAPA should be kept in mind when investigating the etiologies of unexplained mitral insufficiency.

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