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Aortopulmonary collaterals in neonates with d-transposition of the great arteries – Clinical significance early after arterial switch operation☆

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ABSTRACT

Objectives: Purpose of this study is to evaluate the clinical significance of major aortopulmonary collateral arteries (MAPCAs) during the early postoperative course after arterial switch operation (ASO) in d-transposition of the great arteries (dTGA).
Methods: Clinical data of 98 patients with simple dTGA between January 2007 and December 2016 at the University Children's Hospital Zurich, Switzerland were analyzed retrospectively.
Results: 37 from 98 patients (38%) required cardiac catheterization (CC) due to an early complicated postoperative course or difficult coronary transfer due to special coronary anatomy. In 15 (15%) patients, hemodynamically relevant MAPCAs were found during CC and coil embolization was performed. Patients with relevant MAPCAs had a significantly longer PICU stay (7 versus 6 days, $p = 0.021$), longer hospital stay (41 versus 27 days, $p = 0.005$), longer mechanical ventilation time (5 versus 3 days, $p = 0.005$), longer need for inotropic support (5 versus 4 days, $p = 0.001$) and delayed chest closure time (3 versus 2 days, $p = 0.030$) in those in whom it was left open in comparison to all other patients. In patients having CC, pre-surgery oxygen saturation was significantly lower in patients with relevant MAPCAs (58% vs 70%, p 0.019). Echocardiography had a sensitivity of 53% and a specificity of 100% in detecting relevant MAPCAs, accurately.
Conclusions: MAPCAs are frequently found in dTGA patients and can be associated with lower baseline oxygen saturation and a prolonged postoperative course after ASO. Transthoracic echocardiography cannot replace CC as diagnostic tool. If significant MAPCAs are suspected, early CC should be performed for diagnostic and therapeutic reasons.

1. Introduction

Nowadays, standard surgical management for simple type of d-transposition of the great arteries (dTGA) is the arterial switch operation (ASO), if coronary anatomy and valvular morphology allow this surgical technique [1,2]. Despite good short and long-term results with ASO in patients with dTGA, enlarged bronchial arteries/major aortopulmonary collateral arteries (MAPCAs) are possible findings in this patient group [2–4]. MAPCAs are often clinically irrelevant, but may cause congestive heart failure after surgical repair due to significant left-to-right shunting [4–7]. Possible symptoms of hemodynamic relevant MAPCAs are pulmonary volume overload, respiratory failure, left atrial and ventricular dilatation as well as dysfunction, failure to thrive, tachycardia or arrhythmias [6,7]. The gold standard for the diagnosis of hemodynamically significant MAPCAs after ASO is angiography during cardiac catheterization (CC). As CC after ASO is not performed on a routine basis, mostly patients with a complicated postoperative course or difficult coronary transfer due to special coronary anatomy undergo further invasive evaluation. In other case series, treatment of symptomatic MAPCAs was performed by transcatheter coil embolization of the collateral vessels [5–9].

Aim of this study was to evaluate the incidence and clinical significance of MAPCAs during the early postoperative course after ASO for surgical repair of dTGA.

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2. Materials and methods

2.1. Study population

In this retrospective monocentric cohort study, we analyzed clinical data of patients with simple dTGA undergoing ASO between January 2007 and December 2016 at the University Children's Hospital of Zurich, Switzerland. Simple TGA was defined by the International Pediatric and Congenital Cardiac Code of the Association for European Pediatric and Congenital Cardiology (IPCCC-AEPC) [10] ICD-10 Code Q20.3 and could be accompanied by a patent foramen ovale (PFO, ≤ 3 mm), atrial septal defect (ASD, >3 mm), persistent arterial duct (PDA), ventricular septal defect (VSD) and/or vessel and valve abnormalities like coarctation of the aorta (CoA) or pulmonary valve stenosis. Patients with other cardiac comorbidities [e.g. atrio-ventricular septal defect (AVSD) or double outlet right ventricle (DORV)] were classified as complex TGA and were not included in this study. Indications for postoperative CC were difficult intraoperative coronary transfer during ASO, signs of ischemia (ST changes or exceptionally elevated troponin levels), arrhythmia or prolonged intensive care with prolonged mechanical ventilator support and prolonged need for inotropic agents (milrinone or adrenalin).

CC included hemodynamic evaluation and contrast angiography for coronary arteries and MAPCAs. MAPCAs were defined as hemodynamically relevant if injection of contrast agent into the MAPCAs was followed by a clearly visible opacification of the correspondent pulmonary veins. Two groups were created based on these findings: Patients a) with (Group I) or b) without hemodynamically relevant MAPCAs (Group II). The latter includes patients who did not undergo CC (Group II A) and patients without relevant MAPCAs in CC (Group II B) (Tables 1 and 2).

2.2. Treatment techniques

Surgical procedures were performed using moderately hypothermic cardiopulmonary bypass and cold blood cardioplegic arrest. When necessary, repeat cardioplegia by selective cannulation of the coronary buttons was performed. ASO included switching of the aorta and the pulmonary roots, coronary button transfer, obligatory ligation and transection of PDA, and closure of ASD and VSD (as indicated) [11,12]. Lecompte-maneuver (translocation of the pulmonary bifurcation anterior to the neo-aorta) was performed as a rule. Taking into account the limitations that such a retrospective data analysis involves, there were no indicators in the MAPCA group to suggest increased non-coronary blood return during extracorporeal circulation (ECC) which could be indicative of MAPCAs. No routine postoperative angiograms were performed.

CC was performed under general anesthesia and mechanical ventilation. Femoral arterial vascular access was achieved via a 4 Fr introducer sheath in all patients. After hemodynamic assessment and aortic angiography, relevant MAPCAs were selectively cannulated by using a thin-walled end-hole catheter and a 0.014 in. torque wire. In those patients with MAPCAs judged to be hemodynamically relevant (Group I), micro catheters (e.g. Cantata®, COOK Medical/Bloomington/U.S.A.) were used for coil delivery using MRI-compatible coils (BALT Extrusion Spirale® embolization coil). To categorize the risk for the cardiac catheterization procedures we used the "procedure type risk category" published by L. Bergersen et al. [13], which distinguishes 4 different risk categories [1–4], as well as their "definitions for adverse event severity" (adverse event severity level 1–5) [13].

2.3. Data collection

Clinical data (patients' demographics, co-morbidities, data on intraoperative and postoperative course, data on CC) were extracted from patient medical records.

2.4. Statistical analysis

The data is presented as median with interquartile range (IQR) or mean ± standard deviation (SD), as appropriate. Categorical data is expressed as counts and percentages. Comparison of percentages was performed by the use of Pearson's Chi-Square test or Fishers' exact test when appropriate, those of mean values by Students' *t*-test and those of median values by the Mann-Whitney *U* test. If significant differences occurred, post-hoc analysis with appropriate p-value adjustment for multiple testing was performed. Significance testing was 2-sided with the significance level set at $p < 0.05$.

2.5. Ethics

All data was obtained primarily for medical purposes. The study design fulfills the guidelines of the Declaration of Helsinki regarding ethical principles for medical research involving human subjects. The study was approved by the institutional ethical board.

3. Results

3.1. Patient characteristics

103 neonates with simple dTGA were treated with ASO including Lecompte-maneuver between January 2007 and December 2016. 5 patients were excluded from the analysis due to missing parental consent or loss of follow up. Two patients died postoperatively during hospital stay. Both had an emergency ASO, one on the first and the other one on the second day of life due to insufficient arterial oxygen saturation despite adequately sized atrial septal defect and PDA. The first patient died because of severe left coronary artery stenosis with myocardial infarction despite multiple surgical revisions at the age of 12 days (12 days after ASO, 11 days after diagnostic CC) and the other patient due to severe postoperative mitral regurgitation and heart failure at the age of 17 days (16 days postoperatively, 9 days after diagnostic CC). CC showed normal coronary arteries in this patient. Overall,

Table 1
Baseline characteristics of patients with and without relevant MAPCAs.

	Total	Patients with relevant MAPCAs in CC (Group I)	Classified as no relevant MAPCAs (Group II)	Patients without CC (Group II A)	Patients with no or no relevant MAPCAs in CC (Group II B)	p-Value *
Numbers of patients n (%)	98 (100)	15 (15)	83 (85)	61 (62)	22 (23)	–
Male n (%)	70 (71)	7 (47)	63 (76)	46 (75)	17 (77)	**0.021**
GD, days (mean ± SD), n = 96	274.69 ± 12.76	269.00 ± 11.95	275.72 ± 12.70	275.33 ± 13.64	275.82 ± 9.79	0.060
Weight, grams (median (IQR)), n = 98	3455 (2990–3863)	3050 (2720–3460)	3500 (3000–3880)	3600 (3000–3910)	3455 (3070–3680)	**0.050**
Height, cm (mean ± SD), n = 96	50 ± 3	49.47 ± 2.5	49.81 ± 2.7	50 ± 3	49.34 ± 1.8	0.650
Atrial septal with:						0.297
— No atrial septal defect n (%)	1 (1)	0 (0)	1 (1)	1 (2)	0 (0)	
— PFO < 3 mm n (%)	48 (49)	10 (67)	38 (46)	32 (52)	6 (27)	
— ASD II > 3 mm n (%)	49 (50)	5 (33)	44 (53)	28 (46)	16 (73)	
VSD n (%)	30 (30.6)	5 (33)	25 (30)	14 (23)	11 (50)	0.770
Coronary anatomy: n (%)						0.385
— Normal	67 (68.4)	8 (53)	59 (71)	49 (80)	10 (46)	
— LCX ex RCA	10 (10.2)	2 (13)	8 (10)	3 (5)	5 (23)	
— Single-coronary ostium	7 (7.1)	2 (13)	5 (6)	2 (3)	3 (14)	
— Others	14 (14.3)	3 (20)	11 (13)	7 (12)	4 (18)	
Rashkind n (%)	75 (76.5)	13 (87)	62 (75)	49 (80)	13 (59)	0.509
SpO2 before Rashkind (mean ± SD)	63 ± 16	58 ± 12	64 ± 17	62 ± 17	70 ± 15	0.205
SpO2 after Rashkind (median (IQR))	85 (80–90)	85 (79–90)	85 (80–90)	85 (80–90)	87 (84–90)	0.786
SpO2-difference after Rashkind (median (IQR))	20 (14–35)	23 (19–36)	20 (13−33)	20 (13–34)	23 (15–28)	0.339

Group I: "Patients with relevant MAPCAs in CC"; Group II: "Classified as no relevant MAPCAs", includes Group II A ("Patients without CC") and Group II B ("Patients with no or no relevant MAPCAs in CC"). (*) p-value calculated for the comparison of Groups I and II.
n: numbers of patients; SD: standard deviation; IQR: interquartile range; GD: gestation days; PFO: persistent foramen ovale; ASD: atrial septal defect; VSD: ventricle septum defect; LCX ex RCA: left circumflex ex right coronary artery; Data shown as mean ± standard deviation, median and interquartile range (IQR) or number and percent in general study population.

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