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Short Communication

Pulmonary Atresia with Bilateral Ductus Arteriosus: An Unusual Anatomy

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Abstract

Bilateral PDA is an infrequent entity and is seen in a wide range of presentations. The cases previously reported included bilateral PDA in isolation, associated with congenital heart disease including various forms of pulmonary atresia, aortic arch anomalies including aortic atresia or interrupted aortic arch, and isolation of one subclavian artery. Knowledge of detailed cardiac anatomy is a prerequisite for a successful surgery. For this, complementary imaging to echo cardiography such as computed tomography is required to delineate both the intra cardiac and extra cardiac anatomy fully.

Keywords: Bilateral ductus arteriosus; Pulmonary atresia; Computed tomography; Anatomy

Introduction

A 15-day-old female newborn that began a few hours earlier with breathlessness, inability to feed, cyanosis, and marked intercostal recession was referred to the pediatric cardiology department. The patient was born at the 37th week with a birth weight of 3100 gr. On arrival baby was under an oxygen hood, Dopamine, and Dobutamine at 10 mcg/kg/min. On physical examination, The cyanotic infant with a grade 3/6 ejection systolic murmur and oxygen saturation level of about 65% was noted to have decreased pulmonary vascular markings on chest X-ray and left axis deviation on EKG. Following prostaglandin E1 infusion, oxygen saturations of the patient reached 80%.

Transthoracic echocardiography revealed the Tetralogy of Fallot, including subaortic ventricular septal defect and pulmonary atresia and Patent Ductus Arteriosus (PDA). Computed Tomography (CT) was planned for imaging the pulmonary arteries and blood supplies. Following the evaluation of CT images, the first duct could be seen originating from the innominate artery and supplying the right pulmonary artery (Figure 1A and 1C). The second duct was originating from the arcus aorta and continues as the left pulmonary artery (Figure 1B and 1D). The left pulmonary artery has not continued with the main pulmonary artery, but the right. The main pulmonary artery was also hypoplastic. Interventional team discuss the case and proposed stenting both ducts. During the procedure, due to angle and tortoise course, it became clear that the duct's stenting was not a viable option. The patient was referred for surgery. Written informed consent was obtained from the patient's parents.

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At surgery, the mediastinum was approached through a median sternotomy incision. The pericardium opened longitudinally, and the dissection of the pulmonary artery and duct tissues was started. Aortic right atrial cardiopulmonary bypass was commenced, and bilateral ducts were ligated. All ductal tissue was removed from the pulmonary arteries. After excising the fragile ductal tissue, a gap was encountered on the right pulmonary artery, and the left pulmonary artery was positioned to the left away from the pulmonary confluence. The hypoplastic main pulmonary artery is divided and anastomosed end-to-end fashion with the left pulmonary artery. A 4 mm PTFE shunt was reconstructed between the brachiocephalic artery and the right pulmonary artery. The cardiopulmonary bypass ended, and the procedure was finished as routinely (Figure 2).

Bilateral PDA is a very rare entity and is seen a wide range of presentations [1,2]. Bilateral PDA occurs mostly with associated congenital heart disease such as Tetralogy of Fallot or anomalies of

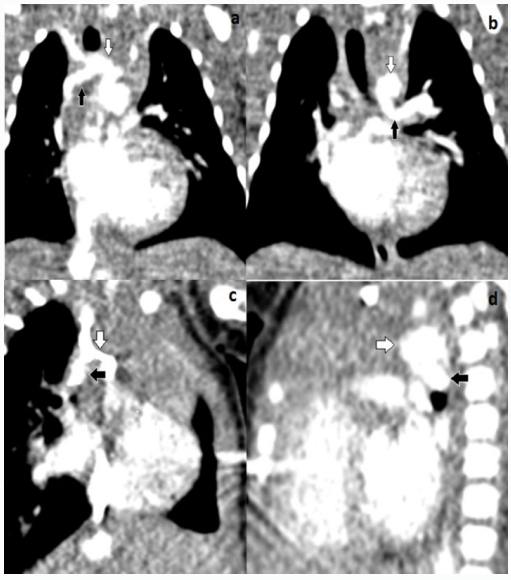

Figure 1: Bilateral Patent Ductus Arteriosus (PDA). CT angiogram (1a, 1c) shows right PDA (black arrow) originating from the right brachiocephalic trunk (white arrow). CT angiogram (1b, 1d) shows left PDA (black arrow) originating from the arcus aorta (white arrow).

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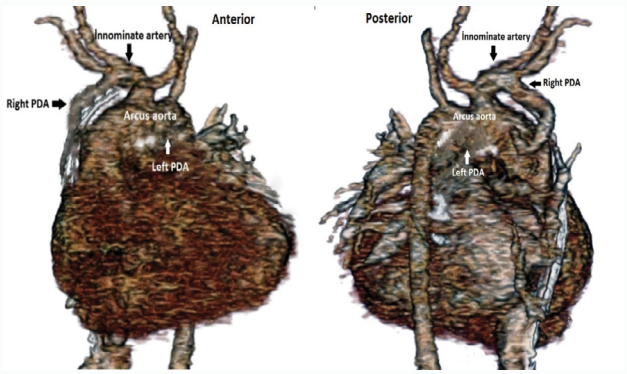

Figure 2: Three-dimensional reconstruction of the aortic arch demonstrates bilateral ductus arteriosus originating from the arcus aorta.

visceral lateralization [2]. Another phenomenon is discontinuous pulmonary arteries with supply from either bilateral duct or aortopulmonary collaterals [2,3]. Current literature includes mainly case reports or small case series of PDA. The cases previously reported included bilateral PDA in isolation, associated with congenital heart disease including various forms of pulmonary atresia, aortic arch anomalies including aortic atresia or interrupted aortic arch, and isolation of one subclavian artery [3-5].

The broad treatment options for bilateral PDA are present such as transcatheter interventions (MAPCA coil embolization, stenting of bilateral duct) and surgical palliation (aortopulmonary shunt and PDA ligation, single or bilateral)) or complete repair [3,5]. The surgical management of this complex anomaly is variable according to arborization of a pulmonary artery, amount of pulmonary blood flow, morphology, size, course of the native pulmonary arteries, and MAPCAs. The presence of pulmonary hypoplasia and MAPCAs, and a single ventricle is vital for surgical decision-make. Thus, a patient without hypoplastic Pulmonary Arteries (PAs) and arborization defects are suitable for one stage of total correction. However, if they have hypoplastic PAs, the approach should be a shunt operation at the first stage before the total correction procedure, as like in our case.

Conclusion

The knowledge of detailed cardiac anatomy is a prerequisite for a successful surgery. For this, complementary imaging to echocardiography such as computed tomography is required to delineate both the intracardiac and extracardiac anatomy fully.

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