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Unilateral absence of left pulmonary artery (UAPA) with ventricular septal defect (VSD) presented on newborn

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Introduction: Unilateral absence of left pulmonary artery is very rare congenital heart defect, often associated with other congenital cardiovascular anomalies, such as Tetralogy of Fallot, VSD, Coarctation of the aorta, Right-sided aortic arch, Truncus arteriosus. Most of the cases are diagnosed in childhood, but in the absence of associated cardiac malformation, the condition may be asymptomatic until adult age. The incidence is about 1/200,000 in live birth.

Case presentation: Seven days old male baby was admitted to University Children's Hospital in Skopje, with clinical signs of tachi-dyspnea, hepatomegaly (sign of cardiac failure) and systolic murmur of 3/6 degree. Complete medical history, physical examinations and imaging examinations were made, which includes: 2D Doppler echocardiography, chest X-rays, cardiac catheterizations and CT angiography. His chest X-rays showed moderate cardiac enlargement and mild prominent pulmonary trunk. The ECG had non significant changes. Echocardiography showed dilated left atrium, dilated left and right ventricle, large ventricular septal defect (VSD) with huge left L-R shunt, dilated pulmonary trunk, right-sided aortic arch and absence of left pulmonary artery. Therefore, CT angiography was performed, where left pulmonary artery was missing and the vascularisation of the left lung was with collaterals from the thoracic aorta. Based on the above findings, a diagnosis of UAPA was made. The initial treatment was with ACE inhibitors and diuretics, he had cardiac surgery with VSD closure when he turned 5 months old. Today, at the age of 2, this toddler is in a good condition, follow up on a regular basis.

Conclusion: In the past 20 years, this is first case in Macedonia. Through UAPA malformation is a rare occurrence, it should be considered as a differential diagnosis when either heart failure or pulmonary hypertension is present. Treatment is unclear and is usually individualized based on symptoms for associated conditions.

Key words: Unilateral absence of left pulmonary artery, Ventricle septal defect, cardiac malformation