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Repair of common arterial trunk: palliation and delayed correction as a viable alternative strategy in selected patients

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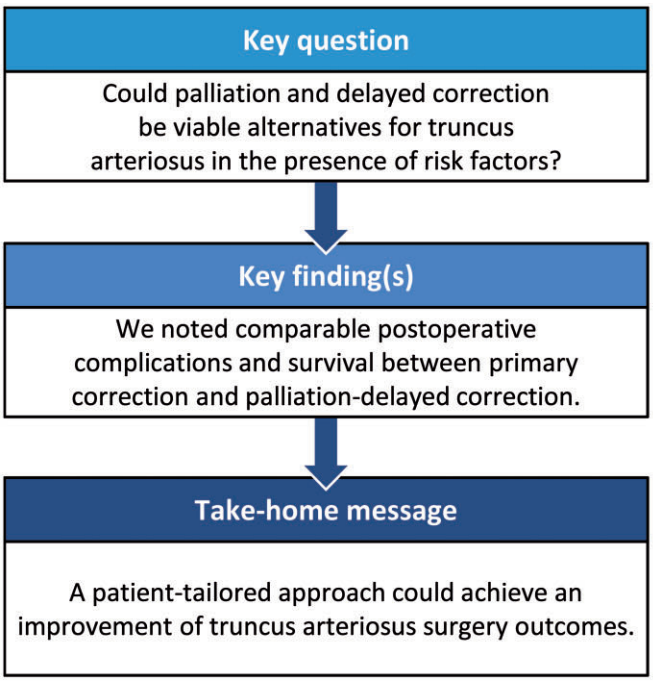

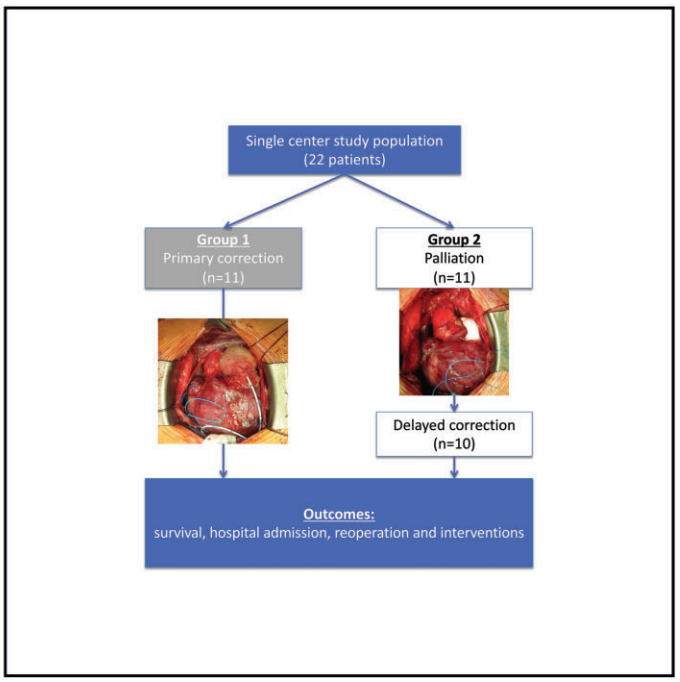

Abstract

OBJECTIVES: Primary repair of common arterial trunk (CAT) is burdened by high mortality rates, especially in the presence of multiple risk factors. Timing, possible palliative methods, optimal management of associated cardiac lesions and handling of a poor preoperative state are still under discussion.

METHODS: We retrospectively analysed all patients who underwent surgery for CAT in our institution between 2008 and November 2020. We included 22 patients, 11 of whom received primary correction (PC) and 11 of whom underwent initial palliation by partial repair, leaving the ventricular septal defect open and connecting the right ventricle to the pulmonary arteries with a small valveless right ventricle-to-pulmonary artery conduit. A delayed correction (DC) was performed after 11.5 ± 3.6 months.

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RESULTS: The overall operative mortality was 4.5%; 1 patient (affected by severe truncal valve stenosis and presenting in a poor state preoperatively) in the DC group died after palliation. The incidence of postoperative pulmonary hypertensive crisis was significantly higher in the PC group ($P = 0.027$). No patient from either group required postoperative extracorporeal support. Survival rates after 6 years differed slightly (PC group, 90%; DC group, 70%; log-rank = 0.270).

CONCLUSIONS: PC of CAT remains an optimal surgical approach for patients with an expected low mortality. However, our data support palliation and DC as a suitable alternative strategy, especially in the presence of significant risk factors like interrupted aortic arch, poor preoperative condition or complex surgical anatomy.

Keywords: Common arterial trunk • Palliation • Sano-Shunt • Outcomes • Patient-tailored therapy • Congenital cardiac surgery

ABBREVIATIONS

BSA	Body surface area
CPB	Cardiopulmonary bypass
CAT	Common arterial trunk
DC	Delayed correction
IAA	Interrupted aortic arch
PC	Primary correction
PAB	Pulmonary artery banding
pHT	Pulmonary hypertension
RVPA	Right ventricle-to-pulmonary artery
VSD	Ventricular septal defect

INTRODUCTION

Common arterial trunk (CAT) accounts for 0.7–3% of all congenital cardiac defects [1, 2] and is associated with interrupted aortic arch (IAA) in 15% [3]. The natural history of patients with CAT is extremely unfavourable, so timely surgical repair remains the only life-saving treatment [4].

Outcomes of the primary correction (PC) of CAT have steadily improved over the years [5]. Nevertheless, associated mortality and morbidity rates remain relatively high. Mastropietro *et al.* [6] reported in a multicentre analysis a 6.9% mortality rate and a 20% rate of postoperative major adverse events. Patients with associated cardiac anomalies and a critical preoperative haemodynamic state often require early surgical repair in the neonatal period, which is associated with higher risk. Moreover, truncal valve dysfunction or the presence of IAA is also associated with increasing mortality and complication rates [7, 8]. Therefore, precise timing, management of associated cardiac lesions and an extremely poor preoperative condition remain controversial topics [9].

Our institution has implemented an alternative approach for high-risk patients (i.e. those with complex anatomy, associated severe cardiac anomalies, postnatal cardiac failure) with immediate palliation and delayed correction (DC) to improve outcomes. The feasibility and the favourable outcomes associated with this approach were reported previously, especially in cases of associated IAA and preoperative low cardiac output [8, 10]. Our goal was to compare both strategies (PC and DC) in terms of safety, survival and prognosis.

METHODS

We performed a retrospective observational study of all patients who underwent surgery for CAT between January 2008 and November 2020 at our centre. The need for informed consent was waived because of the retrospective nature of the collected data and review. Surgical strategies represented a patient-tailored approach, determined individually by the institutional heart team, based on haemodynamic data, anatomy and patient status. We addressed palliation and DC to patients with high-risk constellations and unfavourable prognosis. Criteria favouring palliation were low weight, prematurity, presence of IAA, preoperative cardiogenic shock in combination with complex anatomy [e.g. ventricular septal defect (VSD) not accessible through a limited right ventriculotomy].

Pre-, intra- and postoperative data were collected from patient records. Imaging modalities included echocardiograms, computed tomography scan of the chest (Fig. 1) or cardiac catheterization; the Van Praagh classification was used routinely [11]. Images were used to calculate preoperative Nakata indices [right pulmonary artery area + left pulmonary artery area/body surface area (BSA)]. The mean comprehensive Aristotle score was calculated for each patient.

Follow-up was completed by a routine outpatient medical examination or hospitalization for diagnostic imaging or percutaneous interventions.

The primary outcomes were operative mortality, postoperative major complications (defined as cardiopulmonary re-animation and/or extracorporeal support) and postoperative pulmonary hypertension (pHT) crisis in both study groups. Secondary outcomes were survival and reintervention rates (surgery and/or percutaneous interventions) during follow-up.

Surgical techniques: primary correction

All patients, independently from the chosen strategy, were approached via a median sternotomy, cardiopulmonary bypass (CPB) and bicaval cannulation. Patients were cooled to temperatures of 25.7±1.9°C. After the initiation of CPB and administration of crystalloid cardioplegia, the pulmonary arteries were excised from the truncal vessel. The resulting defect was closed primarily or with a bovine pericardial patch. Right ventricle-to-pulmonary artery (RVPA) continuity was reconstructed with a valveless or valved conduit ($n = 5$ Contegra 12 mm; Medtronic, Inc, Minneapolis, MN, USA; $n = 1$ Labcor 11 mm; Labcor Laboratórios-Ltda, Brazil; $n = 1$ Hancock-II 12 mm; Medtronic, Inc; $n = 2$ Dacron valveless prosthesis; $n = 1$ decellularized homograft 'Espoir' 9 mm; Corlife OHG, Hannover, Germany) and in 1 patient with a direct RVPA anastomosis. The VSD was closed with a patch ($n = 1$ Dacron, $n = 10$ bovine pericardium). Additional procedures such as patent foramen ovale/ASD closure ($n = 7$), VSD enlargement ($n = 1$), persistent left superior vena

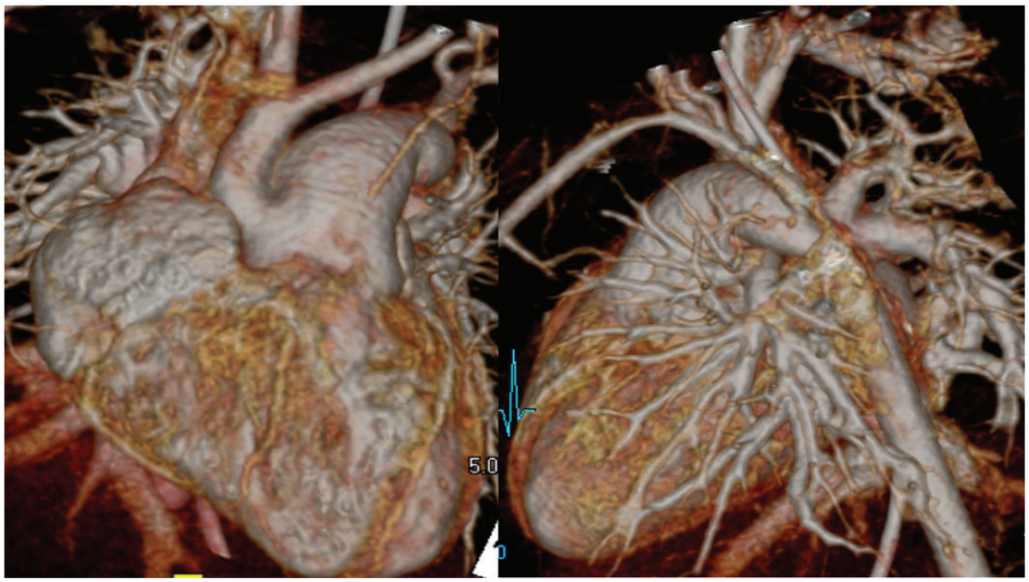

Figure 1: Preoperative computed tomography scan and 3-dimensional reconstruction.

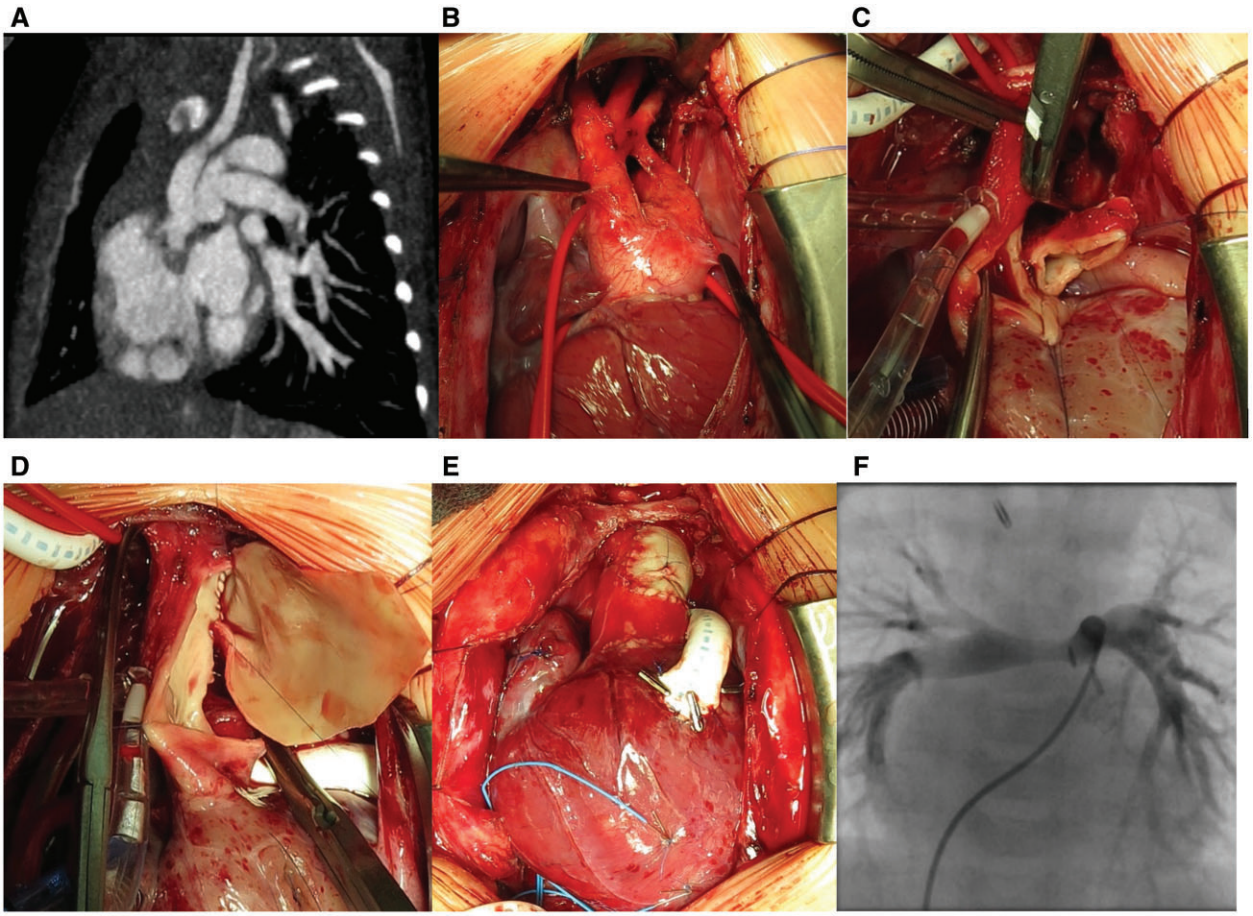

Figure 2: Palliation and aortic arch reconstruction of a complex common arterial trunk type A4. (**A**) Preoperative computed tomography scan; (**B**) intraoperative anatomy; (**C**) pulmonary artery division from the truncal vessel and pulmonary bifurcation reconstruction; (**D**) harmonic augmentation of the aortic arch with a large bovine pericardial patch; (**E**) final result: the right ventricle to pulmonary artery connection was established with a 6-mm valveless Gore-Tex prosthesis; and (**F**) diagnostic catheterization before the scheduled repair.

cava ligature ($n = 1$) and left pulmonary artery plasty ($n = 1$) were also performed.

Surgical techniques: palliation and delayed correction

The mean lowest systemic temperature was 27.7 ± 3.3°C, and myocardial protection consisted of tepid-blood cardioplegia. Division of pulmonary arteries from the truncal vessel was the same procedure as that used for the primary repair. The RVPA continuity was established by implantation of a 6-mm valveless Gore-Tex prosthesis (W. L. Gore & Associates, Newark, DE, USA). Necessary additional procedures, such as correction of an aberrant subclavian artery ($n = 1$) due to tracheal compression, were also performed. One patient with severe truncal valve stenosis underwent valve reconstruction according to the Ozaki procedure with decellularized xenopericardial patches. A total of 36% of the patients ($n = 4$) had a coexisting IAA ($n = 3$ type B; $n = 1$ type A). In these cases, deep hypothermia was required (mean temperature = 24.7°C), and unilateral antegrade cerebral perfusion was performed by the cannulation of the innominate artery through a 3.5-mm Gore-Tex prosthesis. The descending perfusion was ensured through PDA cannulation and later by retrograde perfusion through a femoral cannula during arch repair or in a few cases through direct cannulation of the descending aorta. After resection of ductal tissue, an end-to-end anastomosis of the distal ascending aorta and posterior wall of the proximal descending aorta was performed. The anterior wall was augmented with a large bovine pericardial patch warranting a harmonic good-sized aortic arch. RVPA continuity was established with a 6-mm valveless Gore-Tex prosthesis (Fig. 2). All patients had adequate oxygen saturation after surgery (range: 85–99%). VSD closure in conjunction with a larger valved conduit implant was scheduled for the next 7–19 months (11.5 ± 3.6) after diagnostic catheterization.

After resternotomy, the onset of CPB and the administration of tepid-blood cardioplegia, the VSD was closed with a patch ($n = 6$ bovine pericardium, $n = 4$ Dacron) and a valved conduit was implanted ($n = 5$ Contegra 12 mm; $n = 2$ Contegra 14 mm; $n = 2$ Hancock-II 12 mm; $n = 1$ Matrix-P-plus 13 mm; AutoTissue GmbH, Berlin, Germany). Pulmonary artery enlargement ($n = 2$ left, $n = 2$ right) with a bovine pericardial patch was performed additionally. Central truncal valve regurgitation was repaired by leaflet plication in 1 case.

Statistical analyses

Categorical variables are expressed as frequencies (percentages) and continuous variables, as mean (±standard deviation) or median (interquartile range). Continuous variables were compared by the two-tailed paired *t*-test and categorical variables, by the χ^2 test. Non-normally distributed data were tested using the Mann–Whitney test.

The Kaplan–Meier method was used for survival analysis. The statistical analyses were performed with SPSS software (IBM-SPSS Statistics, Release 20.0.0, SPSS, Inc., Chicago, IL, USA).

Table 1: Baseline characteristics of the study population

	Group 1	Group 2			
	Primary correction ($n = 11$)	At the time of palliation ($n = 11$)	*P*-value (palliation versus PC)	At the time of delayed correction ($n = 10$)	*P*-value (delayed versus PC)
Demographic characteristics					
Age (days)	46 [32–60]	11 [8–21]	***0.005***	369 [214–214]	***<0.001***
Female gender	6 (54%)	4 (36%)	*0.416*	3 (30%)	
Weight (g)	3680 (±420)	3236 (±585)	*0.061*	8198(±161)	*<0.001*
BSA (m^2)	0.23 (±0.02)	0.21 (±0.02)	*0.049*	0.38 (±0.05)	*<0.001*
Prematurity (<37 weeks)	0 (0%)	2 (18%)	*0.152*	–	
Cardiogenic shock	1 (9%)	3 (27%)	*0.291*	0 (–)	
Preoperative inotropic support	2 (18%)	2 (18%)	*1.000*	0 (–)	
Preoperative ventilation[a]	1 (9%)	2 (18%)	*0.557*	0 (–)	
Prior percutaneous intervention[b]	2 (18%)	1 (9%)	*0.557*	4 (40%)	*0.292*
Aristotle score	11 ± 0	12 ± 2	*0.142*	11 ± 0	*1*
Comprehensive Aristotle score	12.4 (±1.9)	14.4 (±2.7)	*0.089*	–	–
Tracheomalacia	1 (9%)	1 (9%)	*1*	1 (10%)	*1*
Van Praagh classification					
A1	6 (54%)	5 (45%)			
A2	4 (36%)	1 (9%)			
A3	1 (9%)	1 (9%)			
A4	0 (–)	4 (36%)			
Associated congenital cardiovascular anomalies					
IAA	0 (–)	4 (36%)			
ASD/PFO	7 (63%)	9 (81%)			
Coronary anomalies	0 (–)	1 (9%)			
Severe truncal valve stenosis/insufficiency	1 (9%)	1 (9%)			

Values are presented as mean (±standard deviation), number (%) or median [interquartile range]. *p* values are formatted in italics; those < 0.05 are formatted in bold.

ASD: atrial septal defect; BSA: body surface area; IAA: interrupted aortic arch; PC: primary correction; PFO: patent foramen ovale.

[a]Included only endotracheal intubation.

[b]Any interventional procedure for pulmonary artery or truncus valve inclusive of a stent implant.

Table 2: Intra- and postoperative outcomes

	Group 1	Group 2			
	Primary correction ($n = 11$)	Palliation ($n = 11$)	*P*-value (palliation versus PC)	Delayed correction ($n = 10$)	*P*-value (delayed versus PC)
CPB time (min)	211 (±38)	169 (±66)	*0.094*	204 (±44)	*0.731*
Aortic cross-clamp time (min)	120 (±37)	94 (±29)	*0.091*	85 (±40)	*0.068*
RVPA conduit type					
Valved	8(73%)	0 (–)		10 (100%)	
Valveless	2 (18%)	11 (100%)		0 (–)	
None	1 (9%)	0 (–)		0(–)	
RVPA conduit size (mm)	12 [10.25–12]	6 [6–6]		12 [12–12]	
RVPA conduit size/BSA (mm/m^2)	49.1 ± 5.7	28.6 ± 3	***<0.001***	32.97 ± 4.5	***<0.001***
Red blood cells (ml)	363 (±105)	375 (±218)	*0.877*	323 (±246)	*0.645*
Platelets (ml)	126 (±61)	179 (±118)	*0.225*	222 (±163)	*0.099*
Fresh frozen plasma (ml)	205 (±129)	100 (±126)	*0.079*	97.6 (±132)	*0.087*
ICU stay (days)	10 [6]	9 [4]	*0.195*	3 [1]	*0.058*
VIS	19.5 [13]	18.9 [7]	*0.295*	8.4 [1.7]	*0.382*
Postoperative pHT	4 (36%)	0 (–)	***0.0027***	1 (10%)	*0.173*
Delayed sternal closure	4 (36%)	3 (27%)	*0.675*	1 (10%)	*0.173*
CPR	1 (9%)	2 (18%)	*0.557*	0 (–)	*0.353*
ECMO	0 (–)	0 (–)	*1*	0 (–)	*1*
AKI	0 (–)	2 (27%)	*0.152*	0 (–)	*1*
Length of stay (days)	19 [10]	16 [6]	*0.292*	8 [6]	*0.106*

Values are presented as mean (±standard deviation), number (%) and median [interquartile range]. *p* values are formatted in italics; those < 0.05 are formatted in bold.

AKI: acute kidney injury; BSA: body surface area; CPB: cardiopulmonary bypass; CPR: cardiopulmonary resuscitation; ECMO: extracorporeal membrane oxygenation; ICU: intensive care unit; PC: primary correction; pHT: pulmonary hypertensive crisis; VIS: vasoactive inotropic score.

Table 3: Pulmonary valve and vessels data

	Group 1	Group 2			
	Primary correction ($n = 11$)	At time of palliation ($n = 11$)	*P*-value (palliation versus PC)	At time of delayed correction ($n = 10$)	*P*-value (delayed versus PC)
PV *z*-values	1.7 (±0.74)	-2.2 (±0.73)	***<0.001***	0.54(±0.52)	***<0.001***
LPA (mm)	4.8 (±2)	4.6 (±0.9)	*0.834*	5.5 (±1.5)	*0.446*
RPA (mm)	5.7 (±2.2)	5.4 (±0.8)	*0.797*	6.4 (±0.7)	*0.361*
Nakata index (mm/m^2)	143.6 (±112)	131.0 (±38)	*0.741*	109.0 (±35)	*0.376*

Values are presented as mean (±standard deviation). *p* values are formatted in italics; those < 0.05 are formatted in bold.

LPA: left pulmonary artery; PC: primary correction; PV: pulmonary valve; RPA: right pulmonary artery.

RESULTS

Data from 22 patients who underwent CAT surgery were compared: 11 after PC and 11 after DC. Preoperative data and patient characteristics are summarized in Table 1. The median age in the PC group was 46 days (range 7–91 days; only 1 patient from Africa was treated at the age of 282 days). Patients in the DC group were significantly younger ($P = 0.005$); the median age was 11 days (range 5–53 days). Three patients in the DC and 1 patient in the PC group presented with preoperative cardiogenic shock. There were 2 (18%) premature infants (<37 weeks) in the DC group and none in the PC group. The associated cardiac lesions are listed in Table 1. IAA (CAT type A4) was reported only in the DC group (4 patients). Each group included 1 patient with severe truncal valve dysfunction and tracheomalacia. The overall operative mortality was 4.5% (=1/22); no patient in the PC group and 1 patient in the DC group died after palliation.

All 10 survivors in the DC group completed DC with a median age of 369 days (range 214–574 days) and a mean weight of 8198 ± 161 g. At time of the delayed repair, all patients were electively hospitalized in the absence of acute symptoms. The mean comprehensive Aristotle score was higher in the DC group (14.4 ± 2.7) but not statistically significant ($P = 0.086$).

Intraoperative and in-hospital outcomes are shown in Table 2. Time on CPB, aortic cross-clamping, number of blood transfusions, incidence of major complications and rethoracotomy were not significantly different between the 2 groups at both times (i.e. palliation and DC versus PC). The incidence of postoperative pHT was significantly higher in the PC group ($P = 0.027$). No patient required postoperative ECMO. Ventilation times until extubation were similar in both groups at the time of the initial procedure (median 5 days). One patient, after primary repair, underwent a tracheostomy due to severe tracheal obstruction. Most patients who underwent delayed repair were extubated on the day of the operation or on the first postoperative day (range: day 0–postoperative day 9). Stays in the intensive care unit and the hospital were shorter in the DC group but were not statistically significant.

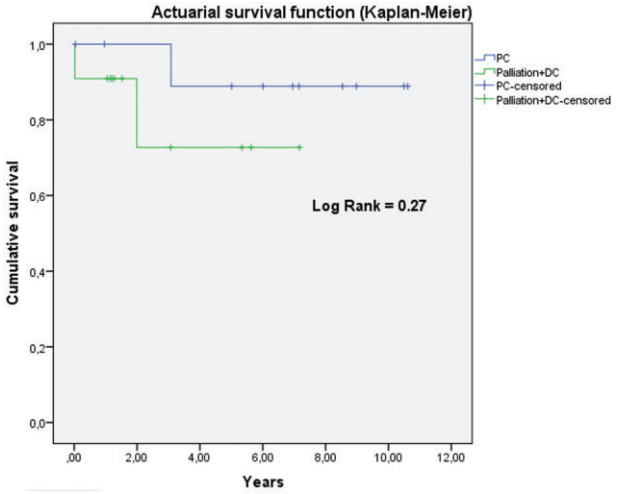

Figure 3: Kaplan–Meier survival curve; blue: primary correction group; green: delayed correction group.

The indexed RVPA conduit size was significantly higher in the PC group (49.1 ± 5.7 mm/m^2) when compared with the DC group at palliation (28.6 ± 3 mm/m^2) and at delayed repair (32.97 ± 4.5 mm/m^2) ($P < 0.001$). Pulmonary valve *z*-values were significantly higher in the PC group accordingly (Table 3).

The mean follow-up duration was 32.5 months (976 ± 810 days, range 385–2618) in the DC group and 75 months (2250 ± 1250 days, range 14–3871) in the PC group. Kaplan–Meier curves are shown in Fig. 3. Until November 2020, we observed an overall mortality of 9.5%: 2 patients out of the 21 survivors, 1 in each group, died late after hospital discharge (log-rank = 0.270). Other outcomes at follow-up are shown in Table 4. To date, reoperation rates for RVPA conduit replacement were lower in the DC group after complete repair but were not statistically significant (30% in the DC group, 64% in the PC group; $P = 0.136$). In addition, 2 patients in the PC group underwent an operation because of severe truncal valve regurgitation (1 truncal valve repair and 1 replacement). Percutaneous angioplasty or stent implants in the pulmonary arteries and/or RVPA conduit and rehospitalization rates were lower in the DC group but were not statistically significant.

DISCUSSION

Outcomes of surgical CAT repair have steadily improved in recent years [5]; however, mortality and morbidity remain relatively high [6, 12, 13]. The estimated risk of death for children affected by CAT, reported from the combined resources of the European Association for Cardiothoracic Surgery Congenital Heart Surgery Database (33 360 operations) and the Society of Thoracic Surgeons Congenital Heart Surgery Database (43 934 patients) between 2002 and 2007, was 14.1% (range 11.4–16.8%). In case of simultaneous IAA repair, the reported mortality was 29.8% (range 17.7–44.3%) [12].

Many reports were able to elucidate factors that contribute to such a high number of deaths. Naimo and colleagues reported an early mortality of 17% for patients undergoing simultaneous CAT and IAA repair [14]. McCrindle *et al.* reported worsened outcomes for patients with CAT and IAA repair in contrast to IAA repair alone [15]. Russell *et al.* [7] identified IAA as the single greatest risk factor for death in patients affected by CAT.

Table 4: Midterm follow-up outcomes

	Group 1, *n* (%)	Group 2, *n* (%)	*P*-value
RVPA conduit first reoperation	7 (64)	3 (30)	*0.136*
RVPA conduit second reoperation	1 (9)	0 (–)	*0.353*
Percutaneous intervention	9 (82)	7 (70)	*0.549*
Rehospitalization	9 (82)	7 (70)	*0.549*

Values are presented as number (%). *p* values are formatted in italics.
RVPA: right ventricle to pulmonary artery.

As previously cited, Mastropietro *et al.* [6] reported in a multicentric analysis 6.9% mortality and a 20% rate of postoperative major complications. However, patients with type A4 CAT were excluded from this analysis.

The goal of our institutional approach was to face the problem of this high mortality through an alternative strategy for selected patients. The resulting overall operative mortality in our small series was 4.5%, which included 4 patients with coexisting IAA.

No PC patient died, which might be explained by the decision to postpone complete repair, whenever possible, from the neonatal period. In fact, Naimo *et al.* recently reported a 23.7% mortality rate for CAT repair in neonates in contrast to 7.4% in older patients. Although early repairs have steadily improved over time, multivariable analysis showed that CAT repair in neonates remains an important risk factor for postoperative death [16].

Operative mortality in the DC group was 9%, not surprisingly higher than that reported by Mastropietro and colleagues, because 36% of patients had associated IAA (type A4 CAT), 18% were premature and 27% presented with preoperative cardiogenic shock. Despite a relatively higher mortality in the DC group, our results are more than satisfying, because we have been able to treat patients with complex issues and achieve outcomes comparable to those reported in previous studies of patients with simpler anomalies [13].

Some authors prefer pulmonary artery banding (PAB) as an initial palliative strategy, analogous to the hybrid approach of bilateral PAB in the setting of hypoplastic left heart syndrome. Although this approach is challenging in small patients [17, 18], the placement of PAB as an initial palliative strategy may be beneficial and even life-saving in high-risk patients.

Although CPB is needed, our alternative strategy has several advantages to control postoperative complications in high-risk patients. The RVPA conduit guarantees a limited pulsatile antegrade flow, which allows somatic and pulmonary artery growth. The use of 6-mm RVPA conduits (significantly smaller in comparison to PC with an average of 12 mm) avoids hypervolaemia in the pulmonary artery system and prevents mismatch between the RVPA conduit and tiny neonatal pulmonary arteries. Moreover, the right ventriculotomy is limited, thus avoiding postoperative myocardial right ventricular failure. The left open VSD preserves cardiac output during phases of elevated pulmonary resistance. All previously mentioned arguments have convinced us to prefer the DC strategy in low-weight or premature patients, who often present with pulmonary impairment and thus could benefit from a limited right ventriculotomy and a smaller RVPA conduit. Although our palliation appears more invasive in comparison to simple PAB, we succeeded in treating these patients and achieving low mortality and morbidity rates. The brief period

of CPB was no problem at all, even in the extremely small patients. As explained previously, our palliation strategy not only comprises a delayed VSD closure but also encompasses a more complex concept, and each detail contributes, in synergy with the other details, to a preferable outcome.

The issue of an oversized RVPA conduit implant and a related large right ventriculotomy was already addressed in previous studies, which showed a trend towards more deaths in patients with a larger RVPA conduit diameter and the association between postoperative cardiac major adverse events and an RVPA diameter indexed to BSA > 50 mm/m^2 [6, 19]. The exact reasons have not yet been identified, but some authors [6] have postulated that larger conduit diameters are associated with a larger sized right ventriculotomy, which directly impairs postoperative RV function. Further postoperative arrhythmias might also be induced, leading to further aggravation [6]. Moreover, larger conduits could cause kinking or distortion of the pulmonary arteries due to a size mismatch, resulting in a relative conduit stenosis [6].

In our experience, patients with a DC showed 70% survival after 6 years, which, in our opinion, represents an excellent outcome, considering the preoperative constellation. The only death observed in the DC group after primary palliation occurred in a patient with coexisting severe truncal valve stenosis and severe myocardial hypertrophy. She presented with preoperative persistent low cardiac output despite inotropic support and respiratory failure, so preoperative ventilation was required. Intraoperatively, we found that the dysplastic truncal valve was thickened with fused commissures and rigid leaflets. As demonstrated in previous studies, truncal valve dysfunction, especially in the presence of moderate-to-severe stenosis, is often associated with preoperative death and is an independent risk factor for postoperative mortality and morbidity [7, 20, 21]. Some authors postulated that early neonatal surgery would avoid any further organ damage [20]. The haemodynamic condition of our patient forced us to perform surgery on the 7th day of life (weight = 2.3 kg). Our initial plan was to perform a truncal valve commissurotomy and apply our palliation strategy. However, the patient's anatomy made a more complex reconstruction necessary (Ozaki technique).

At the midterm follow-up, we observed an overall mortality of 9.5%. One patient who underwent primary repair died late of dehydration that developed during a severe form of gastrointestinal infection, after returning to Africa. Another patient in the DC group who had a severe form of tracheolaryngomalacia died late, which underlines the fact that coexisting non-cardiovascular pathologies play an important role in the long-term outcomes after CAT repair. Buckley *et al.* [22], for example identified DiGeorge syndrome and the need for postoperative tracheostomy as independent risk factors for late mortality after CAT repair.

The postoperative incidence of major cardiac adverse events was similar in both groups: We had 1 case of cardiopulmonary resuscitation in the PC group versus 2 in the DC group; at palliation time, no patient had to be placed on ECMO postoperatively in order to improve outcomes.

Postoperative pHT occurred in 4 (36%; $P = 0.027$) patients who underwent PC: 3 of these (75%) had a relatively large RVPA conduit implanted when indexed to BSA (>50 mm/m^2). Patients who underwent primary palliation and received a 6-mm RVPA conduit had no signs of pHT. This fact is explained by the combined protective effect of our surgical strategy: implanting a 6-mm prosthesis significantly downsizes the RV incision (thus limiting the surgical damage to muscle and preventing the RV failure) and leaving the VSD open preserves cardiac output during pHT. Patients who received PC were significantly older than those in the DC group at palliation (median, 46 days; range 7–91 days; only 1 patient was treated at the age of 282 days), which increases the risk of pHT. Furthermore, PC was not significantly delayed in this study group, and the surgical timing range was similar to that in other studies [16].

Only 1 patient in the DC group presented with postoperative RV dysfunction due to pulmonary restriction after repair, although the indexed RVPA conduit size was <50 mm/m^2. However, this patient had an extremely restrictive pulmonary vascular system, as shown by the Nakata index of 81 mm^2/m^2 and a McGoon ratio of 1.34.

Interestingly, we found that the mean indexed RVPA conduit size in the DC group at delayed repair was significantly smaller than those observed in the PC group. This finding underlines the fact that extremely oversized RVPA conduits were implanted in the PC group.

In our analysis, we did not find a statistically significant difference between the Nakata index at the time of palliation and at the time of delayed repair. We would like to postulate that the 6-mm RVPA conduit ensured the development of the pulmonary vessels as well as the somatic growth of the patients.

At follow-up, we recognized lower (but not statistically relevant) reoperation rates for the RVPA conduit site after a staged correction. However, it is fair to point out that the follow-up period for patients undergoing PC had a longer duration.

Recently, Hames *et al.* [23] reported an interesting large analysis regarding outcome of ECMO in patients after primary CAT correction. They showed that only 55.8% of those patients survived to hospital discharge. The multivariate analysis for mortality for patients on ECMO showed many variables as risk factors, which are neither editable nor predictable. For this reason, the authors advocate avoiding ECMO support in underweight patients. We think that the high mortality rate of these patients signifies failure of the chosen surgical approach rather than a problem with the indication for ECMO. Indeed, the correlation of low weight with worse outcome suggests that an alternative approach (such as palliation with delayed repair) could be a preferable choice [24].

We believe that CAT primary repair is, whenever possible, still the best option. However, in patients with complex pathologies, unfavourable anatomy and postnatal heart failure, an alternative strategy could be extremely helpful to ensure a favourable outcome. Our study demonstrated that palliation and DC could be a viable alternative surgical strategy with at least comparable survival and complication rates, despite the certainty of a rather early necessary reoperation.

Limitations

This study has several limitations. First, it is an observational, retrospective study. Nevertheless, it is the first study reporting clinical outcomes with this alternative strategy (palliation and DC) compared to the traditional approach. Second, it is a single-centre study with a small number of patients, so our results could be affected by a type II error or low statistical power. However, larger studies

involving patients from different centres over a greater period of time could suffer from other biases (e.g. different surgeons, different protocols). We did not perform an adjustment for multiple testing: for this reason, *P*-values may not be interpreted as confirmatory but rather as descriptive. Finally, the follow-up, especially for the DC group, is restricted, which limits our ability to make stronger conclusions. Future studies are needed to better investigate this alternative strategy to improve the outcomes of CAT surgery.

CONCLUSIONS

Complete PC of CAT remains the optimal surgical approach whenever possible. However, our 10 years of experience suggest that palliation and delayed repair represent a safe and efficient alternative for patients affected by CAT. Palliative implantation of a 6-mm RVPA conduit avoids hypervolaemia in the pulmonary system and provides pulsed antegrade flow, allowing sufficient pulmonary vascular and somatic growth. Moreover, implanting a small-diameter RVPA conduit limits the incision in the right ventricle, thus avoiding RV failure and postoperative complications. This alternative approach could be an attractive option, especially for neonates with complex anatomy and/or postnatal cardiac failure and an estimated high mortality risk. A patient-tailored approach could achieve an improvement in CAT outcomes, thereby lowering the overall mortality and complication rates.

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Author contributions

Michela Cuomo: Conceptualization; Data curation; Formal analysis; Investigation; Methodology; Validation; Writing—original draft. **Ariawan Purbojo:** Supervision; Validation. **Robert Blumauer:** Validation; Visualization. **Martin Schöber:** Validation. **Wolfgang Wällisch:** Validation. **Sven Dittrich:** Conceptualization; Supervision; Validation; Writing—review & editing. **Robert Anton Cesnjevar:** Conceptualization; Data curation; Formal analysis; Methodology.

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