Original Research

OUTCOMES OF ARTERIAL SWITCH OPERATION IN PATIENTS WITH INTRAMURAL CORONARY ARTERIES AT NATIONAL CHILDREN'S HOSPITAL

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ABSTRACT

Objectives: *To evaluate the outcomes of arterial switch operation (ASO) in a subgroup of patients who had intramural coronary arteries at National Children's Hospital.*

Methods: *From 2010 to 2016, 304 patients underwent ASO at National Children's Hospital. In which, eighteen (5.9%) had intramural coronary arteries. These medical records of these patients were collected and retrospectively reviewed.*

Results: *There were 10 male and 8 female in this group of patients. Patients with intramural coronary arteries had transposition of the great arteries (83.3%, n = 15) or Taussig - Bing anomaly (16.7%, n = 3). At the time of surgery, mean age was 69.5 ± 81.5 [11 - 321] days and mean weight was 3.9 ± 1.1 [2.5 - 6.3] kg. Mean bypass time and mean aortic cross - clamped time were 235 ± 90 [168 - 564] minutes and 149 ± 29 [100 - 255] minutes respectively. There were 3 deaths (16.7%): two hospital deaths (at 6 days and 26 days after ASO), one death after discharge (2 months later). One patient lost follow - up. Other 14 patients are in a good health status after discharge and free of re - intervention or reoperation related to the total correction with the mean follow - up time of 68.0 ± 38.5 [2 - 113] months.*

Conclusions: *Intramural coronary arteries remain a rare coronary anatomic variant following ASO. The outcomes of ASO in this subgroup of patients is quite favorable. Long - term follow - up is necessary.*

Keywords: *Arterial Switch Operation (ASO), Taussig - Bing Anomaly, Intramural Coronary Artery.*

I. BACKGROUND

The first Arterial Switch Operation (ASO)was performed by Jatene [1] in 1975, since then, the ASO has become the standard surgery for patients with transposition of the great arteries (TGA) andTaussig - Bing heart anomaly [2]. Anatomical variants of coronary artery in which the coronary artery runs within the aortic wall present a challenge for congenital heart surgeons, thus increases surgical complexity, and that is a risk factor for mortality due to complications of coronary artery disease after surgery [3]. From 2010 to 2016, 304 patients underwent ASO at Vietnam National Children's Hospital. In which, 18 patients (5.9%) had intramural course of the coronary artery. A retrospective study was conducted to describe the outcomes after ASO in this group of 18 patients.

II. METHODS

2.1. Subjects

The study included 18 patients (10 male, 8 female) underwent ASO with intramural coronary artery at Heart Center - Vietnam National Children's Hospital from 2010 to 2016.

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2.2. Methods

This is a retrospective, descriptive study with convenience sampling. Patient condition was confirmed by the contact with the family of patient via telephone and patient's examination was checked with hospital computer data or paper medical records at the last visit follow up. Early postoperative mortality was defined as death within 30 days after surgery or before the time the patient was discharged from the hospital after surgery. Patients are followed yearly at the out-patient clinic with regular echocardiography, electrocardiogram and chest X - ray.

The arterial switch operation was conducted with standard cardiopulmonary bypass, bicaval cannulation, and moderate hypothermia (28^{o}C). After cardioplegia administration, the aorta was transected above the commissure 5mm, and the coronary button was harvested in a single disque. The roof of the intramural coronary artery was removed to create an unobstructed coronary ostium. Consequently, the common coronary button could be divided by two individual coronary buttons. The pulmonary trunk was transected, and the Lecompte maneuver was applied to bring the pulmonary bifurcation anteriorly to the ascending aorta. The neo - aorta was reconstructed using 8.0 polypropylene suture, and the two individual coronary buttons were reimplanted to the neo - aorta with modified trap-door technique. The neo-pulmonary was reconstructed with fresh autologous pericardial patch. The intracardiac defectswere closed and a small pattern foramen oval are usually maintained to decompress the left heart if needed.

Data were collected and analyzed using SPSS 20.0 software.Continuous variables were represented by mean, standard deviation, maximum and minimum values. Categorical variables were represented by percentage values. The study was approved by the Ethics committee of the Research Institute of Children's Health, National Children's Hospital. The patient inform consent has been waived due to retrospective study.

III. RESULTS

Before the surgery, all patients had cyanosis, 66.7% of the patient needed artificial ventilation, 55.5% needed balloon atrioseptostomy, and 33.3% needed Prostaglandin E1 infusion. The patient characteristics are shown in **Table 1**.

Table 1: Patient characteristics

Patient characteristics	n (%) / X ± SD [min - max]
Gender	
Male	10 (55.5)
Female	8 (44.5)
Pre-op ventilation	12 (66.7)
PGE1 transfusion	6 (33.3)
Raskind (BAS)	10 (55.5)
LV training	1 (5.5)
Mean age (days)	69.5 ± 81.5 [11 - 321]
Mean weight (kg)	3.9 ± 1.1 [2.5 - 6.3]
TGA subgroup	
TGA - IVS	8 (44.5)
TGA - VSD	7 (33.8)
Taussig - Bing	2 (11.2)
Taussig - Bing CoA	1 (5.5)
Coronary patterns	
2RLCx	10 (55.6)
2RCxL	7 (38.9)
1LRCx	1 (5.5)

LV: left ventricle, BAS: balloon atrial septostomie, IVS: intact ventricular septum, VSD: ventricular septal defect, CoA: coarctation of the aorta.

The mean bypass time and aortic cross - clamped time were 235 minutes, 149 minutes, respectively. The intramural coronary was mostly origin from the right coronary sinus. There were 7 patients (38.9%) who were left chest open after the surgery. Postoperative complications were noticed: 1 patient with phrenic nerve palsy requiring diaphragmatic plication (5.5%), infectious complications: wound infection in 1 patient (5.5%); respiratory infection in 2 patients (11.1%), sepsis in 2 patients (11.1%). The perioperative characteristics are described in **Table 2**.

Table 2: Perioperative characteristics

Perioperative characteristics	n (%) / X ± SD [min - max]
Bypass time	235 ± 90 [168 - 564]
Aortic cross clamp time	149 ± 29 [100 - 255]
Ventilation time	89.0 ± 43.5 [44 - 240]
Concomitant surgeries	
VSD closure	10 (55.5)
ASD closure	17 (94.5)
Aortic arch repair	1 (5.5)
PA bifurcation shifted to the left	3 (16.5)
Delayed chest closure	7 (38.9)
Respiratory infection	2 (11.1)
Wound infection	1 (5.5)
Sepsis	2 (11.1)
Peritoneal dialysis	3 (16.5)
Sinus bradycardia	2 (11.1)
Diaphragmatic palsy	1 (5.5)
Early death	2 (11.1)

ASD: atrial septal defect, PA: pulmonary artery

2 patients died during the postoperative hospital stay due to nosocomial infection, and 1 late death at 2 months after arterial switch operation. The remaining 16 patients had a mean mechanical ventilation time of 89 hours **(Table 3)**. The mean postoperative hospital stay was 21 days.

Table 3: Patient death information.

Name/Gender/ Age/Weight	Diagnosis	Cause of death
Tr.QK/Male/19 days/2.8kg	Taussig Bing - CoA	Nosocomial pneumonia, sepsis. Mortality at POD 6.
Le.HH/Male/ 21d/4.2kg	TGA - VSD	Extubation at POD 4, and re-intubation at POD 6 due to nosocomial pneumonia. Mortality at POD 26.
Tr.TP/Female/ 60days/2.5kg	TGA - IVS	PA band and BT shunt for LV training 14 days before ASO. Extubationat POD 4, discharge at POD 68. Mortalityat 2 months after discharge due to unknown reason.

POD: post-operative days number…,
ASO: arterial switch operation.

The Kaplan - Meier methods shown the estimated survival at 1 year, 5 years, and 9 years follow up are 83.3%., 83.3%, and 83.3%, respectively **(Figure 1)**.

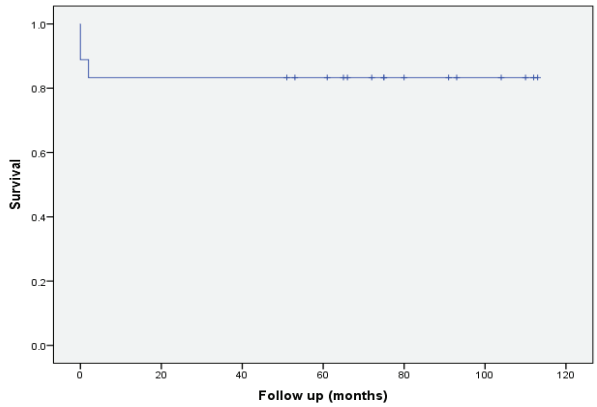

Figure 1: Kaplan - Meier curves estimated survival after surgery.

Except 1 late death and 1 patient loss follow up, the remaining 14 patients are well at the last follow up and the echocardiography show normal cardiac function without residual intracardiac defect. There are no reoperation or reintervention during follow up in this cohort.

IV. DISCUSSION

Intramural coronary arteryrefers to a condition in which the coronary artery lies in the media layer without a clear separation from the adventitia layer of the aorta. The frequency of this abnormality observed in patients underwent ASO varies from 1.7 to 9% [2,4]. During a period of 7 years, 304 patients underwent ASO surgery at Vietnam National Children's Hospital. The prevalence of intramural coronary artery was found in 5.9% of cases (18pts) indicating also that this abnormality is very uncommon. Therefore, a congenital heart surgeon would not frequently see that lesion. A meta - analysis by Pasquali et al [3] reported that the presence of an intramural coronary artery increases more than a six - fold in mortality compared to the usual coronary pattern. The intramural coronary artery obviously remains a challenge for pediatric cardiac surgeons and demands a skillful hand to manage the lesion for better outcomes.

The outcomes of ASO in the presence ofintramural coronary arteries are highly variable between reports from different centers. A report from the Royal Children's Hospital Melbourne - Australia [5] showed that 28 (3.9%) of 720 patients undergoing ASOhad intramural coronary arteries without postoperative mortality. Another report from Boston Children's Hospital - USA [6] showed that 4 patients (1.7%) had this coronary abnormality out of 223 patients undergoing ASO, of which 1 patient (25%) died. The authors from Necker Children's Hospital - France [2] reported 46 patients (5%) with intramural coronary artery, out of 919 patients undergoing ASO. Among this subgroup, 13 patients (28%) diedpostoperatively. The authors from Pusan University Hospital - Korea [4] reported that in 14 patients (8.9%) with coronary artery running in the aortic wall, out of a total of 158 patients undergoing ASO, there was 1 patient(7.1%) died after surgery.

There were some recommendations using an autologous pericardial patch to create a pouch of coronary artery without divided the single coronary button by two individuals [2,4]. In this study group of 18 patients, all of the intramural course of the coronary artery was un - roofted to ensure an un-obstructed flow to the coronary artery, more common was the left coronary artery. We were successful divided the single coronary button by two individual coronary buttons in all case, and each coronary button was easily reimplanted to the neo - aorta. According to the relation of great arteries, despite their discrepancy in size in some cases, the coronary transfer technique varied also which were trap - door technique, simple incision, or pinch - hole. By using the technique we described, we have no coronary hypoperfusion in this specific group. The bypass time and cross - clamped times remained long, but the early outcomes of this subgroup are quite good. There were 2 patients (11.1%) died during the hospital stay (6 and 26 days after surgery) due to sepsis and nosocomial pneumonia. 1 patient died 2 months after discharge with unknown reason. Overall survival rate was 83.3%. During the follow - up period, 1 patient was lost contact, the remaining 14 patients all gained weight well, still went to the doctor regularly and did not have to re - intervention or re - operation with an average follow - up time of 68.0 ± 38.5 [2 - 113] months.

V. CONCLUSIONS

From theresults above, the presence of intramural coronary artery is uncommon in patients undergoing arterial switch operation. The outcomes of ASO in this subgroup of patients is quite favorable. Long - term follow - up is necessary.

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