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Unique Case of Anomalous Anatomy of the Right Coronary Artery from the Pulmnary Artery

Anomalous origin of the right coronary artery from the pulmonary artery is one of the rare congenital coronary diseases. The percentage of population damage is 0.002 %. In most cases this occurrence is asymptomatic and diagnosed accidentally like our case. Here, we report a 9-month-old female infant who was admitted with a diagnosis multiple muscle ventricular septal defects. For diagnostic we used echocardiogram, as a result an anomalous origin of the right coronary artery from the pulmonary artery was suspected. After that, coronary computed tomography angiography confirmed the diagnosis. The patient underwent surgical correction by redeployment of the coronary artery in aorta. The aim of our report is to describe the successful correction of a potentially fatal disease. Early diagnostic can provide effective treatment.

Key words: congenital heart defects; anomalous anatomy of the right coronary artery; right vetricular myocardial ischemia.

Introduction. Anomalous origin of coronary arteries is rare and found in about 1–2 % of the general population [1]. The coverage of coronary arterial anomalies including anomalous origin of the right coronary artery in normal heart ranges from 0.2 % of young patients undergoing echocardiography to 1.2 % of patients at computed tomography (CT) angiography [2]. As of result of the rarity of this pathology, there isn't much information found on this case, in literature. The first discoverer of the disease was John Brooks in 1885 [3]. The understanding of the process of coronary artery formation during development could potentially advance treatments. The embryological origin of anomalous origin of the right coronary artery from the pulmonary artery (ARCAPA) is unclear, but literary sources write about development disorders on the fourth to sixth week of gestation [4]. It is usually an isolated cardiac anomaly but, in 5% of the cases, has been described with coarctation of the aorta, atrial or ventricular septal defects [5]. With a significant left-right shunt, myocardial ischemia of the right ventricle may occur. Here, we report a 9-month-old female infant and this serves as a confirmation that early diagnosis and treatment is the best option for this defect.

Case report. The 9-month-old female with weight of 9,6 kg, was assessed with a diagnosis of multiple VSDs. Physical examination revealed blood pressure of 85/45mmhg. Trans- thoracic echocardiography showed multiple muscle VSDs, indirect sights anomalous of coronary arteries, initial dilatation of the right ventricle with an ejection fraction of 73 %, end diastolic volume of 31ml, total contractility of the left ventricle is prserved. Coronary CT angiography revealed dilated and tortuous right coronary artery (RCA) about 3mm in diameter at the right-upper contour of the proximal segment of the pulmonary artery (PA), pulmonary root 12x12x10 mm, pulmonary trunk with diameter of 14 mm, left and right branch of the PA 9 mm (Fig. 1).

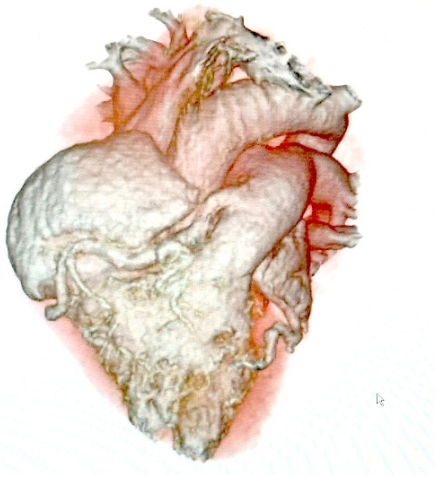

Fig. 1. Coronary CT angiography.

Electrocardiogram was without signs of myocardial ischemia. A Chest Xray showed the usual transparency of the lung fields, absence of fresh infiltrative focal shadows, pulmonary pattern is enriched basally (Fig. 2).

Based on the diagnostic data and review of the available literature the decision to proceed to surgical repair was undertaken. Surgery is the main treatment of ARCAPA. In the operating room, the exposed course of the anomalous right coronary artery was exactly as predicted by imaging. The surgery was performed by a standard sternotomy and surgical pro-

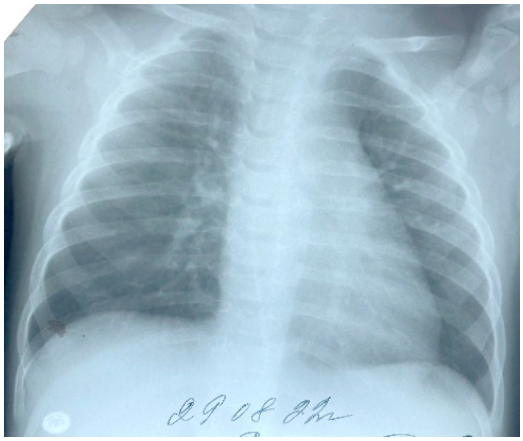

Fig. 2. Chest Xray.

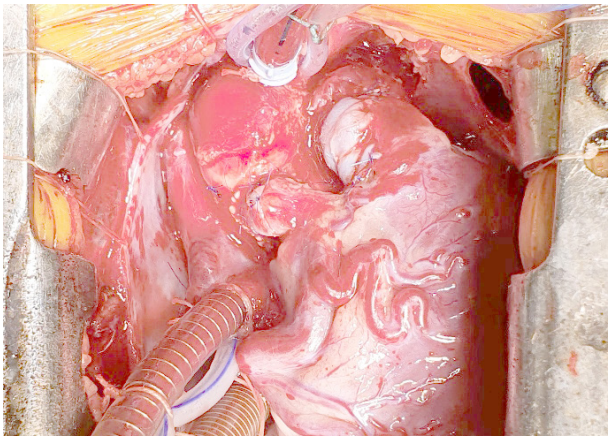

Fig. 3. After implantation RCA in the right coronary sinus of the aorta.

cedure were with circulatory support to hypothermia (28 C). We used standard aortic and bicaval venous cannulations. An aortic cross-clamp was applied. The left and right branches of the pulmonary artery where compressed. Pharmaco-cold cardioplegia was pumped into the root of the aorta. The right atrium was widely opened to vent the right ventricle and for left atrium drainage through the PFO. Further after dissection of the trunk of the pulmonary artery, additional selective cardioplegia was performed in the right coronary artery. The right coronary artery button was dissected from PA and then implanted in the right coronary sinus of the aorta by suture with 6-0 polypoprylene. The pulmonary artery was reconstructed with an autologous pericardial patch with a standard running suture 6-0. (Fig. 3).

The total duration of the operation was 390 min, perfusion time continued 179 min and aortic clamp time was 97 min. Cardioplegia pumped three times. Mechanical ventilation were weaned off 5 hours after surgery. There was no need for inotropic support.

The patient did not develop any post-operative complications. A follow-up echocardiogram revealed the right coronary artery implanted in the aortic artery with a laminar flow and preserved biventricular function. The patient was discharged on the twelve post-operative day, and was prescribed with spironolactone 12.5 mg/kg and furosemide 1 mg/kg and cefuroximum 200 mg/kg. The patient is currently undergoing asymptomatic rehabilitation.

Comment. We describe a rare case of ARCAPA, treated effectively with redeployment of the coronary artery in aorta. Given pathology is one of the rare congenital coronary diseases and in most cases this occurrence is asymptomatic and diagnosed accidentally like our case. For the further selection of treatment, we relied on the results of such diagnostic methods as coronary CT angiography (revealed dilated and tortuous RCA about 3mm in diameter at the right-upper contour of the proximal segment of PA, left and right branch of the pulmonary artery 9 mm). For our patient, the decision to proceed to the surgical correction at 9 months of age was made because this occurrence is the beginning of the development of severe complications. As of today, the patient is undergoing comprehensive rehabilitation and the child's development is taking place according to gender and age.

Authors' Note. The patient's parents consented to publication of the case report

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УНІКАЛЬНИЙ ВИПАДОК АНОМАЛЬНОГО ВІДХОДЖЕННЯ ПРАВОЇ КОРОНАРНОЇ АРТЕРІЇ ВІД ЛЕГЕНЕВОЇ АРТЕРІЇ

У статті наведено опис успішної хірургічної корекції вкрай рідкісного і потенційно смертельного захворювання.
Аномальне відходження правої коронарної артерії від легеневої артерії належить до рідкісних уроджених захворювань коронарної артерії. Відсоток ураження населення становить 0,002 %. Здебільшого це явище перебігає безсимптомно і діагностується випадково, як у нашому випадку. Тут ми розповідаємо про 9-місячну дівчинку, яка була госпіталізована з діагнозом множинні дефекти міжшлуночкової перегородки. Для діагностики використовували ехокардіографію, в результаті якої запідозрили аномальне відходження правої коронарної артерії від легеневої артерії. Після цього комп’ютерна томографія підтвердила діагноз. Дитині проведено хірургічну корекцію шляхом реімплантації правої коронарної артерії з легеневої артерії в аорту. Рання діагностика може забезпечити ефективне лікування.

Ключові слова: уроджена вада серця; аномальне відходження правої коронарної артерії; ішемія міокарда правого шлуночка.