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Case report

Rare case of bilateral ductus with confluent pulmonary arteries in case of d-TGA with intact ventricular septum

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ABSTRACT

Bilateral ductus is a very rare abnormality. Here we present a rare case in which ductus was associated with confluent arteries and dextro-Transposition of the Great Arteries. The probable theory of its origin is also discussed with help of Edward's hypothetical double aortic arch system.

Introduction

Bilateral ductus is a very rare abnormality. It is usually associated with other congenital anomalies. Bilateral ductus is frequently associated with anomalies of pulmonary arteries or arch anomalies. This is a rare case in which ductus was associated with confluent arteries and dextro-Transposition of the Great Arteries. The hypothetical reason of their origin has also been discussed.

Case details

14 day old male child with dextro-Transposition of the Great Arteries (d-TGA), intact ventricular septum was taken for Arterial switch operation. Pre op echocardiography showed dextro-transposition of great arteries with intact ventricular septum and left sided patent ductus arteriosus. Intra-operatively, there was d-TGA with aorta and main pulmonary artery related antero-posteriorly, aorta being anterior to pulmonary artery. There was left aortic arch. Pulmonary arteries were confluent and good sized. Bilateral ductus arteriosus were present. Right ductus was connected to right

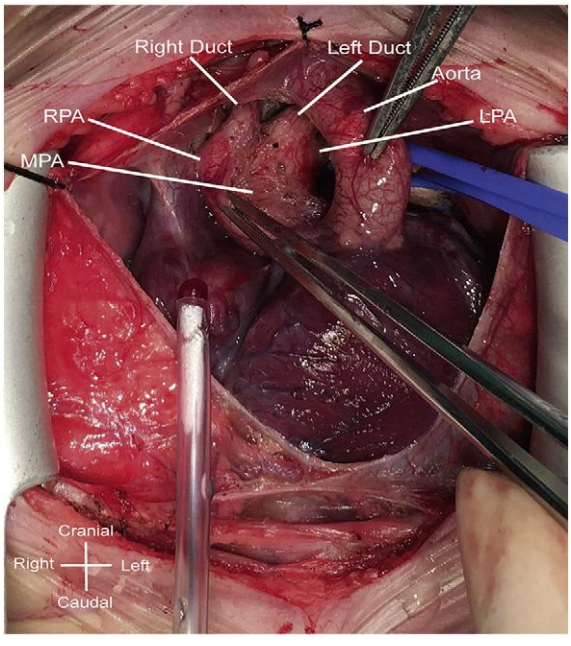

Fig. 1. Intra-operative photograph showing bilateral ductus. MPA is retracted to right and aorta to left with forceps for better view. RPA looped with blue tape. MPA: Main pulmonary artery, LPA: Left pulmonary artery, RPA: Right pulmonary artery.

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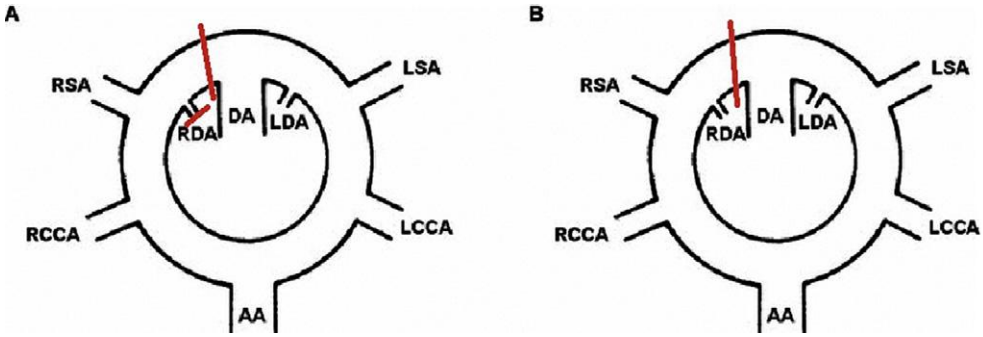

Fig. 2. A: Edward's hypothetical double aortic arch with bilateral PDA. Red lines show normal points of discontinuation of arch components. B: red line shows point of disconnection of arch component in present case. Notice there is no disconnection of right ductus with normal disconnection of right sided arch.

pulmonary artery and right brachicephalic artery. Left ductus was connected to left pulmonary artery and descending thoracic aorta (Fig. 1). There was no vascular ring. After completion of dissection required for arterial switch operation, cardiopulmonary bypass was instituted between aorta and right atrium cannulae. Soon after going on bypass both ducts were ligated and divided. Rest of the surgery was performed routinely. Intra- and post-operative course was uneventful.

Discussion

A persistent bilateral PDA is an uncommon abnormality which occurs during the development of the aortic arch and the pulmonary arteries. It is most commonly seen accompanied by abnormalities of pulmonary valve, pulmonary arteries and aortic arch anomalies.[1,2] Freedom et al. in their study of 27 cases with bilateral PDA reported associated anomalies of pulmonary arteries and aortic arch in all cases.[2] In the present case bilateral PDA were associated with confluent pulmonary arteries and no associated aortic arch anomalies. However bilateral PDA is commonly associated with co-existing intra-cardiac anomalies. Its association with dextro-transposition of great arteries has been reported only once.[3] In that case there was bilateral PDA with dextro-transposition of great arteries associated with right aortic arch. Left ductus was arising from left subclavian artery forming a complete vascular ring. The present is rare because it's first of its kind in which bilateral PDA associated with dextro-transposition of great arteries with left aortic arch with normal pulmonary arteries without anomalies of aortic arch.

Extrapolating from the primitive pharyngeal arch system, there is a tremendous range of potential anomalies of the aortic arch and its branches. With help of Edward's hypothetical double aortic arch (Fig. 2) it is easy to understand origin of bilateral PDA. Normally the segment of right aortic arch and right ductus disappears forming left aortic arch with left sided ductus. But if right ductus persists with only disappearance of right aortic arch then the present anomaly of bilateral PDA with left aortic arch arises.

Bilateral PDA is readily demonstrated on echocardiography. However, because of frequent association of anomalies of pulmonary arteries and aortic arch, some other imaging modality like CT of MRI is necessary.[4] In the present case it was missed on pre-operative echocardiography. With increasing use of fetal echocardiography it can be diagnosed in utero.[5]

The treatment mainly depends on associated anomaly of pulmonary artery, aortic arch and intra-cardiac anomaly. Surgical management frequently requires extensive reconstruction of great vessels. The treatment in present case was very straight forward. Bilateral PDA were ligated and divided and arterial switch operation was done. Absence of associated anomalies of great vessels reduced complexity of procedure.

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