CONVERSION OF MODIFIED FONTAN PROCEDURE TO LATERAL ATRIAL TUNNEL CAVOPULMONARY ANASTOMOSIS

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After modified Fontan procedures with atriopulmonary anastomoses or right atrium–right ventricle conduits, some patients have progressive exercise intolerance, effusions, arrhythmias, or protein-losing enteropathy. Theoretic advantages of a lateral atrial tunnel cavopulmonary anastomosis and published clinical results suggest that conversion of other Fontan procedures to the lateral atrial tunnel may afford clinical improvement for some patients. Eight patients (8 to 25 years old) with tricuspid atresia ($n = 4$), double-inlet left ventricle ($n = 3$), and double-outlet right ventricle ($n = 1$) underwent conversion to a lateral tunnel procedure between December 1990 and November 1994. An arbitrary clinical score was assigned before the lateral tunnel procedure and at follow-up. Before conversion, patients had decreased exercise tolerance ($n = 8$), arrhythmias ($n = 6$), effusions ($n = 4$), and protein-losing enteropathy ($n = 8$). At catheterization, all had a low cardiac index ($1.9 \pm 0.7 \text{ L} \cdot \text{min}^{-1} \cdot \text{m}^{-2}$), five had elevated pulmonary vascular resistance (>3 Wood units), and three had right pulmonary venous return obstruction by compression of an enlarged right atrium. Fenestrated lateral tunnel construction was undertaken 7.3 ± 3.6 years after atriopulmonary anastomosis, with one early death related to low cardiac output. After the lateral tunnel procedure, two patients had no clinical improvement (no change in clinical score) but five patients had either marked or partial improvement. The right pulmonary vein compression present in three patients was resolved after conversion. The mean clinical scores improved from 4.5 ± 1 to 3.0 ± 2 ($p < 0.04$). In conclusion, conversion to a lateral tunnel procedure led to clinical improvement in five of eight patients at short-term follow-up and may be particularly indicated for patients with giant right atria or pulmonary vein compression who have symptoms. Pulmonary vein compression should be looked for in patients after modified Fontan procedures and can be relieved by conversion to the lateral tunnel procedure. (*J Thorac Cardiovasc Surg* 1996;111:1169-76)

After modified Fontan procedures, such as right atrium–right ventricle conduit or atriopulmonary anastomosis (APA), some patients have progressive exercise intolerance, atrial arrhythmias, chronic effusions, or protein-losing enteropathy.¹⁻⁶ The non-

lamellar venous blood flow that results from the postsurgical anatomy may cause energy loss, as suggested by experimental studies,^{7,8} and it may result in low cardiac output and development of symptoms. In addition, markedly enlarged right atria may also compress the pulmonary veins, causing elevated resistance to pulmonary blood flow.

Few therapeutic options have been considered for patients with such problems. Cardiac catheterization may identify anatomic or hemodynamic abnormalities in their circulation, some of which can be corrected either by transcatheter techniques or surgically.¹⁰⁻¹² Heart transplantation is often a consideration,¹³⁻¹⁵ particularly if ventricular dysfunction is present. When ventricular function is preserved, conversion to a lateral tunnel Fontan procedure

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Table I. Clinical score

Clinical feature	0 points	1 point	2 points
Exercise tolerance	No limitations (NYHA class I)	Slight limitation (NYHA class II)	Severe limitation (NYHA class III or IV)
Arrhythmias	None on no medications	None on medications or after RFA	Arrhythmias on medications or after RFA
Effusions	None	Intermittent	Chronic
PLE	No		Yes

NYHA, New York Heart Association congestive heart failure classification; RFA, radiofrequency ablation; PLE, protein-losing enteropathy.

(LT) has been recently proposed,⁹ although indications and possible predictors of success for such a procedure are unknown. The purpose of this report is to describe our experience with eight patients with marked symptoms who underwent conversion from atriopulmonary or atrioventricular Fontan operations to LT.

Methods

Patient population. Before 1989, 245 patients at our institution underwent modified Fontan procedures with APA or right atrium–right ventricle conduits. Between December 1990 and November 1994, eight of these patients had the initial Fontan connection converted to LT; these patients constitute the study group. The age at conversion to LT ranged from 7.9 to 25 years (14.3 ± 6.8 years). The interval between the original Fontan procedure and the conversion procedure was 7.3 ± 3.6 years, ranging from 3 to 13 years. There were four male and four female patients.

Data collection and follow-up. All available data, including medical records, operative notes, cardiac catheterizations, electrocardiograms, echocardiograms, and lung perfusion scans, were reviewed. Lung scans before and after conversion were obtained only for three patients in whom right pulmonary vein stenosis resulting from extrinsic compression by a “giant” right atrium was identified. An arbitrary clinical score (Table I) was assigned to each patient before conversion and at the latest follow-up. Four clinical features were considered as components of the score: (1) exercise tolerance, (2) arrhythmias, (3) effusions, and (4) protein-losing enteropathy. A maximum of 2 points was assigned for each feature, with a higher clinical score indicating a worse clinical condition.

Exercise tolerance. Exercise tolerance was graded according to the New York Heart Association classification for congestive heart failure. In addition, three patients also had exercise testing performed.

Arrhythmias. For evaluation of arrhythmias, clinical history and electrocardiograms were available in all cases, and Holter recordings before and after conversion were available for five and six patients, respectively. Intracardiac electrophysiologic studies were performed in two patients before conversion and in another two after conversion, at which time mapping of the atrial flutter circuits was performed. One of these patients underwent radiofrequency ablation.

Effusions. Effusions were considered to be present only when patients required hospitalization for drain-

age (chest tube placement, pericardiocentesis) or high doses of diuretic therapy (furosemide at more than $3 \text{ mg} \cdot \text{kg}^{-1} \cdot \text{day}^{-1}$). If effusions persisted for more than 3 weeks after onset of therapy, they were classified as chronic; if they were of shorter duration but recurred at least twice in the course of a 6-month period, they were considered intermittent.

Protein-losing enteropathy. Protein-losing enteropathy was considered to be present with a serum albumin concentration of less than 2 gm/dl on more than three separate determinations and a requirement for albumin replacement therapy at least once every 3 months.

Operative technique. All the patients had the original Fontan connection replaced by LT with a 4 mm fenestration.

Statistical analysis. The numeric data are expressed as mean $\pm$ standard deviation. The Wilcoxon signed-rank test was used to compare clinical scores before and after conversion to LT.

Results

Patient characteristics. Descriptive data are summarized in Table II. The diagnoses were tricuspid atresia (four patients), double-inlet left ventricle (three patients), and double-outlet right ventricle with multiple ventricular septal defects (one patient). Before APA, all had undergone previous palliative procedures including Blalock-Taussig shunts (three patients), Glenn shunts (two patients), or pulmonary artery banding (four patients). Five patients had been considered to be at high risk as Fontan candidates when APA was performed because of elevated pulmonary vascular resistance (two patients), pulmonary artery distortion (three patients), systemic venous abnormality (one patient), or moderate atrioventricular valve regurgitation (one patient). None had clinical arrhythmias before the initial modified Fontan procedure.

At the time of the conversion operation, all patients had decreased *exercise tolerance* of New York Heart Association functional class II, III, or IV. Six had arrhythmias involving recurrent intra-atrial reentry (flutter or fibrillation), and one patient had an atrial pacemaker implanted for sinus node dysfunction.

Fig. 1. Pressure tracings recorded at cardiac catheterization before conversion to LT (*Pre*) and after conversion (*Post*). Note the gradient between the pulmonary capillary wedge pressure (*PCW*) and the ventricular end-diastolic pressure (*LV*). After conversion to LT (*Post*), cardiac catheterization hemodynamics demonstrate resolution of pulmonary vein stenosis gradient.

Effusions were present in four patients. Three patients had ascites and pleural effusions and one had pleural and pericardial effusions. Two patients had protein-losing enteropathy necessitating long-term albumin replacement therapy. One patient had an atrial thrombus eight months before conversion; this condition was managed medically with complete resolution by the time of LT.

Cardiac catheterization. Preoperative cardiac catheterization was performed in all cases (Table II). Pulmonary artery mean pressures were higher than 16 mm Hg in six patients (median 18.3 ± 4.6 mm Hg). The median pulmonary vascular resistance was 4.8 ± 4.2 Wood units, and it exceeded 3 Wood units in five patients. All had low cardiac index (mean 1.9 ± 0.7 L $\cdot$ min⁻¹ $\cdot$ m⁻²). Obstruction of the right pulmonary venous return as a result of compression by a markedly dilated right atrium was diagnosed in three patients, with an average gradient of 10 mm Hg (Fig. 1). The pulmonary vein compression was demonstrated hemodynamically and by angiography in all cases as well as by chest magnetic resonance imaging in one (Fig. 2). Lung scintigraphy showed decreased perfusion of the right lung compared to the left.

Fig. 2. Chest magnetic resonance imaging in orthogonal (A) and transaxial (B) views show a giant right atrium after atriopulmonary anastomosis, before conversion to LT. Note the compression of the right pulmonary veins (arrow) against the spine by the posterior wall of the right atrium (*RA*) in part B.

Only one patient had elevated left ventricular end-diastolic pressure but preserved systolic ventricular function, in association with severe subaortic stenosis as a result of a restrictive bulboventricular foramen. In six patients, there was no significant gradient across the APA. Two patients, with right atrium-pulmonary artery and right atrium to right ventricle conduits had mean gradients of 4 mm Hg and 8 mm Hg, respectively. Two stents had previously been placed in two of the conduits, decreasing the gradient.¹⁶

Surgical LT procedure. The period between APA and conversion to LT was 7.3 ± 3.6 years (3 to 13 years). All patients underwent conversion to a fenestrated LT with polytetrafluoroethylene patch,* leaving minimal RA wall exposed to high pressures.

*Gore-Tex patch and tube, registered trademark of W. L. Gore & Associates, Inc., Newark, Del.

Table II. Patient characteristics

Pt. no.	Diagnoses	Previous operations	APA connection	Age at APA (yr)	Age at LT (yr)	Other procedures	NYHA class before LT
1	DORV, MVSDs	PAB	SVC-MPA, TV closure, ASD closure	5	8.8	SAS resection	III
2	Single DILV, BVF	Left BTS	RA to MPA nonvalved conduit, TV patch	5.2	16	—	IV
3	TA, TGA, restrictive VSD, JAA	Right BTS, PA plasty	APA, ASD closure, BVF resection	2.4	8.5	—	III
4	TA, restrictive VSD	Right BTS	RA to RV valved conduit, VSD and ASD closure	11	24	—	IV
5	Single DILV	PAB	APA, TV suprannular patch closure, ASD closure	3	7.9	SAS resection	II
6	TA, TGA, restrictive VSD	PAB, right Glenn	APA, fenestrated ASD closure	9	12	—	II
7	TA, restrictive VSD	Right Glenn	RA to MPA conduit, ASD closure	15	25	PA plasty (GTX)	III
8	Single DILV	PAB	APA, atrial patch	4.6	11	DKS	II
Mean				6.9 ± 4.4	14.3 ± 6.8		

APA, Atriopulmonary anastomosis or initial modified Fontan procedure; NYHA, New York Heart Association congestive heart failure classification; PVR, pulmonary vascular resistance (Wood units); CI, cardiac index ($L \cdot min^{-1} \cdot m^{-2}$); VEDP, systemic ventricular end-diastolic pressure (mm Hg); RAP, right atrial pressure (mm Hg); PVS, pulmonary vein stenosis mean gradient (mm Hg); DORV, double-outlet right ventricle; MVSDs, multiple ventricular septal defects; PAB, pulmonary artery banding; SVC-MPA, superior vena cava-main pulmonary artery; TV, tricuspid valve; ASD, atrial septal defect; SAS, subaortic stenosis; DILV, double-inlet left ventricle; BVF, bulboventricular foramen; BTS, Blalock-Taussig shunt; RA, right atrium; MPA, main pulmonary artery; TA, tricuspid atresia; TGA, transposition of the great arteries; VSD, ventricular septal defect; JAA, juxtaposition of the atrial appendages; PA, pulmonary artery; RV, right ventricle; GTX, pulmonary artery reconstruction with interposition of polytetrafluoroethylene tube; DKS, Damus-Kay-Stansel procedure.

*Fenestration closed at follow-up.

†Average gradient in three patients with PVS by compression.

Additional surgical procedures performed were closure or takedown of APAs, takedown of conduits and a previously implanted stent, atrial septectomy, closure of a tricuspid valve leak, subaortic resection for a restrictive bulboventricular foramen ($n = 2$) and main pulmonary artery to aortic anastomosis for subaortic obstruction ($n = 1$). One patient required interposition of a polytetrafluoroethylene tube* between discontinuous left and main pulmonary arteries.

Postoperative results and follow-up. One early death occurred on the fourth postoperative day related to elevated pulmonary vascular resistance, low cardiac index, and atrial arrhythmias (Table II; patient 4). The seven survivors were followed up for 23 ± 15 months (3 to 42 months). On the basis of the clinical score analyses (Fig. 3), three patients had marked clinical improvement, two patients had evidence of some improvement, and two patients remained unchanged. The average mean clinical score was 4.5 ± 1 before conversion and decreased to 3.0 ± 2 after conversion. The difference between the two was statistically significant by Wilcoxon signed-rank test ($p < 0.04$).

Exercise tolerance. All patients had decreased exercise tolerance before conversion, with New York Heart Association classes II (three patients), III (three patients), and IV (two patients, Table II). After conversion, six patients had either marked or partial improvement in the exercise tolerance. Five patients had improvement by one class and one patient had improvement by two classes.

Protein-losing enteropathy. Neither of the two patients with protein-losing enteropathy had improvement of this condition. One patient underwent cardiac transplantation at another institution as a result of persistent protein-losing enteropathy similar in severity to the preoperative condition. The remaining patient continues to require weekly albumin infusions at follow-up.

Effusions. Only one of four patients with chronic effusions had improvement of this condition. Ascites or pleural effusions persisted in the other three.

Arrhythmias. Of the six patients with atrial arrhythmias before conversion, only two are arrhythmia free (one on a regimen of digoxin and one without any antiarrhythmic medications). The patient who died in the early postoperative period had

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Score before LT	Catheterization characteristics before LT					Score after LT	NYHA class after LT	Follow-up (mo)
	PVR	CI	VEDP	RAP	PVS			
6	3.3	1.7	10	15	0	6	III	42
7	1.5	2.8	10	16	0	7	III	26
4	2.9	2.5	8	22	8	2	I (#)	38
6	3.4	1.5	12	17	0	6	dead	—
3	2.2	2.7	10	12	0	0	I	28
3	14.5	1.1	6	22	12	0	I (#)	18
4	7.2	1	10	16	10	2	II (#)	5
3	3.5	2	16	26	0	1	1	3
	4.8 ± 4.2	1.9 ± 0.7	10.3 ± 2.1	18.3 ± 4.6	10*			23 ± 15

recurrent atrial tachycardia before death. Of the remaining three patients with recurrent tachycardia, one underwent successful radiofrequency catheter ablation of a flutter circuit and two patients continue on regimens of medications.

Pulmonary vein compression. All three patients with pulmonary vein compression before conversion had clinical improvement after LT. In all cases, resolution of the gradient was confirmed by echocardiography and at cardiac catheterization. Although lung perfusion scans are difficult to interpret in patients with modified Fontan procedures as a result of incomplete mixing of the venous blood and streaming, two patients with preoperative preferential perfusion of the left lung now have preferential perfusion of the right lung after conversion to LT.

Thrombotic events. One patient with a complex surgical connection had a large intracardiac thrombus in the intraatrial baffle and left pulmonary artery polytetrafluoroethylene tube. It was managed with surgical thrombectomy and long-term oral anticoagulants and there has been no recurrence to date. No other thromboembolic event was observed after conversion to LT.

Oxygen saturation and fenestration. Among the seven survivors after conversion, the fenestration has been closed in three: by transcatheter clamshell closure in one (in whom a large baffle suture line defect was also similarly closed), intraoperative closure in another, and spontaneously in the remaining patient. The average oxygen saturation for these

three patients is 93%. Of the four patients with an open fenestration, two continue to have symptoms and are without clinical improvement; the other two have improvement, and transcatheter closure of the fenestration is anticipated.

Discussion

Since the initial reports for repair of tricuspid atresia by Fontan and Baudet¹⁷ and Kreutzer and associates,^{18,19} the lesions so treated have increased greatly and many modifications have been developed²⁰⁻²⁸ in an attempt to create a hemodynamically optimal right heart connection.

The total cavopulmonary connection with LT was initially proposed by Puga, Chiavarelli, and Hagler²⁹ and by Jonas and Castañeda³⁰ as an alternative to APA, particularly in the case of complex Fontan operations, such as for patients with left atrioventricular valve atresia or single atrioventricular valve. Several potential advantages became apparent: (1) the procedure was technically simple and reproducible in any atrioventricular arrangement, and (2) maintenance of low pressure is possible in most of the right atrium and coronary sinus, with possible improvement in the incidence of atrial arrhythmias. Subsequently, hydrodynamic studies performed by de Leval and coworkers⁷ suggested that the right atrium is not an efficient pump in nonvalved APA and that atrial pulsations could generate turbulence, leading to a higher energy loss. A reduction of energy loss as a result of lessened turbulence in the

Fig. 3. Bar graph illustrating clinical scores assigned to each patient before and after conversion at the latest follow-up, according to the arbitrary scoring system summarized in Table I. Each patient was assigned two bars, ranging from 0 points to 8 points, representing clinical scores both before (*first bar*) and after (*second bar*) conversion to LT. PLE, Protein-losing enteropathy.

LT potentially would decrease the incidence of atrial thrombosis or even increase the cardiac index. In addition, other experimental studies with animal models^{31, 32} supported the concept that the atrial contraction is not essential to the maintenance of pulmonary blood flow in a total right heart bypass operation. In vitro flow studies have also suggested that the total energy loss in a cavopulmonary model may be less than that seen in an atriopulmonary model,^{8, 33} although the magnitude of these losses is small. Published clinical results of different modified Fontan procedures tend to favor the total cavopulmonary anastomosis, or LT Fontan procedure, as the variation with lowest incidence of late atrial arrhythmias.^{3, 5, 6, 26} It is necessary to recognize the limitations of retrospective studies and the marked differences in follow-up periods for the different techniques. At present, the procedure of choice in many centers consists of a lateral intraatrial tunnel type of total cavopulmonary anastomosis, with or without a fenestration.

A recent report by Kao and colleagues⁹ on conversion of APA to cavopulmonary anastomosis in three patients operated on during 1993 and followed up for 8 months suggests that surgical conversion could be used for the management of atrial arrhythmias or acute atrial thrombosis. In our series, only one patient had had a thrombotic episode before

conversion; this was managed medically and had resolved by conversion. One patient who required a complex LT connection with interposition of a polytetrafluoroethylene tube between the left pulmonary artery and the main pulmonary artery had a thrombosis necessitating surgical thrombectomy after conversion to LT. Our series suggests that conversion does not necessarily eliminate the risk of acute thrombosis, particularly when a complex connection is required.

Conversion to LT seems to have been a beneficial surgical option for some of our eight patients who had symptoms after APA or other modified Fontan procedures, in which most of the right atrium was exposed to high pressures. The patients with protein-losing enteropathy did not significantly benefit from conversion to LT. In addition, only one of four patients with chronic effusions had improvement of this condition after conversion. Despite the theoretic advantages of LT versus APA, persistence of atrial arrhythmias after conversion did occur. The one patient who died had atrial arrhythmias and another two patients had recurrence of atrial flutter, in one case early after operation and in the other 3 years later. Longer follow-up is therefore necessary before the effect of conversion on the incidence of arrhythmias can be determined.

The etiology of atrial tachycardias in these patients is still poorly defined. Possible contributing factors include (1) atrial scars (suture lines and cannulation sites), (2) increased atrial pressure or atrial dilation, and (3) sinus node dysfunction. Conversion to LT does not eliminate all of these issues and may even introduce new suture lines. Recent data from catheter ablation studies³⁴ suggest that "zones of slow conduction" are present in the atrial wall of such patients, and these zones may not be eliminated simply by changing hemodynamics. As data accumulate from arrhythmia mapping and radiofrequency ablation, it may be possible to devise a surgical approach that involves surgical interruption of all potential slow zones, thereby eliminating the true substrate for these arrhythmias.

The most consistent clinical improvement occurred in patients with right pulmonary vein compression before conversion. This finding emphasizes the need for accurate hemodynamic assessment of patients with symptoms after APA or other modified Fontan procedures. Right pulmonary vein stenosis as a result of extrinsic compression by a dilated right atrium has not previously been reported, and its incidence among patients who have undergone Fon-

tan operations is unknown. Pulmonary vein obstruction increases pulmonary vascular resistance, which would seem to be particularly detrimental in the setting of Fontan circulation. Patients with symptoms in whom this phenomenon is present are candidates for conversion to LT. This conclusion is supported by the pulmonary vein gradient relief and the improvement in symptoms achieved in our three patients.

All the patients in this study had a fenestration placed in the atrial baffle, a procedure previously shown to decrease early postoperative morbidity³⁵ and mortality, although it may not necessarily influence long-term results. It is not possible to determine in this group of patients how much of the clinical improvement can be attributed to the presence of a fenestration. In three of the seven survivors the fenestration is now closed and these patients have shown sustained clinical improvement.

Limitations. The small number of patients in the study, their complex lesions, the absence of a control group, and the short follow-up period limit our ability to identify predictors of improvement with conversion to LT. In this retrospective study, selections of diagnostic tests and management were influenced by individual patient's physicians. Although evaluation of arrhythmias before conversion included 12-lead electrocardiography for all eight patients, Holter monitoring was carried out for only five patients and intracardiac electrophysiologic study was carried out for only two. Not every patient was catheterized after conversion, and postconversion hemodynamics were therefore not available in all cases. Additional surgical procedures were performed in some patients at the time of LT, and conversion to LT may thus not have been the only reason for clinical improvement.

Conclusions. Conversion from APA to a fenestrated LT is technically feasible and resulted in clinical improvement in some patients with symptoms after short term follow-up. It did not improve the conditions of those with protein-losing enteropathy, nor those of most with chronic effusions, suggesting that such patients may best be treated by direct cardiac transplantation rather than by conversion to LT. The procedure was successful for patients in whom a giant right atrium resulted in pulmonary vein compression, in all of whom the gradient was relieved. Attention to the possible diagnosis of right pulmonary vein stenosis is there-

fore required because conversion to LT seems to particularly benefit these patients.

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