

# Postoperative arrhythmia in patients with bidirectional cavopulmonary anastomosis

Alessandra Reichlin<sup>a</sup>, René Prêtre<sup>b</sup>, Hitendu Dave<sup>b</sup>, Maja I. Hug<sup>c</sup>, Matthias Gass<sup>a</sup> and Christian Balmer<sup>a,\*</sup>

<sup>a</sup> Department of Cardiology, University Children's Hospital Zurich, Zürich, Switzerland

<sup>b</sup> Department of Cardiothoracic Surgery, University Children's Hospital Zurich, Zürich, Switzerland

<sup>c</sup> Department of Intensive Care and Neonatology, University Children's Hospital Zurich, Zürich, Switzerland

\* Corresponding author. University Children's Hospital, Steinwiesstrasse 75, 8032 Zürich, Switzerland. Tel: +41-44-2667111; fax: +41-44-2667981; e-mail: christian.balmer@kispi.uzh.ch (C. Balmer).

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## Abstract

**OBJECTIVES:** Bidirectional cavopulmonary anastomosis (BDCPA) is part of the staged Fontan palliation for congenital heart defects with single-ventricle morphology. The aim of this study was to describe incidence and characteristics of early postoperative arrhythmias in patients undergoing BDCPA.

**METHODS:** Retrospective analysis of 60 patients undergoing BDCPA at the age of <12 months from 2001 to 2008 at a single centre. Arrhythmias were subclassified in sinus bradycardia, premature atrial/ventricular contraction, supraventricular tachycardia and atrioventricular block. The groups were compared according to age at operation and diagnosis. Postoperative follow-up data were included until Fontan completion.

**RESULTS:** Postoperative arrhythmia was observed in 20 patients: 12 temporary and 8 persisting until hospital discharge. Sinus bradycardia is a common postoperative arrhythmia and occurred in 16 patients (9 transient, 7 persistent until hospital discharge). One patient undergoing BDCPA and a Damus–Kaye–Stansel procedure had a persisting first-degree atrioventricular block. The occurrence of a postoperative arrhythmia was independent of age and diagnostic group (hypoplastic left heart vs non-hypoplastic left heart). After hospital discharge, five of the eight arrhythmia resolved spontaneously resulting in 2 patients with sinus bradycardia and 1 patient with a first-degree AV block immediately before the Fontan completion was undertaken.

**CONCLUSIONS:** Postoperative arrhythmias in patients with BDCPA occur early after surgery and are temporary. Severe and life-threatening arrhythmias are rare although the interventions are complex and the patients very young. The most common arrhythmia is sinus bradycardia.

**Keywords:** Congenital heart disease • Univentricular heart • Bidirectional cavopulmonary anastomosis • Arrhythmia • Infant

## INTRODUCTION

Thanks to recent advances in the operative techniques in cardiothoracic surgery and in the postoperative care, we are nowadays able to offer therapeutic options to almost every neonate with a congenital heart malformation. While a near-to-normal life can be expected in many patients after biventricular repair, there still exists a considerable morbidity after palliative surgery in patients with single ventricles [1–3]. Arrhythmia, especially supraventricular arrhythmia, is a well-known reason for late morbidity and also mortality in these patients [4, 5]. At our centre, the staged Fontan palliation with a bidirectional cavopulmonary anastomosis (BDCPA) at the age of 3–6 months, followed by completion of the Fontan circuit with an extracardiac conduit at the age of 2.5 to 4 years, is the preferred surgical strategy in these patients.

The BDCPA consists of an end-to-side anastomosis of the superior vena cava (SVC) to the pulmonary artery (PA). The risk of postoperative arrhythmia in patients undergoing BDCPA is

increased due to the proximity of the suture lines at the SVC to the sinoatrial (SA) node and due to scar formation at the site of the right atriotomy predisposing to atrial re-entry tachyarrhythmias [4–6]. The multistage operative pathway to Fontan reconstruction further increases the risk of altered sinus node function [7, 8]. In addition, there are also some congenital heart defects with single-ventricle morphology and an inherent risk of arrhythmia due to the abnormal position or course of the conduction system, e.g. SA disease or supraventricular tachycardia in patients with atrial isomerism or high-degree AV conduction disturbances in patients with congenitally corrected transposition of the great arteries [5]. Age to perform an elective BDCPA varies from one centre to the other. Individual risks and haemodynamics have to be taken into account as well. It is generally considered to be a safe operation in patients as young as 2 months of age [9–14]. There is a trend towards an earlier time of operation, especially in patients with hypoplastic left heart syndrome (HLHS) after first-stage Norwood operation or hybrid approach, with the aim of

reducing the risk of inter-stage mortality. Moving the time of operation below 3 months of age may increase the risk of postoperative complications including arrhythmia, however.

In recent literature, there are some articles addressing the topic of postoperative complications in patients with BDCPA [2, 3], but there exist only few published data about the specific nature and incidence of arrhythmia in these patients [4, 6–8]. The purpose of this study is to describe the incidence and characteristics of postoperative arrhythmia in our patients with single-ventricle morphology after BDCPA in the initial time before undergoing the Fontan completion with a special view on age at operation and diagnosis as potential risk factors.

## MATERIALS AND METHODS

### Study patients

This study is a retrospective analysis of all consecutive patients undergoing BDCPA at the age of <12 months from 1 January 2001 to 31 December 2008 at a single tertiary referral centre (University Children's Hospital, Zürich, Switzerland). A total of 60 patients are included in the evaluation.

### Data collection

Data were retrieved from patient's charts including the Children's Hospital electronic database. Intracardiac anatomy at echocardiography was used to classify the diagnosis. The cohort was subclassified into HLHS and non-HLHS patients. HLHS patients are children with the classical HLHS and its variants, such as unbalanced atrioventricular septum defect (AVSD) with hypoplastic left ventricle, hypoplastic left ventricle outflow tract and hypoplastic aorta. Non-HLHS are all other patients with functionally single ventricle morphology, such as double inlet left ventricle (DILV), large ventricular septum defect (VSD)/double outlet right ventricle (DORV), tricuspid atresia, pulmonary atresia and AVSD. The following parameters were collected: cardiopulmonary bypass (CPB) time, duration of mechanical ventilation, duration of chest tube drainage, length of intensive care unit (ICU) stay, total length of hospital stay, prior operations before BDCPA, operations concomitant with the BDCPA, reoperations after BDCPA during the same hospital stay, medication before and after BDCPA and complications after BDCPA with a special focus on the rhythm anomalies. A pre- and postoperative set of data with oxygen saturation, pulmonary arterial pressure and systemic arterial pressure was obtained.

Arrhythmia-specific information was obtained from routine preoperative 12-lead electrocardiogram (ECG) and 24-h Holter monitoring before BDCPA and postoperatively including subsequent outpatient visits until Fontan completion. Intraoperative and early postoperative observations on the standard patient monitoring system were also included in the analysis. Postoperatively, a 12-lead ECG was routinely obtained on the first day and before hospital discharge. 24-h Holter monitoring was obtained in all patients before hospital discharge.

### Definitions

Arrhythmias are subclassified in sinus bradycardia, premature atrial/ventricular contractions, supraventricular tachycardia and

atrioventricular (AV) block. Sinus bradycardia is defined as a heart rate below age-adjusted normal values [15]. There were no patients with bradycardia/tachycardia syndrome or inadequate sinus tachycardia. AV block is separated into two groups: the lower grade AV block including first-degree AV block as well as second-degree AV block type Wenckebach (Mobitz I) and the higher grade AV block including second-degree AV block type Mobitz (Mobitz II) as well as complete AV block. Premature contractions, either of atrial or ventricular origin, were divided upon their frequency into none or rare (<240/24 h) and more frequent extrasystoles (>240/24 h) as seen in 24-h Holter monitoring.

Haemodynamic events/complications were subclassified in systemic arterial and pulmonary arterial hypertension, upper airway infection, pneumonia, pleural effusion, chylothorax, pericardial effusion, pneumothorax and death. Systemic arterial hypertension is defined as an increased blood pressure adjusted to age [16]. Postoperative pulmonary arterial hypertension is defined as a mean pressure of >15 mmHg measured in the SVC. We classified respiratory tract infection into pneumonia, defined by a consolidation in chest X-ray and upper airway infection. Prolonged fluid collection within the pleural space is classified according to the content of fat and white blood cells into chylous or non-chylous pleural effusion (cell count >1000  $\mu$ l with >80% lymphocytes and triglycerides >1.1 mmol/l = chylous effusion). Pneumothorax is noted as a complication, if the amount of air in the pleural space was more than minimal. Pericardial effusion is classified as a haemodynamically relevant fluid collection within the pericardium.

Intravenous inotropic support with adrenaline and norepinephrine is routinely administered within the first postoperative hours for the prevention and therapy of the postoperative low cardiac output syndrome. The evaluation of arrhythmias with Holter monitoring before hospital discharge was always done late after cessation of catecholamines.

### Operative technique and procedure

Through a median sternotomy, the SVC and the PA were dissected, and the circulation supported with a heart lung machine using aortobicaval cannulation. The SVC was transected 2 mm above its insertion into the right atrium, and the lower end was sutured with absorbable sutures. The cephalic end of the transected SVC was anastomosed (endothelium to endothelium) to the superior aspect of the right PA just near the right hilum, in an end-to-side fashion. Construction of the BDCPA was performed on an empty beating heart. Simultaneous procedures such as take down of a Blalock-Taussig (BT) shunt or a RV-PA (Sano) shunt were also performed on an empty beating heart. More complex procedures like transection of the main PA with suture closure of the pulmonary annulus, Damus-Kaye-Stansel procedure and/or atrial septectomy were performed during a period of cardioplegic arrest of the heart. Prior to pericardial closure, bipoles of atrial and ventricular temporary pacing wires were inserted. The pericardium was routinely closed, most often with a bridging polytetrafluoroethylene membrane, to protect the heart from injury during subsequent sternotomy.

### Statistical analysis

Continuous variables are expressed as median and range, categorical data as count and percentages. Significance analysis is made

by using Student's *t*-test or the Fischer's test where applicable. A *P*-value of <0.05 is considered the level of significance (\*\*<0.001, \*<0.05, (>0.05). All statistics were performed on the data with Microsoft Excel 2010 for Windows.

## RESULTS

### Study patients

The study population consists of 60 patients who underwent the BDCPA procedure. The patients' characteristics and diagnosis are summarized in Table 1. HLHS patients were operated on at an earlier age and the younger age group has longer times of CPB and hospital stay. The duration of mechanical ventilation, ICU stay and chest drain did not differ significantly between the various groups. The diagnoses and additional operations/interventions are summarized in Tables 2 and 3. Atrial situs was solitus in 55, right isomerism in 2 patients, left isomerism in 1, ambiguous in 1 and in-versus in 1. HLHS patients had less operations accompanying BDCPA (9 of 29 HLHS patients vs 20 of 31 non-HLHS patients, *P* < 0.011). Patients younger than 4 months had more reoperations during the same hospital stay (3 of 8 in patients <4 months and 4 of 52 in patients >4 months of age, *P* > 0.043). Neither the patient age nor the morphological groups (HLHS vs non-HLHS) were statistically associated with the occurrence of postoperative arrhythmia.

### Occurrence of arrhythmia

A new arrhythmia was observed in 20 patients (33%). In 12 patients, the arrhythmia was transient and resolved before hospital discharge. In 8 patients, the arrhythmia persisted until hospital discharge (Table 4).

Pre-existing arrhythmias were seen in 3 patients: 1 patient with sinus bradycardia, 1 patient with severe ventricular dysfunction and ectopic atrial tachycardia who received a single-chamber AAI pacemaker for amiodarone-induced bradycardia and 1 patient with a dual-chamber pacemaker for pre-existing complete AV block.

Sinus bradycardia was the most frequent arrhythmia observed in our study population. It occurred in 27% of the study population. It shows a trend to be present more often in the older group and in the non-HLHS patients late postoperatively. There was no patient with a higher degree AV block as a consequence of the BDCPA operation and only 1 patient with a newly onset first-degree AV block.

Supraventricular tachycardia was seen in 3 patients during ICU stay (JET in 2 patients and AVRT in 1). All these supraventricular tachycardias did not persist until hospital discharge. Ventricular arrhythmias were not observed. Sinus bradycardia was mild and of no clinical consequence. None of these patients needed a medication or a pacemaker implantation to accelerate the heart rate.

The distribution of the temporary and persisting postoperative arrhythmias are given in Table 4. At hospital discharge, sinus bradycardia is the most common arrhythmia. No significant difference was seen between patients with and those without persistent arrhythmias regarding additional interventions (Table 3), complications and medication.

At the time of Fontan completion, 2 of the 8 patients with persistent arrhythmias showed a persistence of the same kind of arrhythmia as after BDCPA with no clinical relevance (sinus bradycardia in 2 patients and first-degree AV Block in 1).

**Table 2:** Diagnosis in all 60 patients with BDCPA (number of patients)

		Age at operation	
		<4 months	>4 months
HLHS and HLHS variants	29	5	24
Non-HLHS			
DILV	8	0	8
VSD/DORV	8	3	5
Tricuspid atresia	7	0	7
Pulmonary atresia	6	0	6
AVSD	2	0	2

DILV: double inlet left ventricle; VSD: ventricular septum defect; DORV: double outlet right ventricle; AVSD: atrioventricular septum defect.

**Table 1:** A summary of patients' characteristics of all the 60 patients with BDCPA

	Total	Age at operation		Diagnostic group	
		<4 months	>4 months	HLHS	non-HLHS
Number of patients	60	8	52	29	31
Age (months)	5.2 (1-12)	2.7 (1-3)	5.4 (4-12)*	4.9 (1-8)	5.8 (2-12)**
Weight (kg)	5.7 (2.9-8.2)	4.2 (2.9-5.3)	6.0 (3.7-8.2)*	5.3 (2.9-7.9)	6.2 (3.7-8.2)**
Preoperative oxygen saturation (%)	77 (60-90)	80 (70-90)	77 (60-89)	77 (70-89)	78 (60-90)
Cardiopulmonary bypass (min)	135 (66-285)	236 (81-283)	133 (66-285)**	137 (66-283)	133 (82-285)
Mechanical ventilation (days)	3 (1-49)	5 (2-49)	2 (1-15)	2 (1-49)	3 (1-14)
Duration of chest tube (days)	4 (2-69)	6 (3-22)	4 (2-69)	4 (2-69)	4 (2-22)
Intensive care unit stay (days)	6 (3-76)	16 (4-38)	6 (3-76)	7 (3-76)	6 (3-52)
Length of hospital stay (days)	19 (11-166)	79 (15-166)	18 (11-119)**	19 (12-151)	19 (11-166)

Figures are given as the number of patients and median (range), respectively, for the whole patient collectively and for age and diagnosis subgroups individually; significance codes for comparison between the subgroups.

\**P*<0.001 and \*\**P*<0.05.

HLHS: hypoplastic left heart syndrome.

**Table 3:** Patients with cardiothoracic surgical operations/interventions in addition to the BDCPA and type of additional interventions (number of patients)

		All
Previous operations (57)	Pulmonary artery banding	22
	Norwood I	20
	Modified Blalock-Taussig shunt	9
	Central shunt	5
	Coarctation of the aorta	12
	Damus-Kaye-Stansel operation	2
	Repair of TAPVR <sup>a</sup>	2
Operations concomitant with the BDCPA (29)	Atrial septectomy	19
	Damus-Kaye-Stansel operation	6
	Damus-Kaye-Stansel operation + reconstruction of the aortic arch	4
	Modified Blalock-Taussig shunt	1
	Central shunt	2
	Pulmonary artery banding	1
	Implantation of a pacemaker for amiodarone-induced bradycardia	1
Reinterventions during same hospital stay (7)	Operation	5
	Cardiac catheter	2

<sup>a</sup>TAPVR: total anomalous pulmonary vein return.

**Table 4:** Distribution of relevant temporary and persistent postoperative arrhythmias, premature atrial/ventricular complexes excluded (number of patients = 60)

	Temporary arrhythmias	Persistent arrhythmias
Total	12	8
Sinus bradycardia	9	7
Atrioventricular block first-degree	2	1
Atrioventricular block higher degree	1	0
Atrioventricular re-entrant tachycardia	1	0
Junctional ectopic tachycardia	2	0

Patients are possibly multiple mentioned.

Premature contractions were rarely seen. In the late postoperative Holter monitorings before hospital discharge, there was only 1 patient with rare premature ventricular and 4 patients with rare premature atrial contractions. All of them were singular and haemodynamically irrelevant.

## Complications

Major postoperative complications were rare. One patient had a circulatory failure after BDCPA during ICU stay due to a previously unknown thrombosis of the innominate vein. This is the only patient of the study population who died. Minor postoperative complications (upper airway infection, pneumonia, inadequate systemic and pulmonary arterial hypertension, pleural and pericardial effusion and pneumothorax) occurred in 41 of all 60 patients. The outcome regarding in-hospital complications other

than arrhythmia was similar in the subgroups age and diagnosis. The occurrence of arrhythmia was independent of the occurrence of other complications.

## Medication

The following medications with a potential effect on the heart rhythm were necessary in 7 patients during hospital stay and at hospital discharge: Digoxin in 5 patients, beta adrenergic receptor blocking agents in 2 and amiodarone in 2. A proarrhythmic effect was not observed in our study population.

## DISCUSSION

In our study population, we have not seen any severe and life-threatening arrhythmias after BDCPA. Most of the observed arrhythmias were temporary and resolved during the hospital stay. In view of the complexity of the surgical intervention during BDCPA, the low incidence of arrhythmia is reassuring. Also age was not a risk factor for the occurrence of arrhythmia. Therefore, the time to perform the BDCPA is not limited by the occurrence of postoperative arrhythmia but can be adjusted to the individual haemodynamic situation of the patient.

Postoperative junctional ectopic tachycardia (JET) is described to occur with an incidence of up to 9.5% in a similar population [17]. It was observed in our population in 2 patients, resulting in an incidence of only 3%. In general, JET is not well tolerated in the early postoperative hours because of the decreased diastolic ventricular filling due to the high heart rate and to the loss of AV synchrony. It may result in progressive low cardiac output syndrome and therefore needs aggressive therapy with supportive measures and eventually amiodarone. The arrhythmia itself is known to resolve within 72 h postoperatively. Also both patients from our cohort returned to sinus rhythm within the first postoperative days.

Supraventricular tachyarrhythmias such as intra-atrial re-entrant tachycardia, ectopic atrial tachycardia and atrial fibrillation which are typically seen in the long-term follow-up in adults with Fontan circulation [5] did not occur in our study cohort. Long-term haemodynamic stress and multiple atrial suture lines therefore seem to be more important in the genesis of these arrhythmias than the initial surgical trauma during BDCPA.

The most frequent late postoperative arrhythmia after BDCPA is bradycardia due to sinus node dysfunction. It has been previously reported to occur in 6–10% [7, 8] and was observed in our study population in a similar incidence (12%). The sinus node dysfunction is interpreted as a consequence of tissue trauma by the suture line close to the region of the sinus node or its artery. Surprisingly, the avoidance of surgery near the sinus node seems not to have a discernable effect on the development of early sinus node dysfunction which has been shown in a study comparing patients with bidirectional Glenn anastomosis and Hemi-Fontan procedure [7].

As seen in our study population, sinus bradycardia is clinically irrelevant and was of no therapeutic consequence in short-term follow-up. The incidence of sinus node dysfunction increases after the Fontan completion to 20–25% due to repeated dissection in the region of the SA node [8, 18]. In long-term follow-up, however, sinus node dysfunction is a troublesome complication after Fontan completion. It is a known risk factor for the later development of intra-atrial re-entrant tachycardia [19] and may require

the implantation of a permanent pacemaker to treat significant bradycardia.

## LIMITATIONS

The conclusions of this study are limited by its retrospective design and the relatively small number of patients.

**Conflict of interest:** none declared.

## REFERENCES

- [1] Alejos JC, Williams RG, Jarmakani JM, Galindo AJ, Isabel-Jones JB, Drinkwater D *et al.* Factors influencing survival in patients undergoing the bidirectional Glenn anastomosis. *Am J Cardiol* 1995;75:1048–50.
- [2] Hansen JH, Uebing A, Furck AK, Scheewe J, Jung O, Fischer G *et al.* Risk factors for adverse outcome after superior cavopulmonary anastomosis for hypoplastic left heart syndrome. *Eur J Cardiothorac Surg* 2011;40:43–9.
- [3] Kogon BE, Plattner C, Leong T, Simsic J, Kirshbom PM, Kanter KR. The bidirectional Glenn operation: a risk factor analysis for morbidity and mortality. *J Thorac Cardiovasc Surg* 2008;136:1237–42.
- [4] Balaji S, Silja MJ, McAnulty JH. Arrhythmias in patients with congenital heart disease. *CEPR* 2002;6:42–4.
- [5] Walsh EP, Cecchin F. Arrhythmias in adult patients with congenital heart disease. *Circulation* 2007;115:534–45.
- [6] Weindling SN, Saul JP, Gamble WJ, Mayer JE, Wessel D, Walsh EP. Duration of complete atrioventricular block after congenital heart disease surgery. *Am J Cardiol* 1998;82:525–7.
- [7] Cohen MI, Bridges ND, Gaynor JW, Hoffman TM, Wernovsky G, Vetter VL *et al.* Modifications to the cavopulmonary anastomosis do not eliminate early sinus node dysfunction. *J Thorac Cardiovasc Surg* 2000;120:891–901.
- [8] Manning PB, Mayer JE, Wernovsky G, Fishberger SB, Walsh EP. Staged operation to Fontan increases the incidence of sinoatrial node dysfunction. *J Thorac Cardiovasc Surg* 1996;111:833–40.
- [9] Chang AC, Hanley FL, Wernovsky G, Rosenfeld HM, Wessel DL, Jonas RA *et al.* Early bidirectional cavopulmonary shunt in young infants. Postoperative course and early results. *Circulation* 1993;88:149–58.
- [10] Jaquiss RD, Ghanayem NS, Hoffman GM, Fedderly RT, Cava JR, Mussatto KA *et al.* Early cavopulmonary anastomosis in very young infants after the Norwood procedure: impact on oxygenation, resource utilization, and mortality. *J Thorac Cardiovasc Surg* 2004;127:982–9.
- [11] Jaquiss RD, Siehr SL, Ghanayem NS, Hoffmann GM, Fedderly RT, Cava JR *et al.* Early cavopulmonary anastomosis after Norwood procedure results in excellent Fontan outcome. *Ann Thorac Surg* 2006;82:1260–6.
- [12] Petrucci O, Khoury PR, Manning PB, Eghtesady P. Outcomes of the bidirectional Glenn procedure in patients less than 3 month of age. *J Thorac Cardiovasc Surg* 2010;139:562–8.
- [13] Reddy VM, Liddicoat JR, Hanley FL. Primary bidirectional superior cavopulmonary shunt in infants between 1 and 4 months of age. *Ann Thorac Surg* 1995;59:1120–6.
- [14] Reddy VM, McElhinney DB, Moore P, Haas GS, Hanley FL. Outcomes after bidirectional cavopulmonary shunt in infants less than 6 months old. *J Am Coll Cardiol* 1997;29:1365–70.
- [15] Davignon A, Rautaharju P, Boiselle E, Soumis F, Mégélas M, Chouquette A. Normal ECG standards for infants and children. *Ped Cardiol* 1979;1:123–31.
- [16] Park MK, Menard SM. Normative oscillometric blood pressure values in the first 5 years in an office setting. *Am J Dis Child* 1989;143:860–4.
- [17] Zampi JD, Hirsch JC, Gurney JG, Donohue JE, Yu S, LaPage MJ *et al.* Junctional ectopic tachycardia after infant heart surgery: incidence and outcomes. *Pediatr Cardiol* 2012;33:1362–9.
- [18] Cohen MI, Wernovsky G, Vetter VL, Wieand TS, Gaynor JW, Jacobs ML *et al.* Sinus node function after a systematically staged Fontan procedure. *Circulation* 1998;98:352–9.
- [19] Fishberger SB, Wernovsky G, Gentles TL, Gauvreau K, Burnett J, Mayer JE *et al.* Factors that influence the development of atrial flutter after the Fontan operation. *J Thorac Cardiovasc Surg* 1997;113:80–6.