ORIGINAL CONTRIBUTION

MODIFIED FONTAN PROCEDURE WITH THE SIMULTANEOUS BIDIRECTIONAL CAVOPULMONARY SHUNT

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ABSTRACT

We reviewed our experience of 56 patients from 1989 to 1992 who underwent a modified Fontan procedure and a bidirectional cavopulmonary shunt simultaneously. There were 39 male and 17 female patients and their weight ranged from 6.54 to 29kg (mean weight 13.58 ± 3.96kg). Patient age ranged from 16 to 135 months (mean age 42.8 ± 3.7 months). Diagnoses included single ventricle in 29, tricuspid atresia in 11, double outlet of right ventricle in 10, hypoplastic left heart syndrome in 4, and pulmonary atresia with intact ventricular septum in 2 patients. The techniques of inferior vena cava to pulmonary artery (IVC-PA) connection were anastomosis of proximal superior vena cava (SVC) to pulmonary artery (PA) in 27 (group 1), direct atriopulmonary anastomosis with roof formation in 29 patients (group 2). There were significant differences in postoperative 1-hour right atrial (RA) pressure and period of chest tube drainage between group 1 and group 2. The early mortality was 12.5% (7/56), and 2 late deaths (4.1%) occurred with a mean follow-up period of 22.4 months. Risk factors for the late postoperative arrhythmia were immediate postoperative arrhythmia and prolonged pleuro-pericardial effusion. Direct connection of the remaining proximal SVC to PA with the bidirectional cavopulmonary shunt may have less pleuro-pericardial effusion and late arrhythmia than atriopulmonary anastomosis.

(Asian Cardiovasc Thorac Ann 1995;3:29-34)

INTRODUCTION

In 1971 Fontan and Baudet[1] reported the first clinically successful procedure for total bypass of the right heart as a treatment of tricuspid atresia. Since their attempt, many other modifications, including Kreutzer and coauthors modification[2] were introduced for the variety of complex congenital heart lesions.

The bidirectional cavopulmonary shunt may be defined as an operation that diverts the systemic venous return from the superior vena cava (SVC) or cavae to both lungs. The cavopulmonary shunt usually consists of an end-to-side anastomosis between the SVC and the undivided pulmonary artery (PA). Variations include the anastomosis of bilateral venae cavae to both pulmonary arteries and the total cavopulmonary shunt type of operation[3] and others[4,5]. The cavopulmonary shunt approach is a useful adjunct to completion of Fontan-type repairs[6].

We assessed postoperative hemodynamics in simultaneous bidirectional cavopulmonary shunt during the Fontan procedure. To assess the possible benefits of such an approach, we reviewed the case histories of 56 patients who underwent the modified Fontan procedure with bidirectional cavopulmonary shunt simultaneously from 1989 to 1992. This group made up an increasing percentage of all patients undergoing Fontan procedures at our hospital during this period, constituting 70% of all Fontan-type procedures we performed in the same period.

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METHOD

STUDY PATIENTS.

Clinical and hemodynamic data were drawn from 56 patients from 1989 to 1992 who had a modified Fontan operation and simultaneous bidirectional cavopulmonary shunt. Thirty-nine patients were male and 17 patients were female. Patient ages at operation ranged from 16 months to 165 months, with a mean of 42.8 ± 3.7 months. Of these 56 patients, 10 were between 1 and 2 years old, 24 were between 2 and 3 years old, 8 were between 3 and 4 years old, and 14 were over 4 years old. Patient weight at the operation ranged from 6.54 to 29kg, with a mean of 13.58 ± 3.96kg. The largest group included 29 patients (52%) with single ventricle. Ventricular morphology was like right ventricle in 28, and like intermediate type ventricle in 1 patient. Eleven patients (19%) had tricuspid atresia (TA): TA (1a) in 1, TA (1b) in 6, TA (2b) in 4 patients. Ten patients (18%) had double outlet right ventricle. Six patients had hypoplastic left ventricle (LV), 3 patients had endocardial cushion defect (ECD), and 1 patient had severe mitral valve straddling. Four patients (7%) had hypoplastic left heart syndrome, and 2(4%) had pulmonary atresia with intact ventricular septum. Thirty patients underwent 1 or more prior palliative procedures (Table 1). Anomalous systemic venous drainage (Table 2) was recorded when there existed systemic venous connections to the heart other than right-sided superior and inferior venae cavae (IVC)(or left-sided in the cases of atrial situs inversus). Anomaly of pulmonary venous drainage was considered to be present when any or all of the pulmonary veins were connected to the sites other than the morphological left atrium. Atrioventricular (AV) valve regurgitation was graded as mild, moderate, or severe according to the angiogram.

TABLE 1. PREVIOUS PALLIATIVE OPERATIONS

RMBT		10
LMBT		11
+PDA ligation	2	
RMBT+LMBT		2
PA banding		6
+Correction of TAPVC	3	
+Atrial septectomy	2	
PDA ligation		1
Total		30

RMBT = right modified Blalock-Taussig, LMBT = left modified Blalock-Taussig, PDA = patent ductus arteriosus, PA = pulmonary artery, TAPVC = total anomalous pulmonary venous connection

TABLE 2. ANOMALOUS SYSTEMIC VENOUS DRAINAGE

Bilateral SVC		29
Interruption of IVC	4	
Abnormal drain of hepatic vein	1	
Abnormal drain of IVC to LA	1	
Interruption of IVC		3
Abnormal drain of hepatic vein		1
Total		33

SVC = superior vena cava, IVC = inferior vena cava, LA = left atrium

INTRAOPERATIVE VARIABLES.

All operations were performed with hypothermic cardiopulmonary bypass (CPB) and hemodilution. CPB time was taken as the total time of extracorporeal circulatory support, including circulatory arrest time (when it was used).

All patients had bidirectional cavopulmonary shunts. The SVC was divided at the portion proximal to the cavoatrial junction, and an end-to-side anastomosis was performed between the cephalic end of SVC and the superior aspect of the right PA. When bilateral SVC were present, the procedures were done in both sides.

Several techniques were used for separating the systemic venous return to the heart from the pulmonary venous return. The term "lateral tunneling" was used to describe a technique in which a tubular pathway was created from the ICV orifice to the SVC orifice with a prosthetic tube graft to form the medial wall of this tunnel. The lateral wall of the atrium formed the lateral wall of the pathway[7]. The term "septation" was used to describe a conventional septation patch constructed from a flat piece of prosthetic material (usually Dacron or Gore-Tex patch) to place the atrial septal defect in continuity with the tricuspid valve.

Several techniques were used for the connections between the remaining systemic venous return (mainly from IVC and hepatic veins) and the pulmonary arteries. Patients were grouped by these techniques. In group 1, the superior vena caval orifice of proximal SVC (previously divided for bidirectional shunt) was anastomosed to the inferior aspect of the pulmonary artery (proximal SVC-PA group). In group 2, the superior vena caval orifice of proximal SVC was closed with continuous sutures. The PA was so fully mobilized that the open distal end of the pulmonary trunk could be passed rightward beneath the ascending aorta. Direct atriopulmonary anastomosis was performed at the right atrial appendage or right atrial roof with a roof of autologous pericardial or Gore-Tex patches.

STATISTICAL ANALYSIS

Statistical analysis included Fisher's exact test, chi-square test of association for comparing proportions, general linear model (GLM), and logistic regression analysis for assessing multivariate relations. For the survival analysis, the date of the Fontan operation was used as time 0. Early death was defined as death during the first 30 days. SAS 6.04 statistical software system was used throughout. Only 2-tailed *P*-values were used. When incomplete numeric data were present, the missing value indicators were used to allow evaluation of variables.

We evaluated risk factors affecting survival and late arrhythmia. The variables included patient age less than 3 years at operation, abnormal systemic venous drainage, abnormal pulmonary venous drainage, moderate or severe insufficiency of atrioventricular valve, preoperative medi-

cation for treatment of heart failure, preoperative absence of sinus rhythm, immediate postoperative arrhythmia, postoperative 1-hour RA pressure, chest tube drainage greater than 20 days, duration of postoperative inotropic support, techniques of atrial partitioning, and techniques of connection to pulmonary artery (by groups).

RESULTS

There were 7 early deaths (12.5%, 7/56). One operative death occurred 3 hours after operation. The patient had an episode of cardiac arrest before the operation. The operation was performed after resuscitation. There was difficulty in weaning cardiopulmonary bypass and in spite of the maximal support of inotropics the patient died of ventricular failure. The second early death occurred after an episode of accidental respiratory arrest after early extubation on postoperative day 2. The patient was resuscitated but suffered a severe neurological insult and died on postoperative day 7. The 3rd early death occurred after an episode of ventricular tachycardia and cardiac arrest on postoperative day 2. Three other patients died of persistent low cardiac output and cardiac arrest on postoperative day 1, 8, and 10 respectively. The patients also suffered acute renal failure and hypoxia brain damage. Another patient died 4 weeks later. The patient had an intracardiac thrombosis postoperatively requiring antithrombotic therapy. There was also sign of increased intracranial pressure probably due to the impaired intracranial circulation. He suffered low cardiac output, acute renal failure, ongoing sepsis, and multisystem organ failure.

All patients were treated with inotropic agents in the immediate postoperative period. Mean duration of the inotropic support (dopamine or dobutamine) was 4.6 ± 4.2 days (range 0 to 26 days). Maximum dose of inotropics (dopamine + dobutamine) was 12 ± 8.3μg/kg/min (range 2 to 35μg/kg/min). No patients required intraaortic balloons or ventricular assist devices. Mean time to extubation was 3.2 ± 2.7 days (range 1 to 12 days). Mean duration of chest tube drainage was 19.7 ± 20.9 days (range 3 to 109 days). The average right atrial pressure measured at 1 hour postoperative was 16.7 ± 4.4mmHg (range 9.5 to 29.3mmHg) and the average left atrial (LA) pressure was 13.1 ± 3.4mmHg (range 4.4 to 22.3mmHg). The average length of hospitalization was 28.8 ± 25.7 days (range 10 to 142 days). The perioperative hemodynamic and clinical data were compared between group 1 and group 2 (Table 3, 4). There were significant differences in duration of chest tube drainage and in average postoperative 1-hour right atrial pressure (RAP-1h). There were no significant differences in other variables.

Prolonged pleural effusion (more than 20 days) developed in 11 patients. Two patients had low cardiac output and suffered transitory acute renal failure. Five patients underwent additional operations in the immediate postoperative periods: plication of the diaphragm due to phrenic nerve palsy (2), pericardiostomy due to cardiac tamponade caused by serious pericardial effusion (1), debridement and irrigation due to the mediastinitis (1), exploration due to postoperative bleeding (1). Two patients had minor wound problems. Early postoperative rhythm analysis of survivals revealed normal sinus rhythm in 36, permanent pacing rhythm in 5, 1 degree AV node block in 4, paroxysmal supraventricular tachycardia (PSVT) in 4 patients. Five patients underwent pacemaker insertion simultaneously with other procedures due to the malignant preoperative arrhythmias.

Follow-up periods ranged from 2.2 to 53.5 months; mean follow-up was 22.3 months. There were 2 late deaths. One patient had recurrent PSVT attacks with progressively worsening ventricular function. He died at 10 weeks postoperatively. A second patient had a poorly functioning pacemaker. The pacemaker had functioned well at the

TABLE 3. PREOPERATIVE CLINICAL DATA

Variable	Overall	Groups Group 1	Group 2	*P*-value
Diagnosis (*n*)				0.50
Single ventricle	29	15	14	
Tricuspid atresia	11	7	4	
DORV	10	3	7	
Hypoplastic left heart	4	1	3	
Pulmonary atresia	2	1	1	
Sex (male:female)	39:17	19:8	20:9	0.91
Age (month)	42.7	48.3	37.1	0.11
Body weight (kg)	13.6	14.6	12.8	0.27
Hematocrit (%)	53.2	54.0	52.1	0.23
Systemic vein anomaly (*n*)	33	15	18	0.29
Preoperative arrhythmia (*n*)	7	4	3	0.24
AV valve regurgitation				0.10
Mild	6	2	4	
Moderate	17	12	5	
Severe	9	2	7	

(n) = number, DORV = double outlet of right ventricle, AV = atrioventricular

TABLE 4. POSTOPERATIVE CLINICAL DATA

Variable	Overall	Groups Group1	Group 2	*P*-value
Inotropic support duration (d)	4.2	4.3	4.1	0.54
Maximum dose of inotropic (μg)	12	10.8	12.2	0.8
Duration of pleural effusion (d)	19.7	14	26.4	0.04
Immediate postoperative arrhythmia (*n*)	13	7	6	0.88
Average RAP 1 hour postop (mmHg)	16.7	15.0	18.4	0.003
Duration of hospital stay (d)	28.8	23	34.9	0.11

(d) = days inotropics: dopamine or dobutamine, (n) = number, RAP = right atrial pressure

immediate postoperative period. Holter monitoring revealed that function of the pacemaker was poor and there was ventricular tachycardia 28 months postoperatively. Another pacemaker insertion was planned, but the patient had an unexpected ventricular tachycardia attack and cardiac arrest before pacemaker insertion.

Twenty-nine patients were in normal sinus rhythm (Table 5). The rhythms of 7 patients had changed during follow-up periods from sinus rhythms to arrhythmias (2 with junctional escape beats, 2 with PVCs, 2 with SA node dysfunctions, and 1 with atrial flutter). One patient with PSVT from the immediate postoperative period underwent pacemaker insertion at the postoperative 10th week. Leakage of the septation patch was confirmed in 1 patient. The leakage was not severe and the patient had been under observation. Two patients needed reoperation for AV valve regurgitation. The results of the multivariate analysis of stepwise logistic procedure are shown in Table 6. In this model, the immediate postoperative arrhythmia and prolonged chest tube drainage were recognized as the important variables for late arrhythmia.

DISCUSSION

Bidirectional cavopulmonary shunt improves systemic arterial oxygen saturation without increasing ventricular work or pulmonary vascular resistance. Bidirectional cavopulmonary shunt is defined as an operation that diverts the systemic venous return from the superior vena cava or cavae to both lungs. It is well known that cavopulmonary shunt does not increase the ventricular work and pulmonary vascular resistance, and does not distort the pulmonary arteries[6]. When the bidirectional cavopulmonary shunt is used as an adjunct to Fontan procedure, the term total cavopulmonary anastomosis[7-8] is usually used and described as the lateral tunnel technique. We use the term simultaneous bidirectional cavopulmonary shunt with Fontan operation to include all the Fontan procedures with cavopulmonary shunt regardless of the separation techniques. Also, it is different from total cavopulmonary shunt[3] in that the hepatic blood flow is included in the pulmonary circulation.

TABLE 5. LATE RHYTHM CHANGES

Preoperative Rhythm		Late Rhythm		
Sinus	42	Sinus	29	
		Arrhythmia	13(1)	
		PVC		2
		Junctional escape beats		2
		PSVT		3(1)
		AV node block (1 degree)		2
		SA node dysfunction		2
		Atrial flutter		1
		Pacing rhythm		1
Benign Arrhythmia	2	Arrhythmia	2	
		AV node block (1 degree)		2
Malignant Arrhythmia	5	Pacing rhythm	5(1)	
Total			49(2)	

() = Late death, PVC = premature ventricular contraction, PSVT = paroxysmal supraventricular tachycardia, AV = atrioventricular, SA = sinoatrial

TABLE 6. RISK FACTORS FOR LATE ARRHYTHMIA

Summary of stepwise logistic procedure

Variables	*P*-value
Immediate postoperative arrhythmia	0.0015
Chest tube drainage	0.0164

Variables not in the model

Variables	*P*-value
Age < 3 years at operation	0.21
Anomalous systemic venous drainage	0.73
Pulmonary artery distortion	0.74
AV valve regurgitation	0.78
History of preoperative medication	0.98
Preoperative absence of sinus rhythm	0.21
Techniques of atrial partitioning	0.85
Techniques of RA-PA connection	0.50
Postoperative 1-hour RAP	0.41

AV = atrioventricular, RA = right atrium, PA = pulmonary artery, RAP = right atrial pressure

We initially expected there would be benefits in the postoperative hemodynamics with the simultaneous bilateral cavopulmonary shunt. The early mortality (12%) is a little higher (7 to 10.5%) than that of recent reports[8,10-12]. With improvements in surgical technique and postoperative care, early mortality for Fontan procedures has steadily declined[8,10,12,13]. Our data also show the yearly decline of early mortality. In 1992, there was only 1 early death (n = 21,4.7%).

A number of techniques have been proposed for the creation of atriopulmonary connections for the Fontan operation[14-17]. Most methods use the right atrial appendage or right atrial roof for the site of the anastomosis, and we also used these sites for anastomosis until 1989. Since 1989, we occasionally used the SVC for the anastomosis, especially with the simultaneous bidirectional cavopulmonary shunt in the cases of large SVC or bilateral SVC, and recently we usually selected this site for anastomosis because of the increasing use of the lateral tunneling technique.

We expected the proximal SVC-PA technique would result in a lower incidence of atrial arrhythmias and more improved hemodynamics. We believed the avoidance of postoperative arrhythmia and improved hemodynamics with lower systemic venous pressure might result in a lower mortality and an improved late outcome. There seemed to be significant difference of early mortality between group 1 and group 2, but with the consideration of the partitioning technique and time factor, it was deemed to be insignificant. There was no mortality with the lateral tunneling technique. With the septation technique, the early mortality rate was 11.1% (1/9) in group 1 and 20.7%

(6 /29) in group 2 (P = 0.51). The early mortality was 16.7% (1/6) in 1989, 21.43% (3/14) in 1990, 13.3% (2/15) in 1991, and 4.7% (1/21) in 1992. There was no significant difference between group 1 and group 2 when the early mortality was compared yearly (P = 0.44 in 1989, 0.43 in 1990, 0.28 in 1991, 0.67 in 1992). Pearl and associates[8] reported that early mortality in the lateral tunnel group is not significantly different than conventional Fontan group; it is low with both procedures.

With the proximal SVC-PA technique, we also expected to reduce the central venous pressure postoperatively and the stimulus for atrial natriuretic peptide (ANP) production, decreasing the incidence of pleuropericardial effusion. Several reports demonstrated that patients after Fontan operation had higher ANP levels[18,19]. Anderson and associates[19] demonstrated that ANP decreased both the rate and force of contractions of the thoracic duct. We also speculated the flow direction of proximal SVC-PA technique would reduce atrial turbulence, improving hemodynamics like the lateral tunneling technique. The postoperative 1-hour RAP of group 1 is significantly lower than group 2, but there are no differences in the postoperative 3-hour and 24-hour RAP. The duration of chest tube drainage was significantly different between the groups. However, the incidence of prolonged pleural effusions (4 in group 1, 7 in group 2) was not significantly different (P -value 0.51). Interestingly, when the variables were compared on the basis of the partitioning techniques, the postoperative 1-hour, 3-hour and 24-hour right atrial pressures were significantly different, but the duration of the chest tube drainage and the incidence of the prolonged pleural effusions were not different. Similar findings were reported by others [8,20].

The electrophysiologic findings after Fontan repair include abnormal sinus node function, prolonged atrial refractoriness, delayed intraatrial conduction and inducible atrial arrhythmia[21]. The development of atrial arrhythmias as sick sinus syndrome and AV node block in the conventional modified Fontan patients may result from the extensive suture lines of atriotomy and intraatrial baffle, with resultant atrial scarring and fibrosis[8,9]. Proximal SVC-PA technique has the potential for sinus node dysfunction because the areas of transection of the vena cava and the necessary suture lines are in the vicinity of the sinus node and the sinus node artery[17].

Immediate postoperative arrhythmias developed in a similar number of patients in both groups. However, the nature of the arrhythmia was different. In group 2, the incidence of paroxysmal supraventricular tachycardia (PSVT) attack was more frequent. It was thought that it was related to the difference of the central venous pressure between group 1 and group 2. The combination of prolonged atrial conduction time and right atrial enlargement may increase the possibility of re-entrant atrial arrhythmias like PSVT[21].

For late arrhythmia, the variables of immediate postoperative arrhythmia and prolonged chest tube drainage were recognized as important factors in multivariate analysis. Once an arrhythmia appeared in the early postoperative period, the arrhythmia did not disappear and, furthermore, usually became a serious complicating factor. Although the relation between late arrhythmia and prolonged pleural effusion cannot be defined precisely, some common factor may exist. High central venous pressure and extensive atrial suture line can be avoided by proximal SVC-PA technique.

It seemed there were improved late results with proximal SVC-PA technique. There were 2 late deaths in group 2 and no late deaths in group 1. Late onset arrhythmias developed in a similar incidence. In both groups, all patients had the manipulation of the SVC and risk of sinus node injury during manipulation was not avoided. However, the need of pacemaker placement during follow-up was different. In group 2, 1 patient died of malignant PSVT and another underwent pacemaker insertion due to the malignant PSVT. For the patient with PSVT in group 1, the hemodynamics were stable, and the attacks were infrequent. Although our results suggest an improved late outcome with proximal SVC-PA technique, follow-up is short and further follow-up is necessary to clearly reveal this advantage.

This study does have limitations from a statistical viewpoint. Firstly, the number of patients in this study is relatively small. Additional factors might have been identified as significant risk factors if there were many patients. Secondly, this study is a retrospective analysis. Patient management has continuously evolved during the period of the study. Some factors may have been overlooked. Thirdly, some data are incomplete so some variables are not included in the analysis.

CONCLUSION

Modified Fontan procedure with the simultaneous bidirectional cavopulmonary shunt can be done with relative technical ease. Although the outcome of the procedure seems to be similar to that of the conventional modified Fontan procedure, the procedure is a useful adjunct to Fontan-type repairs, especially in the complicating anomalies. Direct connection of the remaining SVC to PA after the bidirectional cavopulmonary shunt may have less pleuropericardial effusion, fewer late pacemaker insertions, and a lower late mortality rate than direct atriopulmonary connection.

This paper was presented at the 2nd Annual Meeting of the Asian Society for Cardiovascular Surgery, *Nagoya, Japan, February 11, 1994.*

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