Anomalous origin of right coronary artery from the left coronary sinus in association with type A interrupted aortic arch

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June 22, 2022

Abstract

In this case, we reported a 24-day-old infant with a rare combination of the anomalous origin of coronary artery and the type A interrupted aortic arch, who presented with significant shortness of breath. Besides these anomalies, further cardiac examination including transthoracic echocardiography and CT angiogram revealed ventricular septum defect, patent foramen ovale and patent ductus arteriosus at the same time. Surgical procedures including repair of ventricular septum defect, ligation of patent ductus arteriosus, unroofing of right coronary artery and end-to-side aortic anastomosis were performed to cure the infant. This patient stayed in ICU for few days without any major complications and discharged from hospital at the tenth day after surgery.

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In this case, we reported a 24-day-old infant with a rare combination of the anomalous origin of coronary artery and the type A interrupted aortic arch, who presented with significant shortness of breath. Besides these anomalies, further cardiac examination including transthoracic echocardiography and CT angiogram revealed ventricular septum defect, patent foramen ovale and patent ductus arteriosus at the same time.

Surgical procedures including repair of ventricular septum defect, ligation of patent ductus arteriosus, unroofing of right coronary artery and end-to-side aortic anastomosis were performed to cure the infant. This patient stayed in ICU for few days without any major complications and discharged from hospital at the tenth day after surgery.

Keywords : Congenital heart disease

A 24-day-old male infant with persistent shortness of breath since birth was referred to hospital for further treatment. His mother did not have regular antenatal care, while pregnancy complications were either not found. Physical examination revealed the respiratory rate of 30 bpm and blood pressure of 80/40 mmHg. On presentation transthoracic echocardiography (TTE) revealed ventricular septum defect (VSD), patent foramen ovale (PFO) and patent ductus arteriosus (PDA). The aorta was discontinuous at the level of descending aortic arch, and the significantly enlarged arterial duct connected the descending aorta and the pulmonary artery. The right coronary artery originated from the left coronary sinus with an intramural segment could be found on TTE images (Figure 1, *panel A*). Contrast computed tomography angiogram (CTA) and three-dimensional reconstruction was carried out, which demonstrated the interrupted aortic arch (IAA) combined with anomalous aortic origin of a coronary artery (AAOCA). The site of aortic interruption was at the distal side of the left subclavian artery, and the blood flow to the descending aorta was via an enlarged PDA which diameter measured about 5mm (Figure 1, *panel B*). CTA also clearly revealed the right coronary artery originated from the left coronary sinus with a 3mm-length intramural segment (Figure 2,*panel C*). Besides, a subarterial ventricular septal defect with the diameter of approximate 8mm was found on imaging. Thus, the patient underwent procedure including repair of VSD, ligation of PDA, unroofing of right coronary artery and end-to-side aortic anastomosis which combined the descending aorta to the aortic arch. Postoperative TTE revealed no residual shunt at the level of the interventricular septum and the aorta. The boy recovered successfully and discharged from hospital at the tenth day after surgery.

AAOCA is the most commonly reported arterial malformation in cases of sudden death, which was usually caused by an intramural segment of coronary artery. AAOCA is traditionally defined as coronary artery originates from abnormal sinus including right, left or no coronary sinus. Due to the variable reported prevalence in different population, the true incidence remains unknown[1,2]. IAA is a relatively rare congenital anomaly with a incidence of 2/100000 approximately in live births, which is characterized by complete anatomic discontinuity between the ascending and descending aorta, while descending aorta usually receive flow form pulmonary artery via PDA[3]. IAA has been classified into three types (A, B and C) according to the site of aortic interruption. These two main anomalies combined with VSD, PFO and PDA conspired to cause the clinical presentation of the boy. Considering extremely severe symptoms and anatomical anomalies, surgical procedure was essential for this patient.

CONFLICT OF INTERESTS

All authors declare that there are no conflict of interests.

ETHICS STATEMENT

This manuscript and all of its content meet the ethical guidelines, including adherence to the legal requirements of the study country. The need for patient consent was waived.

FUNDING

Not appliable.

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Figure

Figure 1. ***Panel A*** , TEE revealed AAOCA with a significant intramural segment (red arrow). Panel B , three-dimensional reconstruction demonstrated the interruption was at distal side of the left subclavian artery, and the blood flow to the descending aorta was via an enlarged PDA (red arrow). Panel C , CTA clearly demonstrated that the right coronary artery originated from the left sinus (red arrow). (AO, aorta)