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AN INFANT WITH CORONARY CAMERAL FISTULA – CASE PRESENTATION AND A REVIEW OF THE LITERATURE

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ABSTRACT

Coronary cameral fistulas (CCF) are rare, mostly congenital, entity where they involve abnormal termination of a coronary artery, usually the right coronary, into a cardiac chamber, usually the right ventricle. In adult most of CCFs are discovered incidentally during angiographic evaluation for coronary vascular disorder. In infants and children predominate clinical and lab signs of right to left shunt, anginal symptoms or rare, or signs of heart failure. We present a 3-months-old boy with CCF without clinical signs despite that echocardiography and selective coronary arteriography showed important flow through the fistula.

KEYWORDS: coronary arteries anomaly, congenital heart disease, coronary fistula.

INTRADUCTION

Coronary-cameral fistula (CAF) is an anomalous connection between a coronary artery and cardiac chamber. Symptoms associated with the lesion include those caused by volume overloading of the heart or by coronary artery steal secondary to the fistulous communication. Traditional imaging techniques to diagnose a CAF include echocardiography and catheter angiography.[1] However, echocardiography is operator dependent and limited by availability of a good acoustic window and it can presented dilated proximal part of coronary artery but it may not show the entire course of the fistula. Catheter angiography is invasive and overlap between a tortuous fistula and adjacent cardiovascular structures may hamper complete evaluation of the lesion.[2]

Case presentation

An absolutely health infant, aged 3 month, weighed 5.2-kilograms, from the normal pregnancy and normal delivery, with positive family history with congenital heart disease (the baby's cousin was detected with tetralogy of Fallot), during the routine paediatric examination systolic murmur was noted and for cardiologic examination at tertiary level was presented. There was no sign of cardiac failure. Physical examination revealed a short systolic murmur grade 3-4/6 in left, radiating to the right side and imitating the restrictive ventricular septal defect. The electrocardiogram was normal. A chest radiograph showed mild non typical cardiomegaly.

Transthoracic echocardiography demonstrated normal morphology and function of both ventricles, normal left and right outflow tract. Orifice of left coronary artery was normal but right orifice and presented proximal part of right coronary artery was large but not tortuous, measuring 6-7 mm in orifice and 5-6mm in distal presented part (Figure 1). During the systole with continuous flow on color Doppler, an unusual short turbulent flow near lateral wall of the right ventricle was presented with maximal systolic pressure 76mmHg.

A selective coronary angiography was performed and revealed normal left coronary artery and its branches in origin, course and calibre but injection in the right coronary artery (RCA) revealed a dilated tortuous right coronary artery draining into the right ventricle, near the lateral wall, involving mostly flow from the RCA.

We did follow-up sonographic examinations on the boy every 2 to 3 months. The progressive dilatation of the RCA was recorded. The size of the RCA at its opening from the aortic root was 6 mm at 3months of age and 8-9 mm at age of 9 months.

Catheter closure techniques with devices have been performed and at age of 11 months procedure successfully was done. There was excellent final angiographic result with obliteration of flow into the fistula. The patient was monitored three days and discharged home in stable condition.

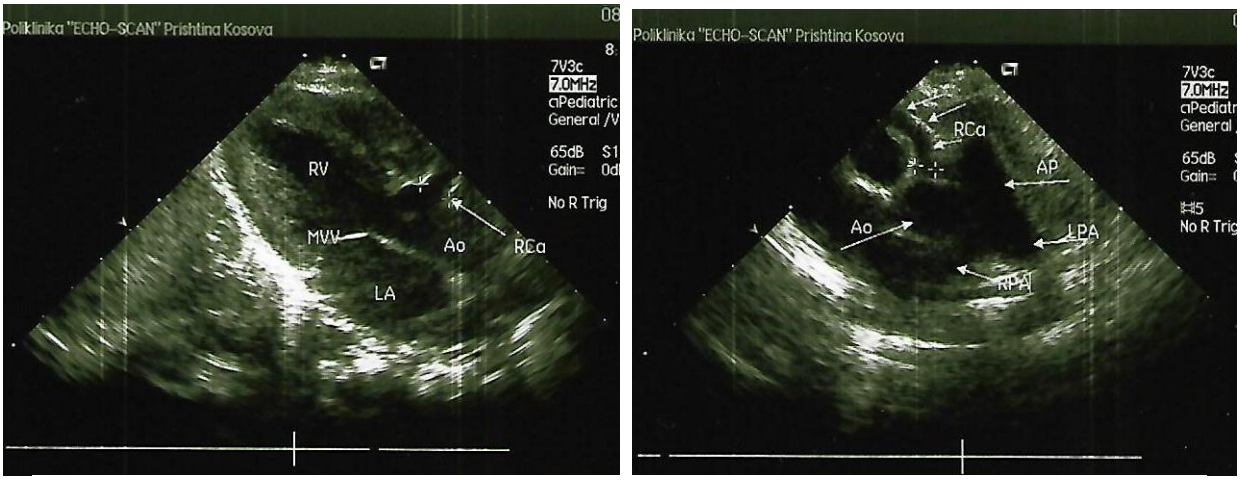

Figure 1. Transthoracic echocardiography presented dilated proximal part of the right coronary artery

DISCUSSION

A coronary artery fistulas are communications between one of the coronary arteries and a cardiac chamber or a major vessel (venae cavae, pulmonary artery, veins, or coronary sinus). Coronary artery-left ventricular fistulae are exceedingly rare with the incidence being reported as 1.2% of all CAFs.[1] Major sites of origin of fistula are the right coronary artery (55%), left coronary artery (35%) and both coronary arteries (5%). Major termination sites are the right ventricle (40%), right atrium (26%), pulmonary arteries (17%), and less frequently the superior vena cava or coronary sinus, and least often the left atrium and left ventricle.[3]

A coronary artery fistula can be identified prenatally by color and spectral Doppler imaging only when they are large. This anomaly should be sought in fetuses with outflow tract obstructive cardiac lesions and an intact interventricular septum. During embryonic development of the heart, some branches of the coronary arteries communicate with the intramyocardial trabecular spaces, which in turn connect to the ventricular lumen. In hypoplastic right or left hearts, the occurrence of a communication between a ventricular cavity and the coronary system is thus considered persistence of the embryonic microvascular pattern.[4]

Cardiac catheterization with coronary angiography remains the gold standard for the diagnosis of coronary artery fistula. It can demonstrate the size, anatomy, number, origination and termination site of the fistulas. Cardiac echocardiography is also useful for diagnosis. Magnetic resonance imaging and multidetector computed tomography are also used to evaluate the anatomy, flow and function of CCF.[5,6]

Clinical presentation will generally depend on the haemodynamic significance of the anomaly and most commonly coronary artery fistulae are asymptomatic and are found incidentally. Anginal symptoms may be the presenting feature, particularly in patients with multiple fistulae and in those patients with a single fistula, exertional dyspnoea is more likely to predominate. Inducible ischaemia has been well demonstrated in these patients and is thought to occur as a result of left to left shunting causing a coronary steal phenomenon and diastolic overload. The hemodynamic consequence of the coronary cameral fistula depends on the size of the fistula and the communicating chamber.[7] Most coronary artery fistulae are small and usually do not cause any ischemic symptoms and has excellent long-term prognosis.[3]

Coronary cameral fistulae may also cause myocardial infarction, congestive heart failure, arrhythmias and aneurysmal formation and rupture of affected vessels may also occur. The best way to manage cameral fistulae is uncertain largely due to the rarity of the condition. Patients in whom focal fistulae with large shunts exist may benefit from closure of the shunt and if this is to be performed it is probably best done as early as possible.[7]

Surgery has long been the accepted treatment of choice for fistula closure. Complications of surgery include myocardial infarction, arrhythmia, transient ischemic changes and stroke.[8]

Catheter closure techniques have been performed to treat coronary fistulas with devices, including detachable balloons, stainless steel coils, controlled-release coils, controlled-release patent ductus arteriosus (PDA) coils and Amplatzer PDA plug. The advantages of the transcatheter approach include less morbidity, lower cost, shorter recovery time and avoidance of thoracotomy and cardiopulmonary by-pass. Moreover, multiple coils may be needed to close large fistulas, which can increase fluoroscopy time, contrast load and chances of failure to occlude the fistula.[5]

Conflict of interest

We declare that we have no financial or personal relationships that may have inappropriately influenced the writing of this paper.

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