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Case report

An Unusual Coronary Artery Complication after Surgery for a Complex Congenital Heart Disease: A Case Report

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Abstract

Background: Unexpected events in cardiac surgery may increase morbidity and mortality, which may be more complicated and difficult to manage especially in redo complex congenital heart surgery in children. This report presents a case of unusual coronary artery complication after redo surgery for a complex congenital heart disease. ***Case Presentation:*** A female baby with ventricular septal defect, transposition of great arteries and coarctation of the aorta, underwent coarctation repair and pulmonary banding in neonatal period then at one year old, she underwent arterial switch operation and ventricular septal defect repair. In the pediatric cardiac intensive care unit, she required high doses of vasoactive inotropic drugs and echocardiography revealed sever right ventricular dysfunction without significant gradients or shunts. Accordingly, a diagnostic cardiac catheterization was performed and the aortic angiography revealed that the right coronary artery was not inadvertently transferred into the neo -aortic root during the step of arterial switch operation. The surgeon declared that he came upon confounding factors during the intraoperative assessment, which led him to identify a single coronary artery. Immediate reoperation performed to translocate the non-transferred right coronary artery into the neo aortic root and postoperatively the case required extracorporeal membrane oxygenation till she stabilized, and she was discharged in an acceptable condition. ***Conclusion:*** Early diagnosis and management of any complications related to coronary artery during pediatric cardiac surgery could lower risk and achieve favorable outcome.

Keywords: ***Unusual; Coronary artery; Complication; Congenital heart Surgery.***

Background

Uncommon anomalies or complications related to coronary artery increase risk of cardiac surgery. Successful transfer of the coronary arteries into the neo-aortic root represents the key point of arterial switch operation (ASO) and the related results are constantly improving. Elassal [1] reported a successful technique to achieve arterial-level repair of transposition of great arteries (TGA) without coronary artery transfer due to the too small ascending aorta, which precluded harvesting of coronary buttons. He aimed to achieve biventricular repair for a complex case presented with TGA combined with hypoplastic aorta, interrupted aortic arch and ventricular septal defect (VSD) [1]. Nevertheless, there is still a risk of early and late coronary complications, particularly in patients with unusual coronary patterns or if the ASO is combined with other surgical steps to correct a complex congenital anomaly [2-4]. This report presents a case of successful management of an unexpected coronary artery complication after redo surgery for a complex congenital heart disease.

Case presentation

A one-year-old baby had ventricular septal defect (VSD), transposition of great arteries (TGA), and coarctation of the aorta (COA) (**Fig: 1**).

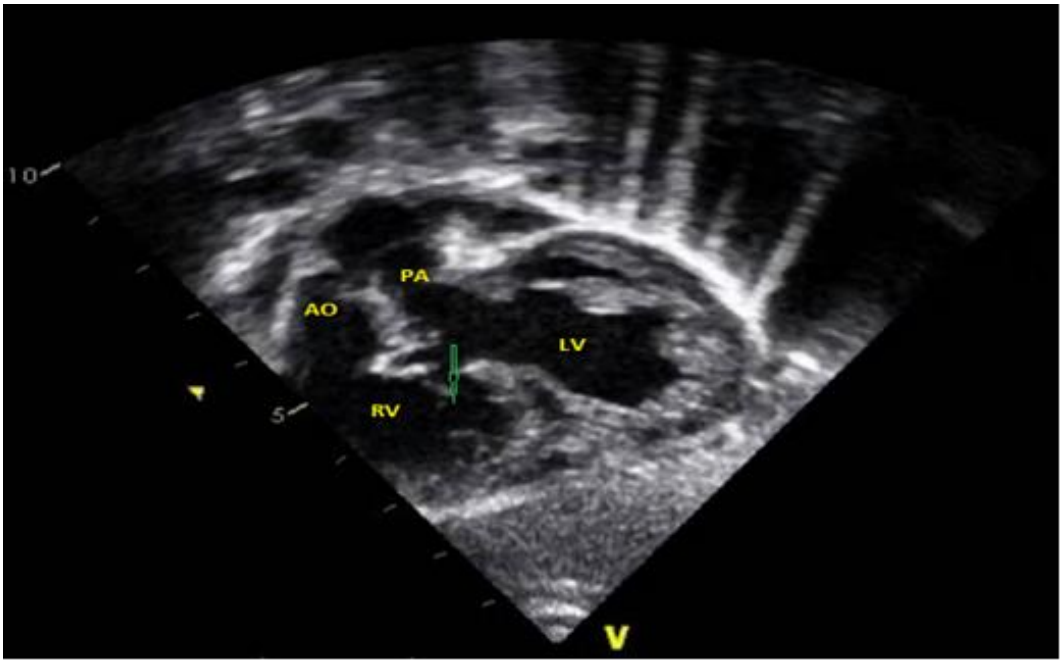

Figure 1: Preoperative echocardiography before the first redo surgery

It revealed transposition of great arteries (TGA) and ventricular septal defect (VSD). **AO:** Aorta, **PA:** Pulmonary artery, **LV:** Left ventricle, **RV:** Right ventricle, **VSD:** Green arrow.

The patient underwent pulmonary artery (PA) banding and CAO repair in the neonatal period. The pediatric cardiology and cardiac surgery team agreed that the echocardiographic data is sufficient, as a diagnostic tool, to perform a complete corrective surgery for the patient at that age. The surgical consent was obtained as a high-risk redo complex surgery and the Risk Adjustment for Congenital Heart Surgery -1 (RACHS-1) was scored as 4 [5]. First redo surgery was performed in the sequence of VSD closure, pulmonary artery disbanding, and arterial switch operation (ASO). The patient transferred to pediatric cardiac intensive care unit (PCICU) with chest closed and moderate doses of vasoactive inotropic drugs (VIDs) due to borderline hemodynamic state. After 8 hours of PCICU admission, the patient became hypotensive despite escalading the doses of VIDs. The electrocardiogram (ECG) revealed dysrhythmias with ST segment changes despite rhythm controlling drugs. Lactate accumulated and urine output diminished. Echocardiography (ECHO) showed sever right ventricular (RV) dysfunction, fair left ventricular (LV) function and no significant gradients or residual lesions. Accordingly, the decision was to prompt the case for a diagnostic cardiac catheterization. Aortic angiography revealed a missed non-transferred right coronary artery (RCA) and only the single coronary artery was transferred into the neo aortic root during ASO step (**Fig: 2**).

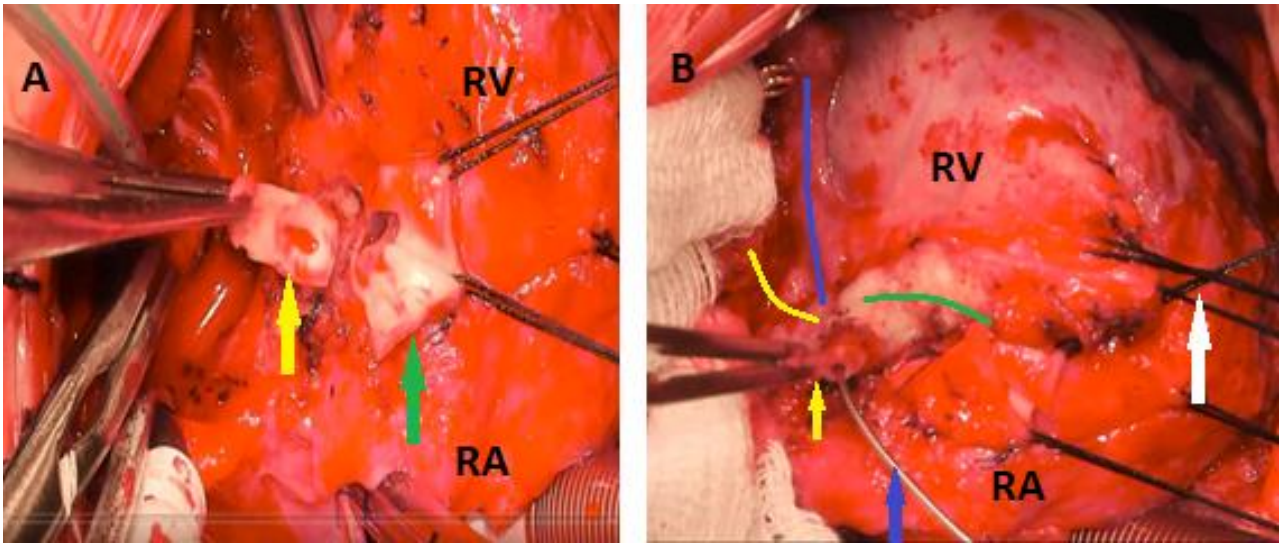

Figure 2: Intraoperative images from surgeon view of the first redo surgery:

A: Harvested button of the identified single left coronary artery (yellow arrow) and exposure of the aortic root coming from the RV (green arrow). **B:** Harvested button of the identified single left coronary artery (yellow arrow), probing through the identified single left coronary artery (blue arrow), the prob passed through trifurcated arteries [left anterior descending artery (LAD) (imaginary epicardial blue line), left circumflex artery (LCX) (imaginary epicardial yellow line) and conus branch misdiagnosed as right coronary artery (RCA) (imaginary epicardial green line)], and silk stitch suspensions retracted the aortic root (white arrow). **RV:** Right ventricle, **RA:** Right atrium.

The surgeon reviewed the intraoperative videos of the ASO steps to find out why the RCA was not identified. The surgeon explained that he encountered confounding factors during the intraoperative assessment, which led him to identify a single coronary artery. Those confounders were the extensive adhesions and the right coronary orifice was too small to be seen. Another important confounding factor was that the surgeon probed the identified single left coronary artery that revealed trifurcated coronary pathways that were interpreted as left anterior descending artery, left circumflex artery, and conus branch misdiagnosed as right coronary artery (**Fig: 3**).

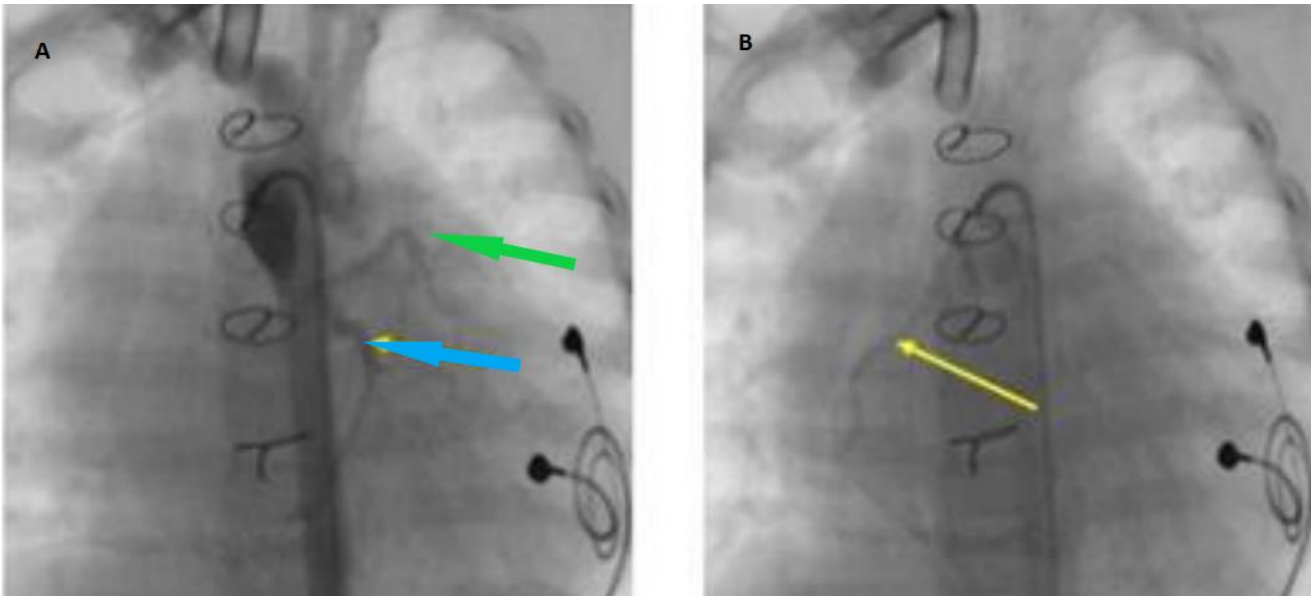

Figure 3: Neo-aortic angiography after arterial switch operation (ASO) during the first redo surgery:

A: Early antegrade filling of transferred left coronary artery bifurcated into left anterior descending artery (LAD) (green arrow) and left circumflex artery (LCX) (blue arrow), **B:** Missed non-transferred right coronary artery (RCA) (yellow arrow) appeared later by retrograde filling.

Hence, a single coronary button reimplantation was performed. After discussion with the team and the family was consented, the surgeon took the case urgently from the Cath Lab to the operating room to translocate the non-transferred RCA into the neo aortic root. Second redo surgery was performed on conventional cardiopulmonary bypass (CPB) through neo-aortic cannulation and bicaval cannulation. Aortic cross clamp was applied and after the heart arrested by antegrade cold blood-based Del Nido cardioplegia, the neo- pulmonary artery was incised and the non-transferred RCA was identified and probed to confirm its course then it was harvested from the neo- pulmonary artery (**Fig: 4**).

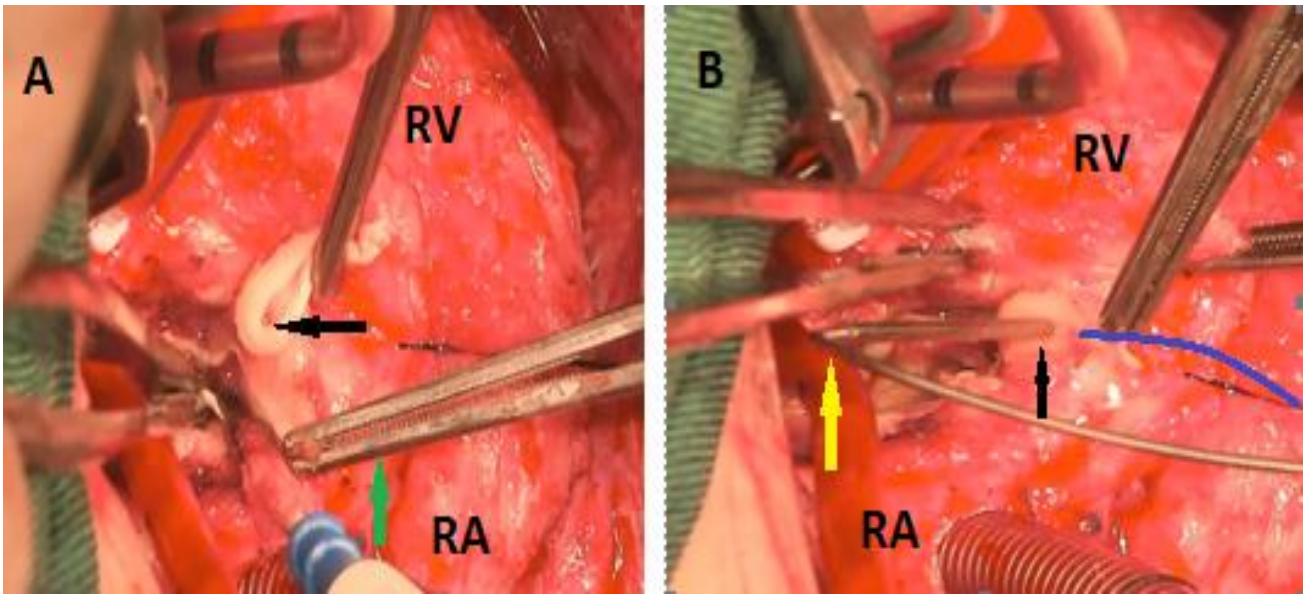

Figure 4: Intraoperative images from surgeon view of the second redo surgery:

A: Harvested button of the missed non-transferred right coronary artery (RCA) (black arrow) and atraumatic forceps grasping the edge of the anterior wall of the neo-pulmonary artery (green arrow). **B:** Harvested button of the missed non-transferred right coronary artery (RCA) (black arrow), probing through the missed non-transferred right coronary artery (RCA) (yellow arrow), and the prob passed through its usual course (imaginary epicardial blue line). **RV:** Right ventricle, **RA:** Right atrium.

The defect in the neo- pulmonary artery was repaired with equine pericardial patch. The neo-aorta was incised and a small round aortotomy hole was done. The RCA was successfully anastomosed to the neo-aortic root. After the aorta declamped, the heart regained sinus rhythm but the weaning from the CPB was very difficult due to hypotension despite maximum doses of VIDs, so connecting the patient to the extracorporeal membrane oxygenation (ECMO) was a reasonable decision. Cross clamp time recorded 35 minutes and conventional CPB time recorded 50 minutes. Patient admitted to the PCICU in a critical condition on moderate doses of VIDs with the chest was left open and ECMO support connected. After 5 days of second admission into the PCICU, the ECMO was successfully disconnected and the chest was closed after 12 days from disconnection of ECMO. After 26 days, the case was extubated and on the 31st day she was transferred to the pediatric cardiology ward in a fair clinical condition. Hospital discharge was on day 57 of the date of first hospital admission with an acceptable clinical outcome on mild doses of captopril and furosemide.

Discussion

The most critical and highly sensitive step in ASO is coronary artery transfer and reimplantation. It has been not only an intraoperative challenging element, but also a major predictor of morbidity and mortality [6,7]. Compromised myocardial perfusion through the reimplanted coronary arteries was early recognized as an important predictor of surgical outcome after correction of TGA, especially when complex coronary artery anatomy was present or the ASO step was combined with other surgical steps in one stage corrective operation [8]. Technical difficulty in translocating is well identified in uncommon coronary artery patterns like single, intramural, double orifice originating from the same sinus of Valsalva and inverted coronary arteries [9,10]. Additionally, performing ASO in combination with other surgical steps to repair complex congenital heart lesions increases the challenge of coronary harvesting and translocation specially in redo surgeries [1-7]. A single common coronary ostium was present in 16 patients (2.2%) in one cohort study [11], none of whom experienced early or late death. In our patient with redo surgery for arterial switch after PA band and CAO repair, visual identification of the RCA was missed and the patient was considered as having a single coronary ostium. The stormy sequelae raised the importance of identifying the coronary anatomy before arterial switch particularly in redo surgery and complex congenital heart surgery. Such redo surgery and complexity resulted in anatomical distortion, which made visual identification of coronary arteries intra-operatively a difficult mission. Cardiac catherization with aortic aortography helped to diagnosis this uncommon coronary artery complication and immediate surgical management succeeded to avoid catastrophic outcome.

Conclusions

While early management of common anomalies and complications related to coronary artery may decrease risk of cardiac surgery, unusual coronary artery events during congenital heart surgery need immediate capture of diagnosis and rapid management to avoid life-threatening complications. Surgery that needs coronary artery translocation in children is still at risk of terrible outcomes related to coronary artery. Aortic angiography confirmed the diagnosis. Early surgery plays a major and effective role to treat them. Success to address these unexpected scenarios is a real gauge to assess the efficacy of multidisciplinary coordination between pediatric cardiology and cardiac surgery services.

List of abbreviations

ASO: Arterial switch operation.
TGA: Transposition of great arteries.
VSD: Ventricular septal defect.
VIDs: Vasoactive inotropic drugs
DORV: Double outlet right ventricle.
COA: Coarctation of the aorta.
RACHS-1: Risk Adjustment for Congenital Heart Surgery -1.
PCICU: Pediatric cardiac intensive care unit.
ECG: Electrocardiogram.
ECHO: Echocardiography.
CPB: Cardiopulmonary bypass.
RCA: Right coronary artery.
ECMO: Extracorporeal membrane oxygenation.
PA: Main pulmonary artery.

Declarations

Ethics approval and consent to participate: This study has been approved by Ethics Committee (EC) and the consent of patients obtained.

Consent for publication

Not applicable.

Availability of data and materials

All data generated or analyzed during this study are included in this published article

Competing interests

There are no financial competing interests.

Funding

The research has no funding.

Authors' contributions

The author analyzed and interpreted the patient data and is the person who edited the manuscript. The manuscript has not previously been published in print or electronic form and is not under consideration by any other publication. The author has read and approved the submission of the manuscript to The Cardiothoracic Surgeon journal. There is no ethical problem or conflict of interest.

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