

Fontan Deterioration in Pediatric Cardiac Surgeon's View

세종병원 흉부외과
이창하

Fontan deterioration

Failing Fontan

Failed Fontan

Fontan Failure

Fontan operation

'This procedure is not an anatomical correction, which would require the creation of a right ventricle, but a procedure of physiological pulmonary blood flow restoration, with suppression of right and left blood mixing'

(F. Fontan and E. Baudet, Thorax, 1971)



Outcome After a “Perfect” Fontan Operation

Francis Fontan, MD, John W. Kirklin, MD, Guy Fernandez, MD, Francisco Costa, MD,
David C. Naftel, PhD, Francesco Tritto, PhD, and Eugene H. Blackstone, MD

A study was undertaken to determine the early and long-term outcomes dictated by the Fontan state per se (a state in which the force driving pulmonary blood flow is solely or largely a residue in the systemic venous pressure of the main ventricular chamber's contractile force)

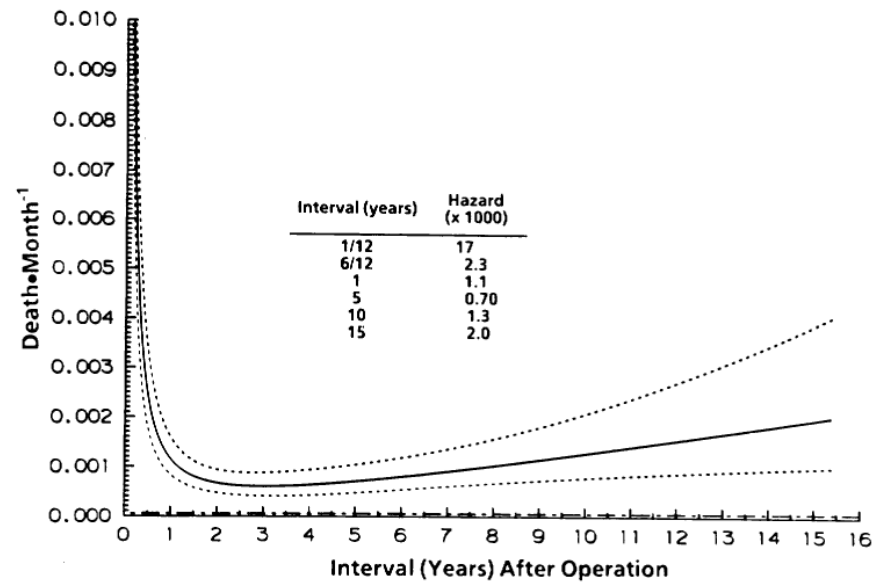
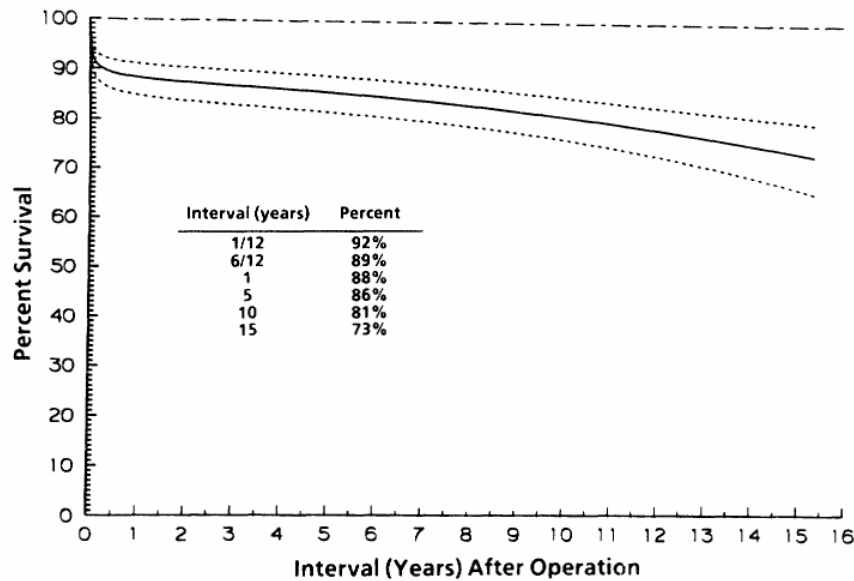
1975 - 1988

334 patient underwent Fontan procedure under optimal conditions

years, respectively, after the Fontan operation. The hazard function (instantaneous risk of death at each moment in time after the operation) had an early rapidly declining phase of hazard that at about 6 months began to give way to a late hazard phase, which was rising by about 6 years after surgery. A secondary study design, using the theory of competing risks, yielded survival and hazard function information very similar to that of the primary study design. The functional capacity of the patients as expressed by New York Heart Association class was less, the longer the period of follow-up. No risk factors (other than older age at time of surgery) were found for the late decline in survival or the decline in functional status. The inference is that the premature decline in survival and functional status and the late rise in hazard function are from the Fontan state per se and that the Fontan operation is, therefore, palliative but not curative. (*Circulation* 1990;81:1520–1536)

Outcome after a "perfect" Fontan operation

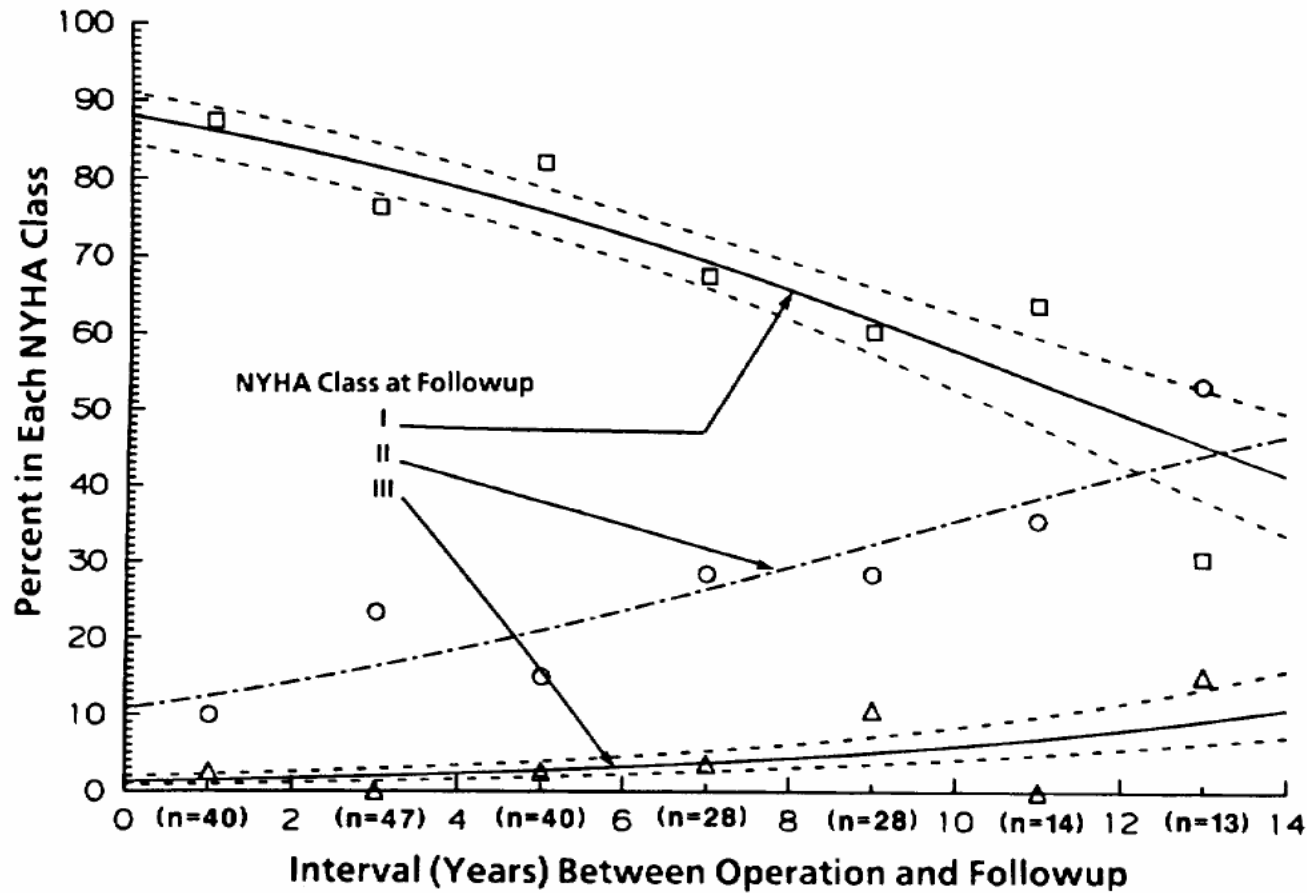
Survival After Perfect Fontan Operation



* The appreciable late hazard (instantaneous risk of death at each moment in time after the operation) gradually began to increase about 6 years after surgery.

Outcome after a "perfect" Fontan operation

Functional Status



Outcome After a “Perfect” Fontan Operation

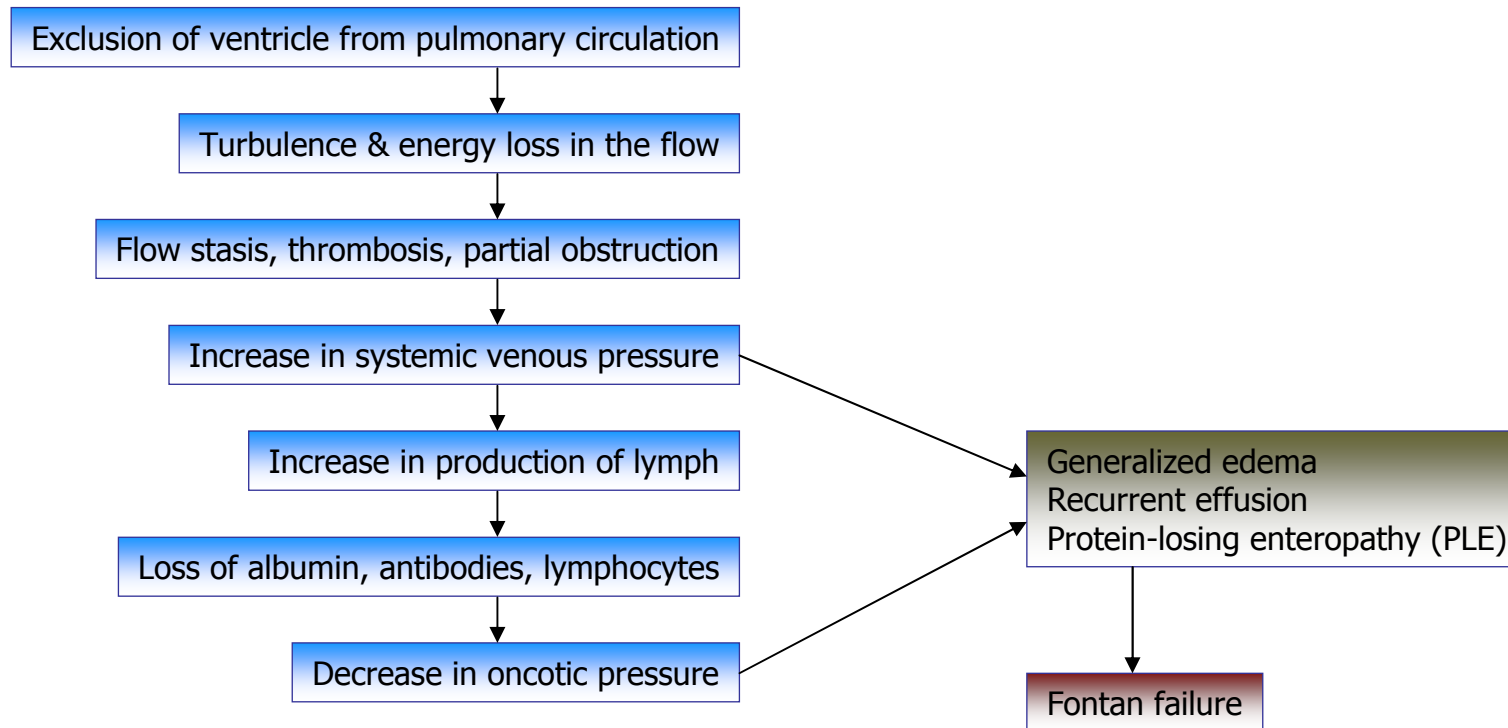
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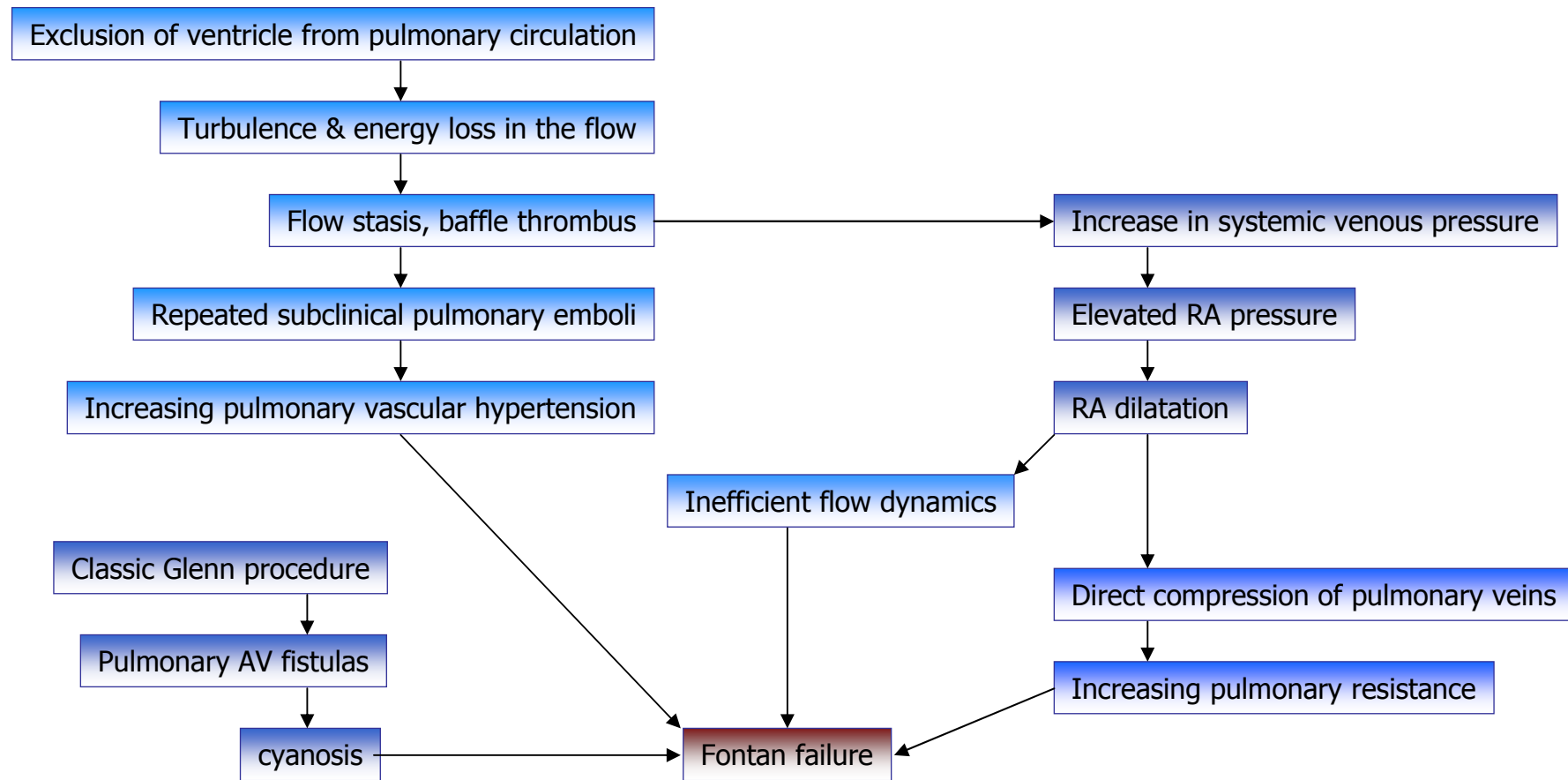
Fontan circulation

-Physiologic & anatomic sequelae-



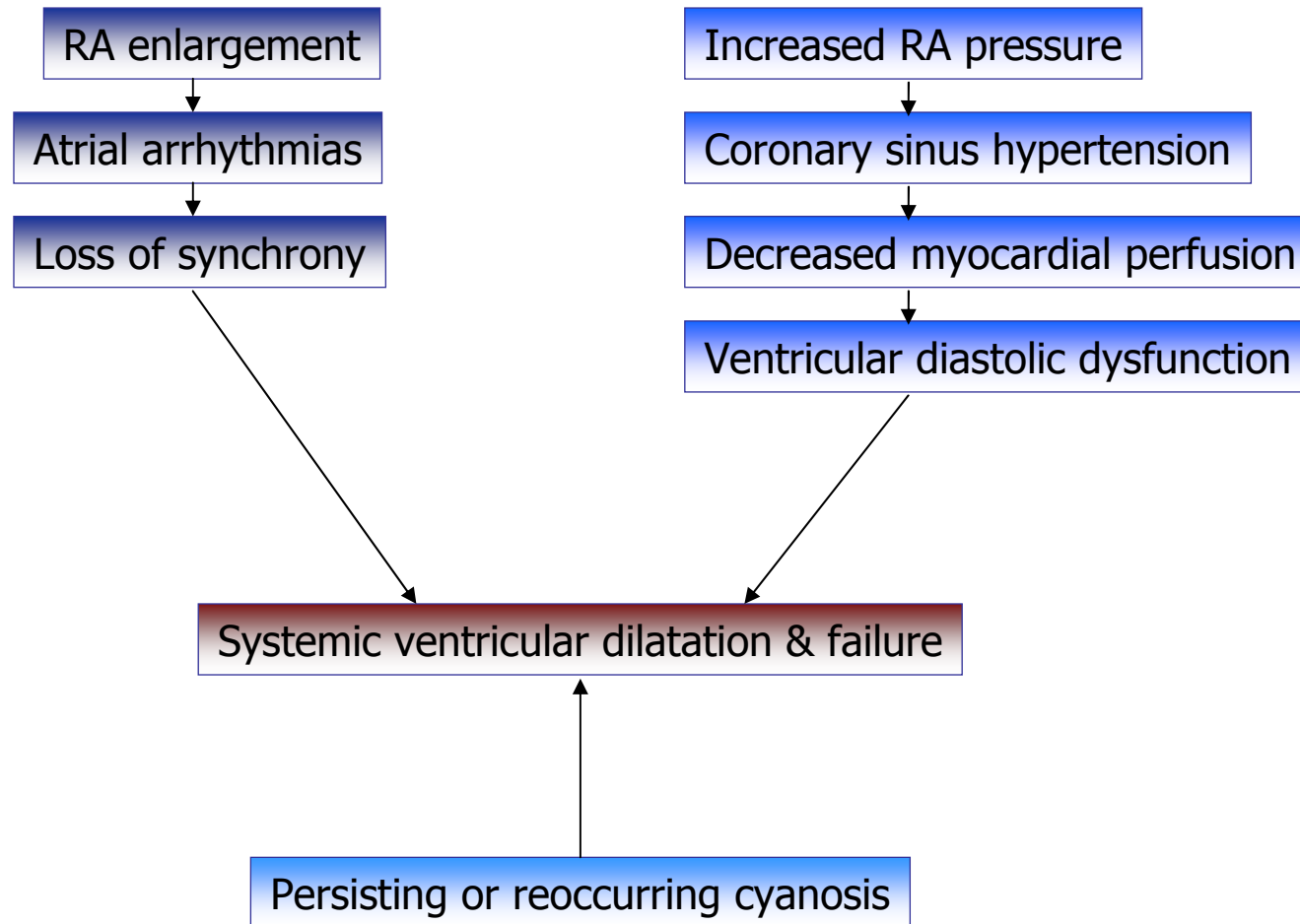
Fontan circulation

-Physiologic & anatomic sequelae-



Fontan circulation

-Systemic ventricular failure-



Surgical modifications

- Not original Fontan or its modifications
- deLeval (1988)
 - ‘Lateral wall’ TCPC; experimentally & clinically to be hemodynamically more efficient
- Early 1990, extracardiac conduit Fontan procedure

Further surgical modifications

- Prior volume unloading via superior cavopulmonary connection (i.e. bidirectional Glenn, hemi-Fontan operation)
- Fenestration
- Better myocardial preservation techniques

Lateral Tunnel Versus Extracardiac Conduit Fontan Procedure: A Concurrent Comparison

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Background.

Outcomes of lateral tunnel (LT) versus extracardiac conduit (ECC) Fontan procedures over the same period were compared.

Methods.

70 Fontan procedures were performed between 1995 and 2002.

All were performed in a single center.

Superior vena cava anastomosis was performed in all patients.

ECC patients were divided into class I and class II.

3.9 ± 2.5 years.

hypoplastic left heart syndrome (57% vs 21%; $p < 0.01$),

and a longer aortic cross-clamp time (55 ± 13 vs 26 ± 15 min; $p < 0.01$).

Weight, gender, preoperative cardiac catheterization values, and cardiopulmonary bypass time did not differ between the two groups.

Results.

Operative mortality was 2.8%, 1 patient in each group ($p = 1.0$).

Over the first 24 hours following operation the mean Fontan pressure, transpulmonary gradient, and common atrial pressure did not differ between LT and ECC patients.

The median duration of mechanical ventilation (LT 12 vs ECC 18 hours), intensive care unit

1995-2002

70 Fontan procedures

37 LT / 33 ECC

Operative mortality 2.8%

Survival at 5yr; 97% for LT, 91% for ECC, $p = 0.4$

LT patients (LT 10 vs ECC 12 days) did not differ.

Incidence of sinus node dysfunction during the follow-up period was similar.

at hospital discharge.

LT patients.

low-up was similar.

ECC patients.

survival at 5 years.

($p = 0.4$);

in class II,

with no difference between groups.

Sinus node dysfunction was seen during follow-up in 15% LT vs 28% ECC patients ($p = 0.2$).

Conclusions.

The LT and ECC approaches had comparable early and mid-term outcomes, including operative morbidity and mortality, postoperative hemodynamics, resource use, and mid-term survival and functional status.

ECC patients had a higher incidence of sinus node dysfunction early after operation.

(Ann Thorac Surg 2003;76:1389-97)

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Fontan Operation: A Comparison of Lateral Tunnel with Extracardiac Conduit

Andrew C. Fiore, MD, Mark Turrentine, MD, Mark Rodefeld, MD,
Palaniswamy Vijay, PhD, Theresa L. Schwartz, MD, Katherine S. Virgo, PhD,
Laurice K. Fischer, MD, and John W. Brown, MD

Divisions of Cardiothoracic Surgery, St. Louis University School of Medicine/Cardinal Glennon Children's Hospital, St. Louis, Missouri, and Indiana University School of Medicine/James Whitcomb Riley Children's Hospital, Indianapolis, Indiana

Background. The outcome of the lateral tunnel (LT) Fontan operation was compared with the extracardiac conduit (EC) Fontan operation.

Methods. From 1990 to 2004, 162 Fontan operations were performed: 49 EC and 113 LT. The EC patients were older and had a greater frequency of heterotaxy syndrome, and LT patients had a higher incidence of hypoplastic left heart syndrome. Preoperative transpulmonary gradient, ventricular end-diastolic pressure, McGoon index, room air saturation, and cardiac rhythm were similar. EC patients underwent superior caval pulmonary connection, and LT patients underwent hemi-Fontan. Cardiopulmonary bypass time was similar, but fewer EC patients needed aortic cross-clamping. Fenestration was more frequent in LT patients (EC, 16% versus LT, 73%; $p < 0.01$).

1990-2004

162 Fontan procedures

49 ECC / 113 LT

Overall operative mortality 1.8%

Survival at 5yr; 90% EC, 95% LT, $p = 0.08$

The EC patients were older and had a greater frequency of heterotaxy syndrome, and LT patients had a higher incidence of hypoplastic left heart syndrome. Preoperative transpulmonary gradient, ventricular end-diastolic pressure, McGoon index, room air saturation, and cardiac rhythm were similar. EC patients underwent superior caval pulmonary connection, and LT patients underwent hemi-Fontan. Cardiopulmonary bypass time was similar, but fewer EC patients needed aortic cross-clamping. Fenestration was more frequent in LT patients (EC, 16% versus LT, 73%; $p < 0.01$). There were seven late deaths (EC 4 versus LT 3; $p = NS$). Actuarial survival at 5 years was not significantly different (EC, 90% versus LT, 95%; $p = 0.08$).

Conclusions. The EC and LT operation had comparable early and late mortality, readmission for chyloous effusion, preservation of sinus rhythm, and frequency of all neurologic events. The more frequently fenestrated LT cohort used fewer resources.

(Ann Thorac Surg 2007;83:622-30)
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Outcomes of current practice

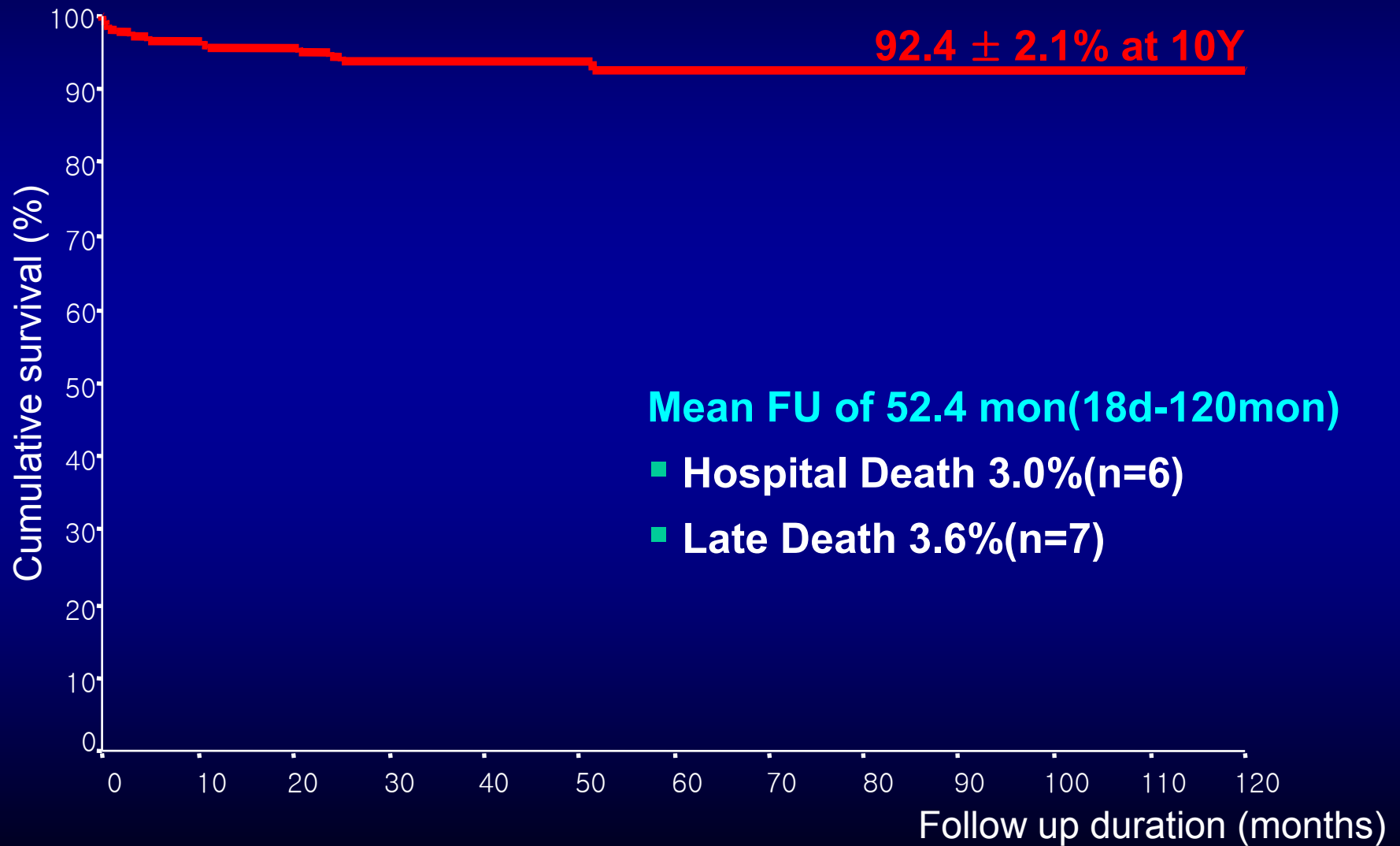
Extracardiac conduit Fontan

Sejong experiences

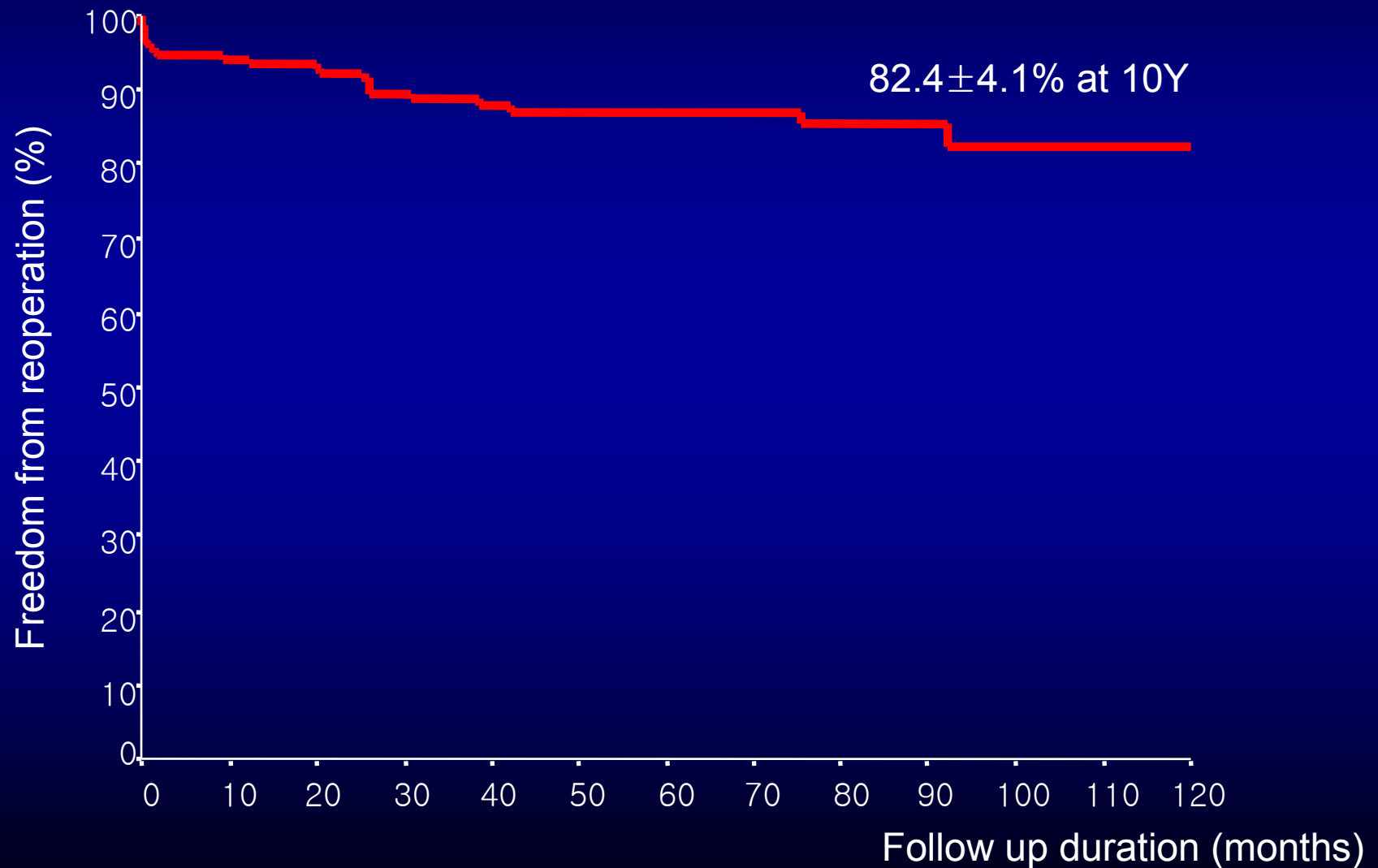
Patients

- Aug. 1996 ~ Aug. 2006
- 200 patients with extracardiac Fontan Op.
- Age : median 3.4Y (16M~35.7Y)
- Previous Op. before BCPS 94.0% (188)
- Staged Op. 89.5% (179)
- Fenestration 42.5% (85)

Overall Survival



Freedom from Reoperation



Reoperation

- Reoperations 24(12%)

Reoperation

Conduit revision 7

Fenestration 6

AV valve operation 5

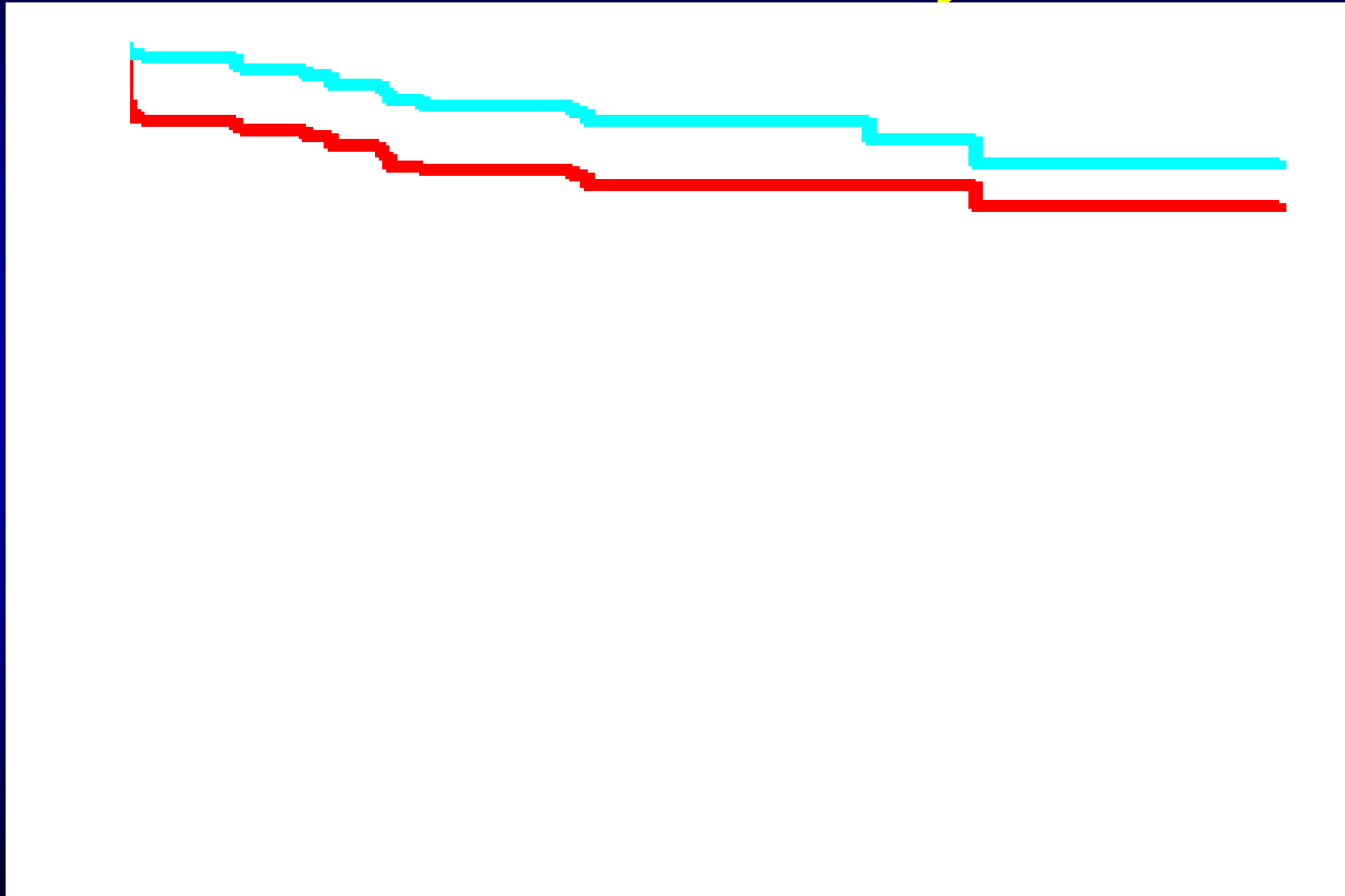
PPM insertion 5

PA angioplasty 4

HV-azygos v 3

Other: Atrial septectomy(1),Azygos v-LPA(1),CS
unroofing(1),LVOTO relieve(1)

Freedom from Arrhythmia

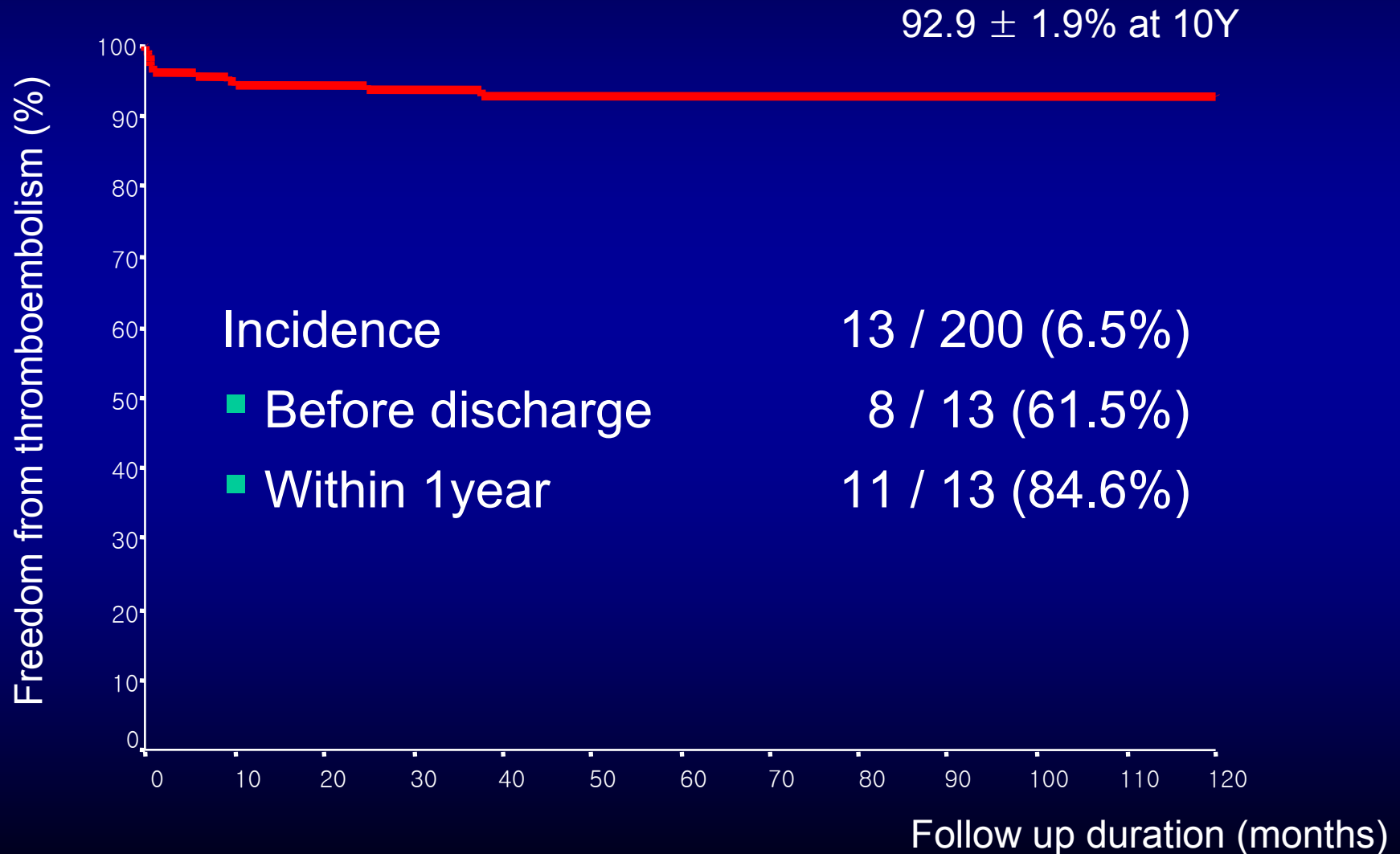


Follow up duration (months)

Arrhythmia

Type	Overall	Early	Late
Bradyarrhythmia	26	10	13
SN dysfunction	13	5	8
PPM	12 (3)	5	4
SN dysfunction	9 (3)	3	3
AV block	3	2	1
Sinus bradycardia	1	0	1
Tachyarrhythmia	10	7	3
PSVT	8	5	3
AF	2	2	0
JET	1	1	0
Total	32/200 (16.0%)	15/200 (7.5%)	15/194 (7.7%)

Freedom from Thromboembolism

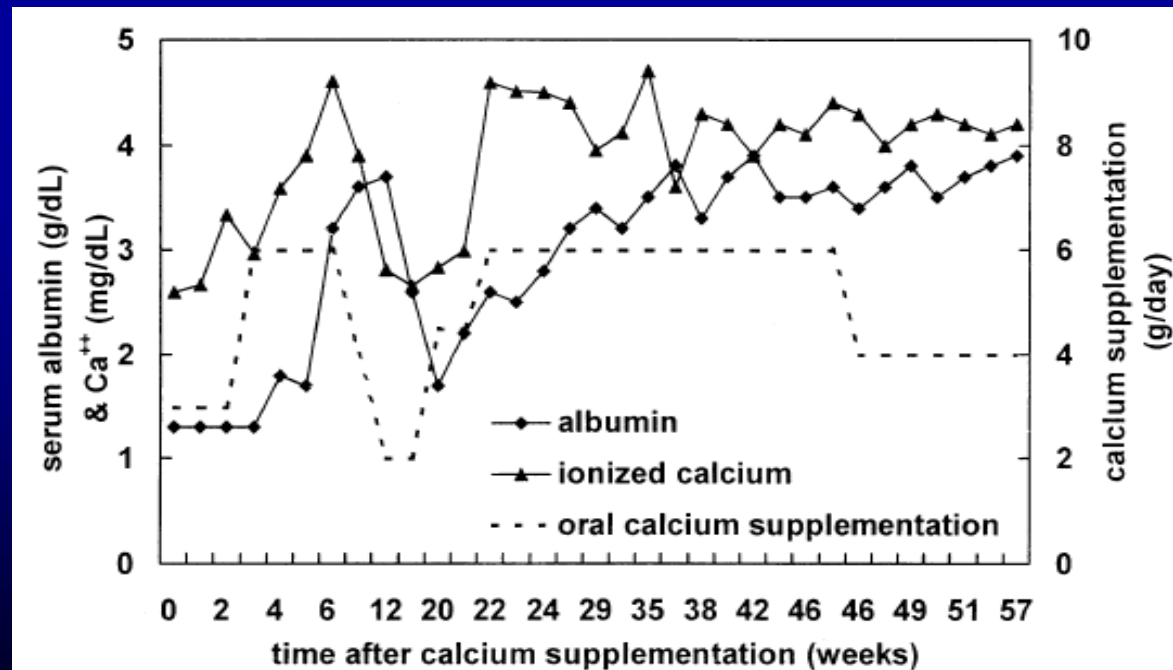


Protein-losing Enteropathy (n=5)

- Recovery 3
- Death 2

Reversal of protein-losing enteropathy with calcium replacement in a patient after Fontan operation.

Case



Surgical reinterventions following Fontan procedure



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European Journal of Cardio-thoracic Surgery 24 (2003) 255–259

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Surgical reinterventions following the Fontan procedure[☆]

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1995-2001

Philadelphia,

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123 procedures in 71 patients

Boulevard.

Median time from Fontan to reoperation 3.6yr

Abstract

Objective: The Fontan procedure is utilized as a final reconstructive procedure for patients with functional single ventricle. Short- and long-term outcomes have improved significantly, however, some patients require additional cardiac procedures following the Fontan operation. The outcomes for these reinterventions are not known. **Methods:** Cardiac Surgery and Cardiac Intensive Care Unit databases at The Children's Hospital of Philadelphia were reviewed to identify all patients who underwent cardiac surgery after a previous Fontan operation between January 1, 1995 and December 31, 2001. **Results:** During the study period, 123 procedures were performed in 71 patients. The median time from Fontan to reoperation was 3.6 years (range 0.1–20 years). Indications for reintervention included arrhythmia, cyanosis, 'failing' Fontan circulation or exercise intolerance, protein losing enteropathy, atrioventricular valve (AVV) regurgitation, and other indications. Procedures included pacemaker insertion or revision ($n = 59$, 48%), reinclusion of previously excluded hepatic veins ($n = 16$, 13%), revision to either a lateral tunnel or extra-cardiac conduit Fontan ($n = 13$, 11%), cardiac transplantation ($n = 9$, 7%), enlargement or creation of a baffle fenestration ($n = 6$, 5%), isolated AVV repair or replacement ($n = 2$, 2%), and other procedures ($n = 18$, 14%). There were five early and five late deaths. Hospital mortality was greatest for patients undergoing cardiac transplantation (4/9, 44%), accounting for 80% of the early deaths. **Conclusions:** Surgical reinterventions following the Fontan procedure may be necessary for multiple indications which result in impairment of the Fontan circulation. Most reinterventions can be performed with minimal morbidity and mortality. Survival for patients requiring cardiac transplantation following the Fontan procedure remains poor.

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Keywords: Fontan procedure; Reoperation; Protein losing enteropathy; Pacemaker; Heart transplantation

Surgical reinterventions following Fontan procedure

Indications for reintervention

Arrhythmia/pacemaker

- Bradyarrhythmia
- Tachyarrhythmia
- Generator malfunction
- Lead malfunction

Cyanosis

- Hepatic vein exclusion
- Baffle leak

'Failing' Fontan circulation

- Progressive exercise intolerance
- Congestive heart failure

Protein losing enteropathy

Atrioventricular valve regurgitation

Other

- Fontan pathway obstruction
 - Pleural/pericardial effusion
 - Osteomyelitis (pacing system related)
 - Thromboembolic events
-

Surgical reinterventions following Fontan procedure

- Pacemaker insertion (n = 38)
- Hepatic vein reinclusion (n = 16)
- Fontan revision (n = 13)
- Heart transplantation (n = 9)
- Fenestration creation/enlargement (n = 5)
- AVV repair/replacement (n = 7)

Surgical reinterventions following Fontan procedure

- Other procedures
 - Placement of a pericardial window for pericardial drainage
 - Pleural drainage for late effusions
 - Peripheral thrombectomy
 - Reconstruction of discontinuous pulmonary arteries
 - Repair of Fontan baffle leak
 - Takedown of the Fontan circulation to a superior cavopulmonary connection



10-year survival after Fontan-type operation

	Number of patients	Years of operation	Survival (%)
Fontan (1990)	334	1975-1988	69%
Driscoll (1992)	352	1973-1984	70%
Cetta (1992)	339	1987-1992	81%
Gentles (1997)	500	1973-1991	79%
Weipert (2004)	162	1978-1995	83%
Giannico (2006)	221	1988-2003	85%



Clinical Outcome of 193 Extracardiac Fontan Patients

The First 15 Years

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Fabrizio Drago, MD,* Attilio Turchetta, MD,† Roberto Di Donato, MD,*
Stephen P. Sanders, MD, FACC*

Rome, Italy

OBJECTIVE To report the clinical outcome of 193 patients who underwent extracardiac Fontan procedure between 1988 and 2003.

BACKGROUND The extracardiac Fontan procedure is a well-established surgical approach for the treatment of single ventricle anomalies.

METHODS A retrospective analysis of 193 patients who underwent extracardiac Fontan procedure between 1988 and 2003. The mean age at surgery was 2.2 years (range 0.1-17.9 years). The median follow-up was 72.2 months (range 1.2-179 months).

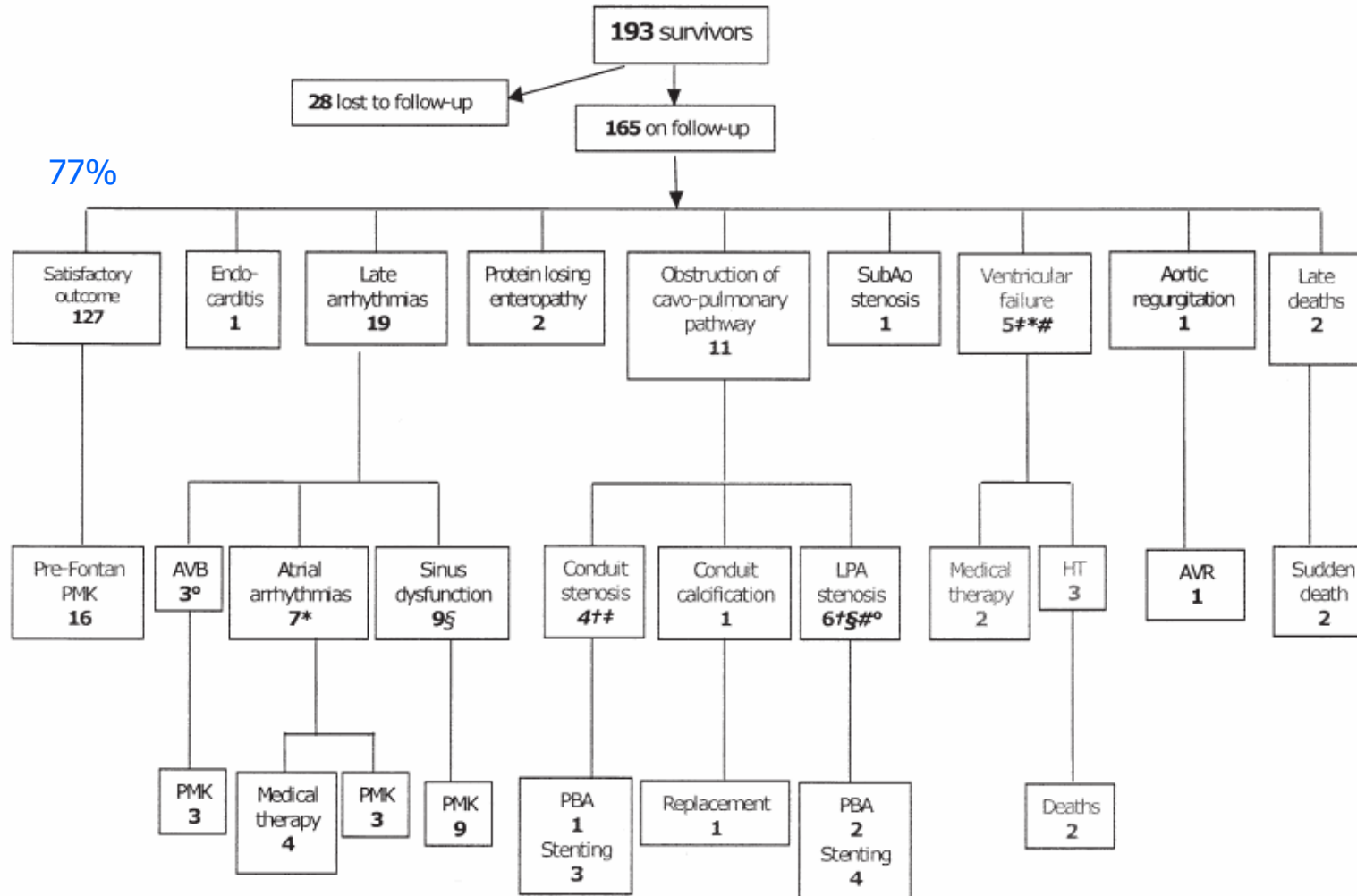
RESULTS The overall survival was 92% at 15 years. A total of 127 of 165 survivors (77%) were in New York Heart Association functional class I. The incidence of late major problems was 24% (42 major problems in 36 of 165 patients): 19 patients had arrhythmias (11%), 5 patients had obstruction of the extracardiac conduit (3%) and 6 of the left pulmonary artery (3.5%), and 5 patients experienced ventricular failure (3%), leading to heart transplantation in 3 patients. Protein-losing enteropathy was found in two patients (1%). The incidence of late re-interventions was 12.7% (21 of 165 patients, including 15 epicardial pacemaker implantations). Four patients died (2.3%), two after heart transplantation.

CONCLUSIONS After 15 years of follow-up, the overall survival, the functional status, and the cardiopulmonary performance of survivors of the extracardiac Fontan procedure compare favorably with other series of patients who underwent the lateral tunnel approach. The incidence of late deaths, obstructions of the cavopulmonary pathway, re-interventions, and arrhythmias is lower than that reported late after other Fontan-type operations. (J Am Coll Cardiol 2006; 47:2065-73) © 2006 by the American College of Cardiology Foundation

1988-2003
221 patient underwent extracardiac Fontan
193 early survivors
Median F/U 50mo (1 -179mo)

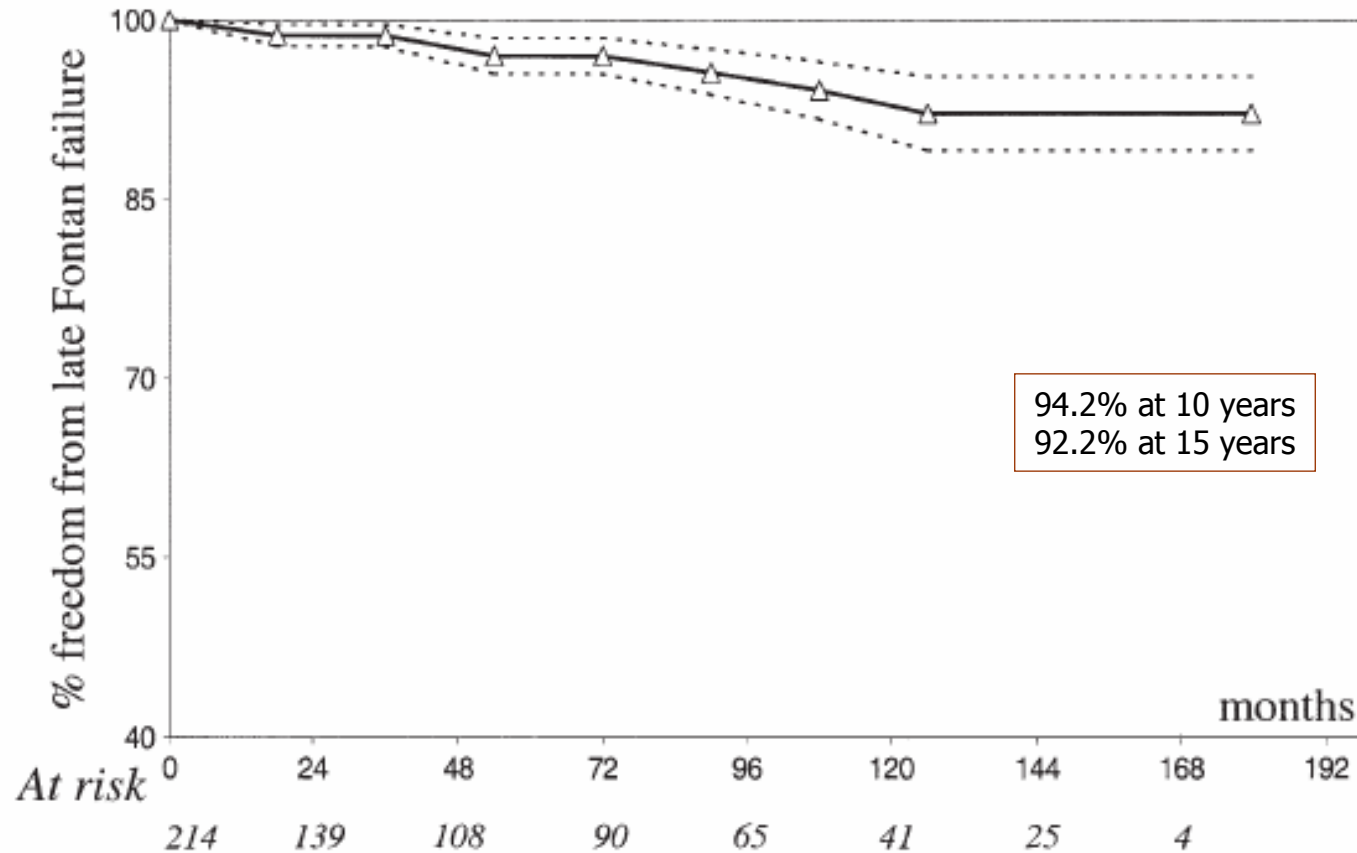
Outcome of 193 survivors

(mean 63mo, median 50mo)

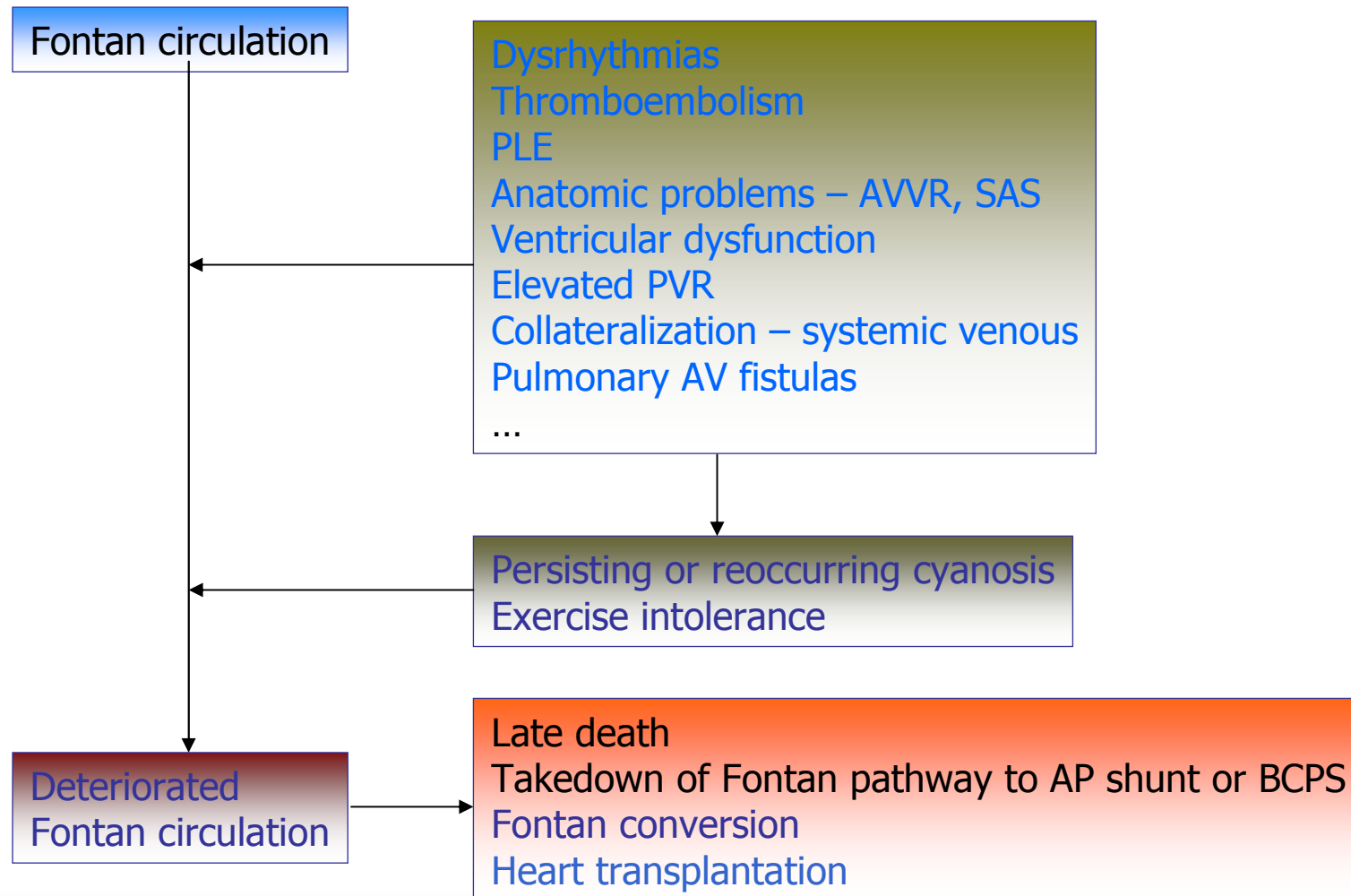


Late Fontan failure

- Death, takedown, or heart transplantation -



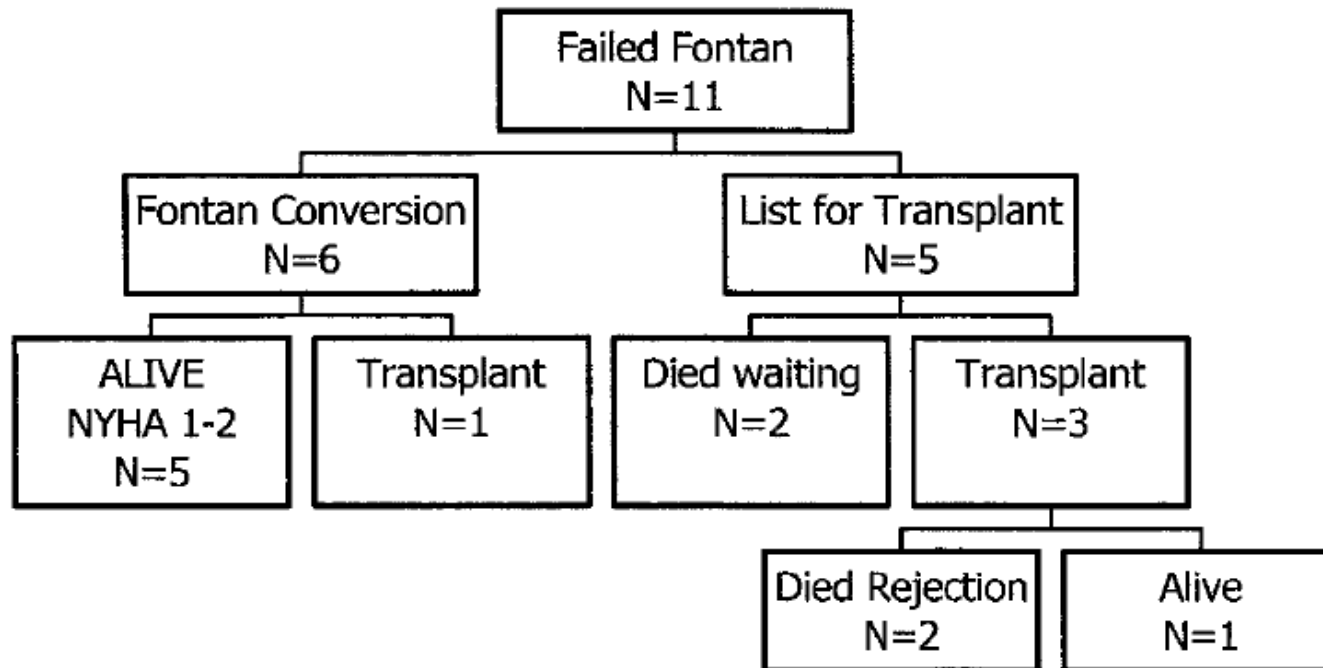
Failing Fontan circulation



Failing Fontan circulation

Fontan conversion vs. Heart
transplantation

Failed Fontan



Fontan conversion vs. transplantation

(Mavroudis et al. J Thorac Cardiovasc Surg 2001;122:863-71)

Fontan conversion vs. transplantation

- The presence of substrates that can be repaired
 - A valve lesion
 - AV valve or aortic valve
 - An obstructive lesion
 - Baffling obstruction
 - Pulmonary vein compression
 - Ventricular dysfunction associated with arrhythmogenic or anatomic substrates

Fontan conversion vs. transplantation

- Isolated systemic ventricular dysfunction
 - Ventricular dysfunction
 - $VEDP \geq 12\text{mmHg}$
 - Underloaded ventricle
 - Low cardiac output
 - Poor transit of systemic venous blood into & through the lungs
- PLE
- NYHA class IV heart failure

Fontan conversion

이창하3

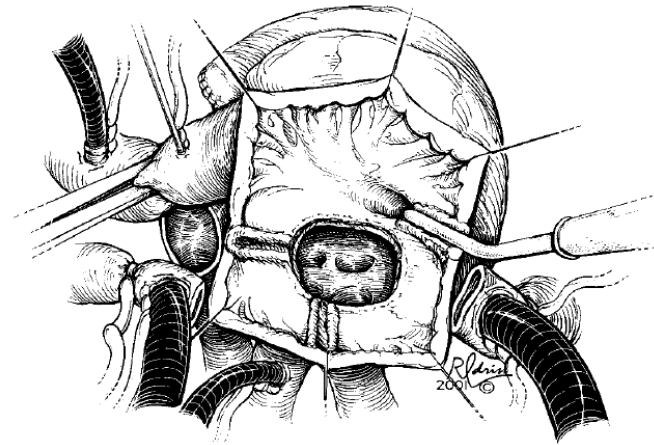
이창하3 heart failure in children and young adult 참조하고 있음
이창하, 2007-04-12

Surgical management protocol

- Takedown of old Fontan connection
- Creation of TCPC
 - Extracardiac conduit
 - Intra-arterial lateral tunnel
- Preexisting atrial arrhythmias
 - Intraoperative ablation surgery including cryoablation
- Atrial debulking
- Correction of residual or recurrent lesions
 - AVVR, SAS, distorted pulmonary arteries

Surgical management protocol

- Intraoperative ablation surgery
 - Atrial reentry tachycardia or flutter
 - Right-sided maze
 - Atrial fibrillation
 - Maze–Cox III
 - Permanent pacemaker should be placed
- Prophylactic arrhythmia ablation surgery
 - No data
 - However, extracardiac connections limit intracardiac access for subsequent arrhythmia therapy whether it is for catheter ablation or pacemaker implantation.



Fontan conversion

Author	Year of revision	Series size	Mode of revision	Arrhythmia surgery	FU (mo)	Results	Last FU status
McElhinney (1996)	1992-1995	7	EC 5 IC 2	Not done	17	1 Early death 1 OHT	Improved NYHA class, 4 PPM, 2
Kreutzer (1999)	1990-1994	8	LT 8	Not done	23	1 Early death	Improved NYHA class, 5
Mavroudis (2001)	1994-2001	40	EC 32 LT 6	Isthmus cryoablation 10 RA maze 16 Maze-Cox III 14	30	1 Late death (after OHT) 2 OHT	NYHA class I/II, 37 PPM, 38
Setty (2002)	1997-2001	6	EC 6	Limited RA Maze 6	29	No mortality	Improved NYHA class, 6 PPM, 6
Weinstein (2003)	1999-2002	10	EC 10	RA Maze 8 Full Maze 2	17	2 Early deaths	NYHA class I/II, 8 PPM, 9
Sheikh (2004)	1997-2002	15	EC 11 LT 4	Cryoablation 11	43	1 Late death	Improved NYHA class, 14 PPM 11
Kim (2005)	1996-2004	16	EC 11 LT 5	Isthmus cryoablation 10 RA Maze 3	27	No mortality	NYHA class I/II, 16 PPM, 9
Morales (2005)	1997-2004	35	EC 19 LT 16	RA Maze 28	29	2 Late deaths	NYHA class I/ II, 32 PPM, 29

* EC; extracardiac conduit, IC; intra-atrial conduit, LT; lateral tunnel, OHT; orthotopic heart transplantation, FU; follow-up, PPM; permanent pacemaker

Lessons learned

- Fenestration
 - Unnecessary
 - Except patients with PLE
- PLE
 - Not good candidate
- Aggressive management for atrial arrhythmias
- Adequate timing

Timing ?

- Intervening during early stages of failing Fontan
- At the first signs of atrial arrhythmias, conduit obstruction, significant atrial enlargement, or valvular dysfunction
- Poor outcomes in marginal transplantation candidates (i.e. NYHA class IV)
- Optimally, revision should be undertaken early in symptomatic patients before irreversible ventricular failure ensues

이창하4 timing인지 p549 suggested management protocol 인지 확실히 해야함
이창하, 2007-04-12

Fontan conversion

Sejong experience

- 2001-2005
- 6 patients underwent Fontan conversion
 - AP Fontan (n = 5); lateral tunnel (n = 1)
 - BT shunt (n = 1); classic Glenn (n = 1); bidirectional Glenn (n = 1)
 - Conversion to extracardiac conduit Fontan in all

Fontan conversion

Sejong experience

- Indication
 - AP Fontan (n = 5)
 - DOE, dilated RA & atrial dysrhythmia in all
 - RA thrombi (n = 2); RPV compression (n= 1); Cyanosis (n = 1)
 - Lateral tunnel (n = 1)
 - Total occlusion of lateral tunnel with hepatomegaly
 - Sinus node dysfunction

Fontan conversion

Sejong experience

Patient	Diagnosis	Type of original Fontan	Duration of Fontan-conversion (year)	Age at conversion (year)	F/U duration (year)
1	TA IIb	AP Fontan	12.5	18.9	5.4
2	DORV PS restrictive VSD	AP Fontan	15.7	18.1	5.1
3	TA IIb	AP Fontan	14.5	20.8	4.1
4	TA Ib	Lateral tunnel	7.9	10.8	3.8
5	TA IIb	AP Fontan	22.7	36.2	2.8
6	RV type UVH	AP Fontan	18.8	24.6	1.4

Fontan conversion

Sejong experience

- Arrhythmia surgery
 - Cryoablation (n = 2) / Modified right-sided maze (n = 3)
 - PPM insertion (n = 3)

Fontan conversion

Sejong experience

- Follow-up (median 3.9 yr, 1.4 – 5.4 yr)
 - Improved functional class in all
 - Sinus rhythm (n = 3), DDD pacing (n = 3)



Heart transplantation

Reported outcomes of transplantation for Fontan failure

Study	n	Hospital mortality	F/U (mo)	Survival estimate
Hsu (1995)	9	33%	23	67%
Carey (1998)	9	33%	17	67%
Lamour (1999)	8	38%	35	50%
Michielon (2003)	6	67%	ND	ND
Mitchell (2003, AATS)	15	7%	60	82%
Gamba (2004)	14	14%	65	77%

* ND, data could not be derived from report

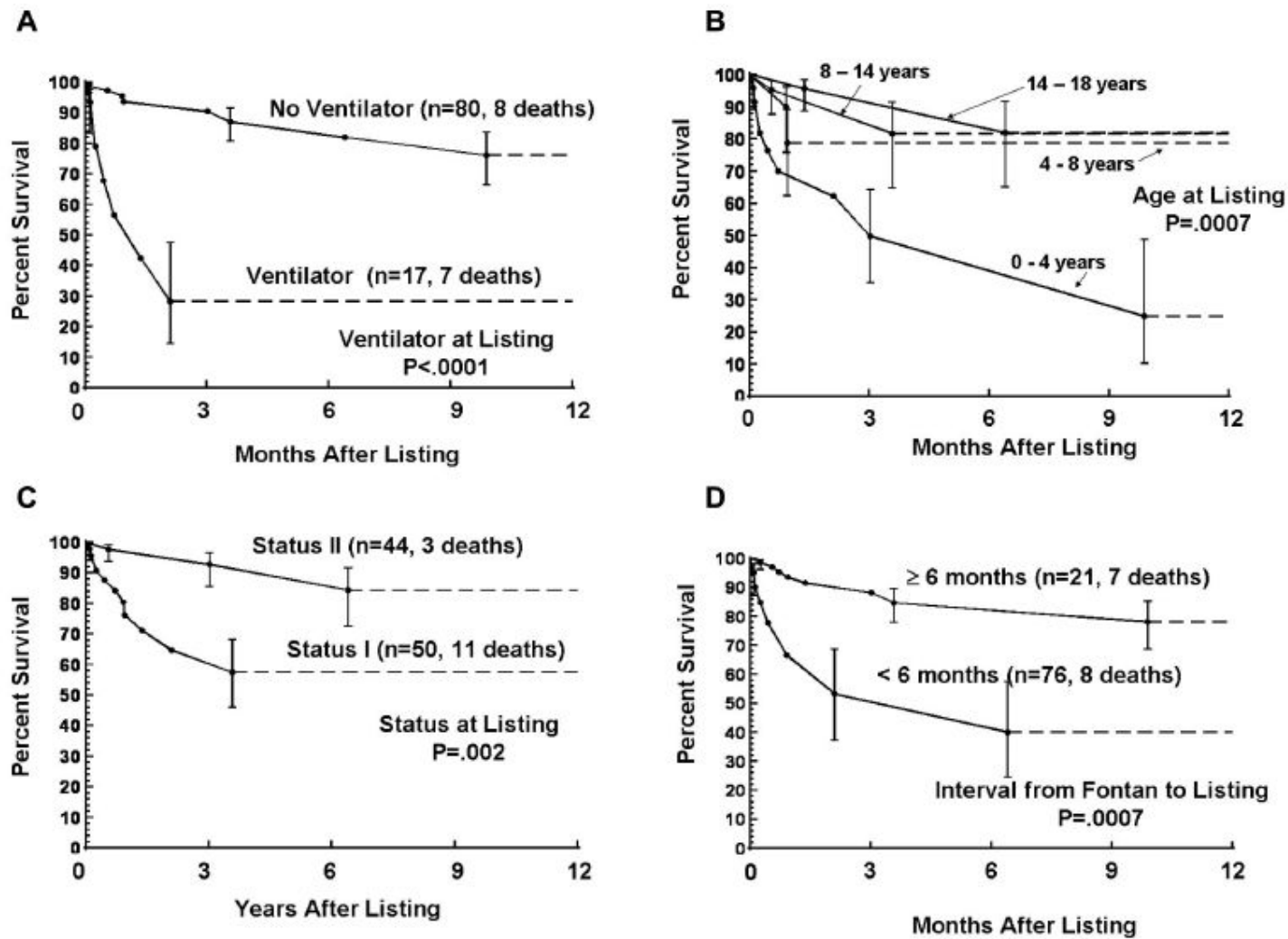
Congenital Heart Disease

Outcome of Listing for Cardiac Transplantation for Failed Fontan

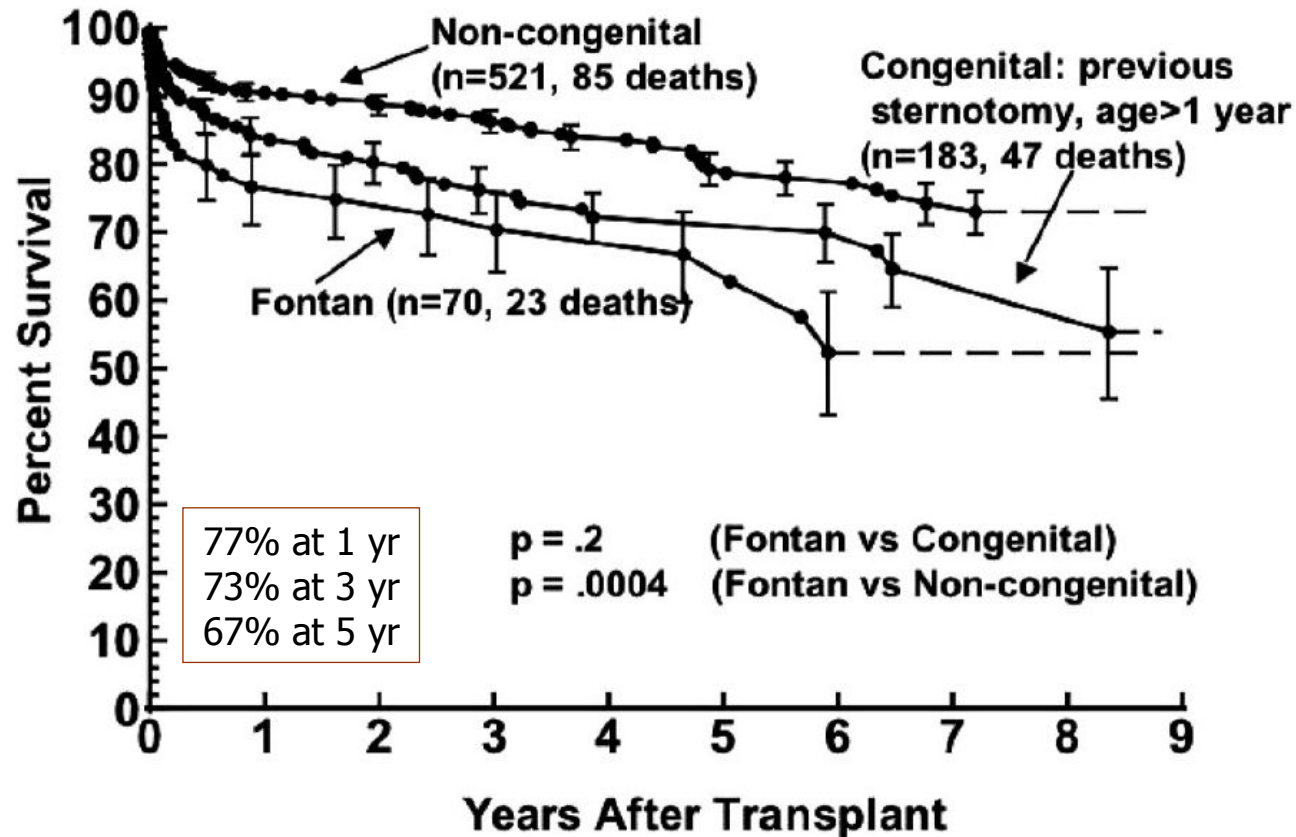
A Multi-Institutional Study

- 1993-2001, 17 Pediatric Heart Transplant Study centers
- 97 Fontan patient <18 yr of age listed
 - mean interval from Fontan to listing; 4.9 ± 4.4 yr (range, 0 to 15 yr)
 - 22% < 6 months after Fontan
 - 31% < 1 year
 - 26% between 1 and 6 years
 - 40% > 6 years
- 70 patients underwent heart transplantation
 - mean interval from Fontan to transplantation; 5.7 ± 4.4 yr (range, 0.02 to 15.6 yr)

Risk factors for death while awaiting transplantation

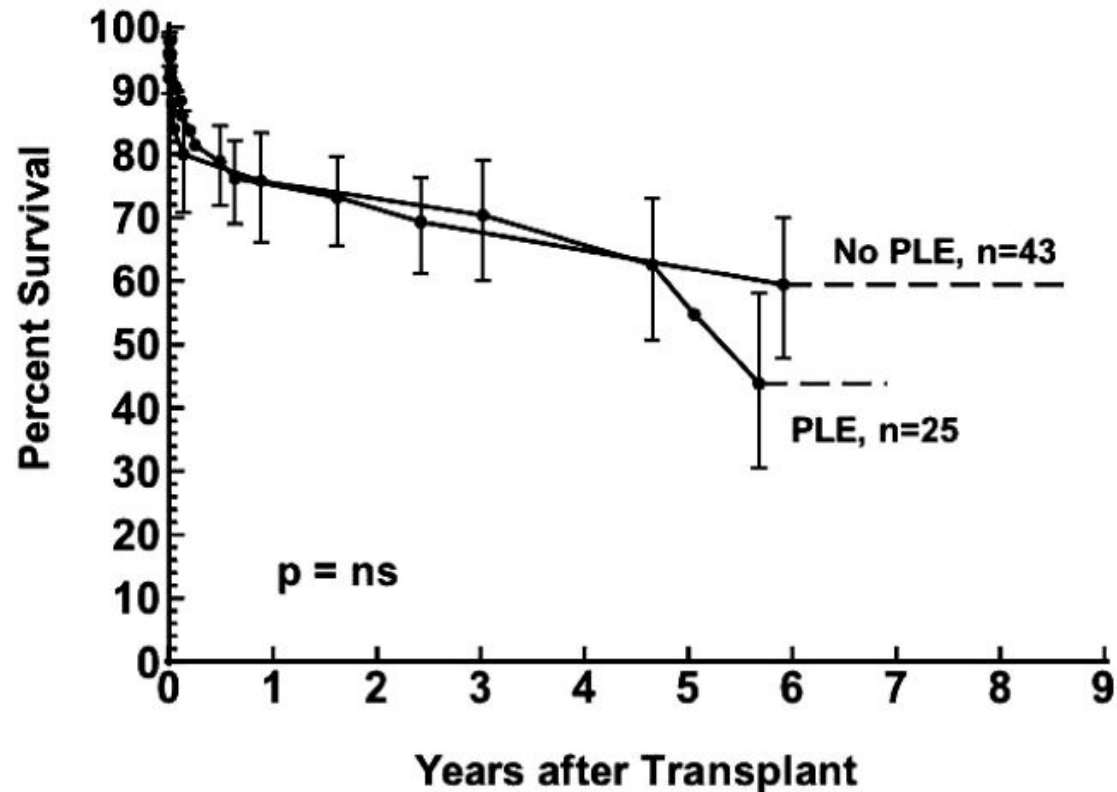


Outcome of Listing for Cardiac Transplantation for Failed Fontan A Multi-Institutional Study



Outcome of Listing for Cardiac Transplantation for Failed Fontan A Multi-Institutional Study

History of PLE



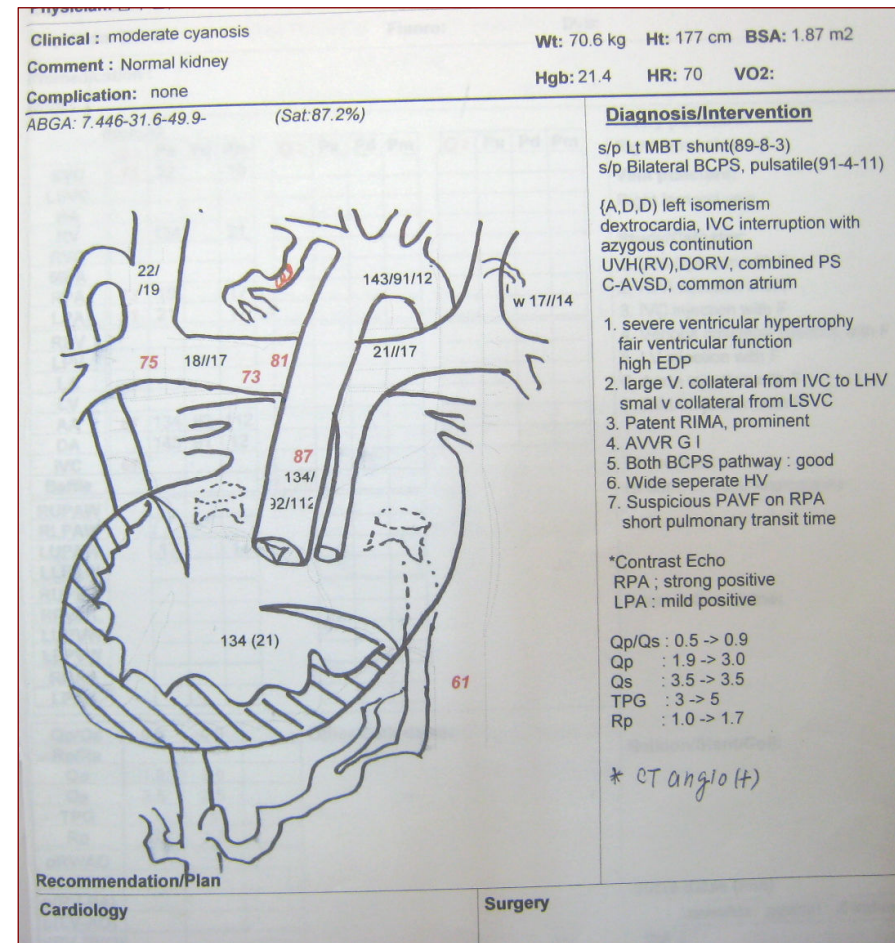
Pretransplant considerations in failing Fontan

- Multiple prior operations
- Hepatic dysfunction
- Cardiac & extracardiac vascular anatomy
- Evaluation of the extent of ventricular dysfunction
- Evaluation of pulmonary vascular disease & prediction of PVR
 - Acute right heart failure

Case

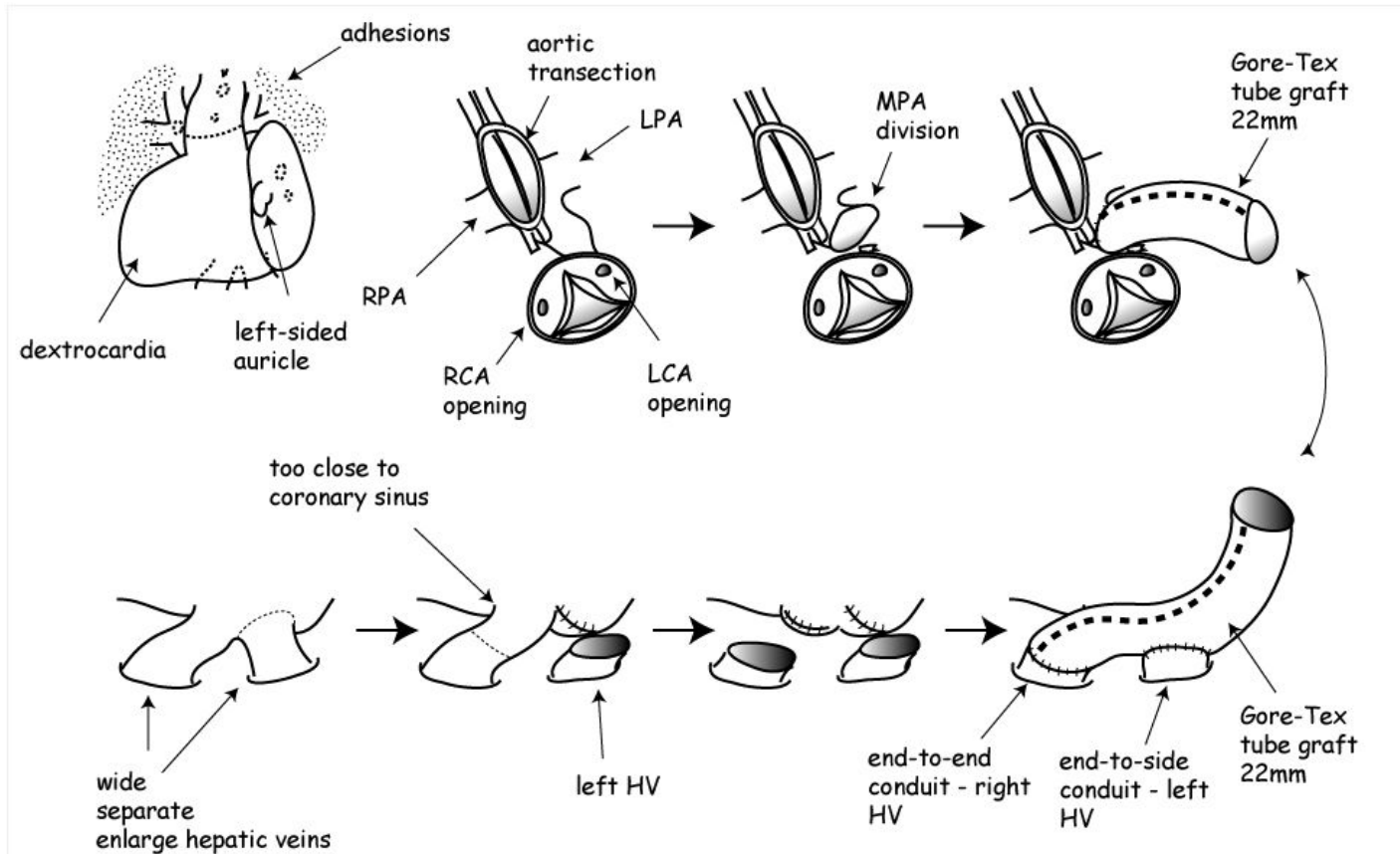
- Ventricle dysfunction with dysrhythmia -

- M / 19 (1987)
 - Left isomerism, bilateral SVC, dextrocardia
 - UVH, common AV valve
 - IVC interruption with azygos continuity
 - s/p LMBT shunt (89)
 - s/p Kawashima operation (91)
- Preoperatively
 - Moderate cyanosis
 - Pulmonary AV malformation, right
 - Severe ventricular hypertrophy with high VEDP (21mmHg)



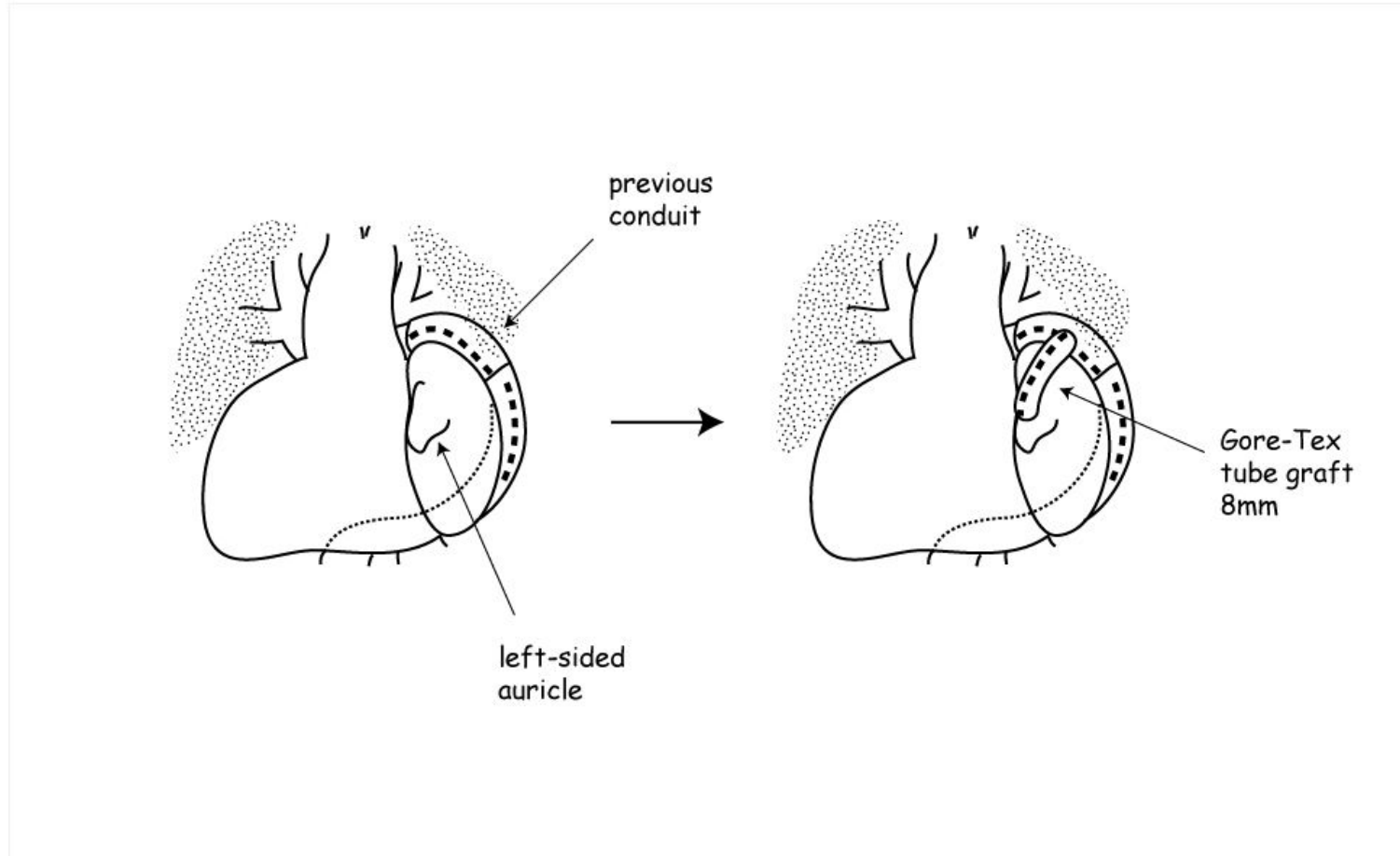
Hepatic vein inclusion

06-12-30

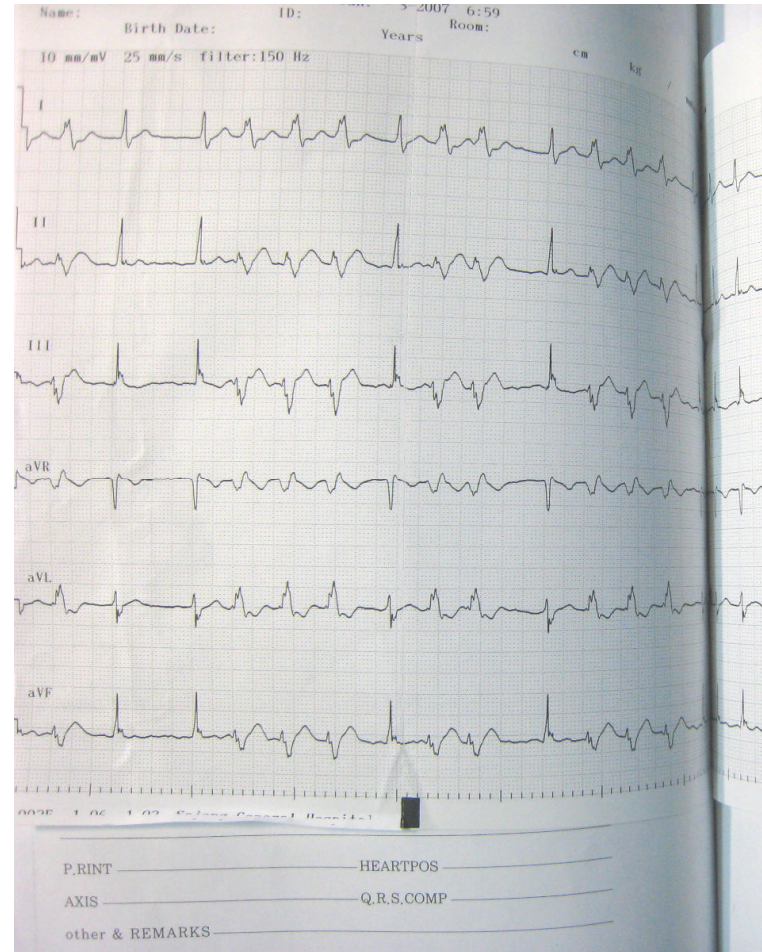
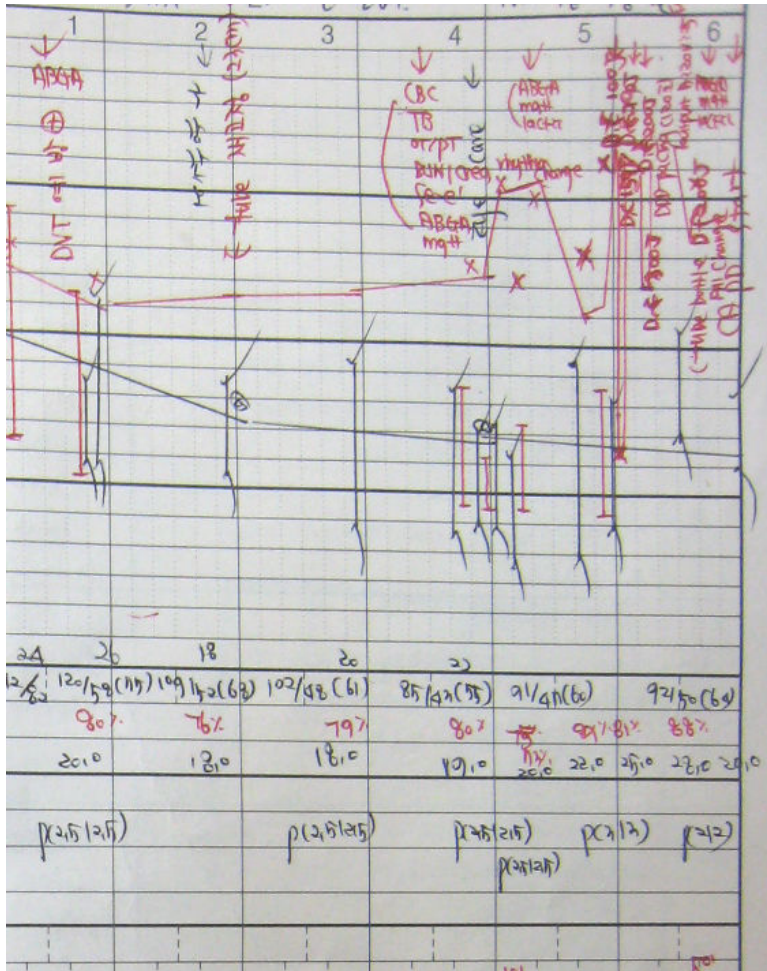


Fenestration

POD #2



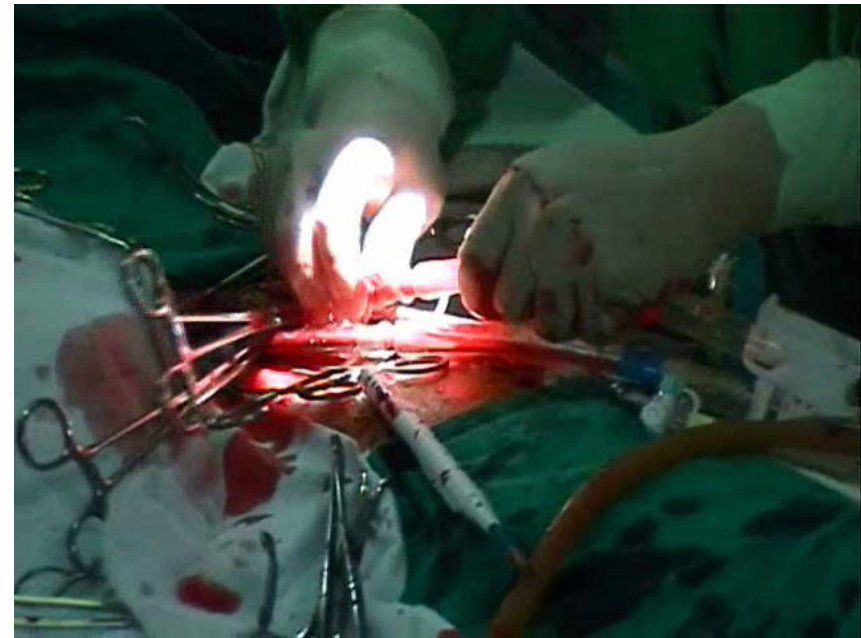
Ventricular tachyarrhythmia



Ventricle dysfunction with tachyarrhythmia



POD #0 Bleeding control
POD #2 LCO - fenestration (8mm)



POD #6 ECMO support with Esmolol
POD #10 ECMO weaning
POD #17 Extubation
POD #23 Transferred to general ward
POD #52 Discharged with sinus rhythm

Conclusion

- Recently, early outcomes for Fontan procedures have been improving outstandingly.
- Fontan circulation has various limitations inherently, so adequate and timely interventions should be advocated in failing Fontan patients.
- Also, current practices of TCPC such as lateral tunnel or extracardiac conduit Fontan procedure should be carefully followed up.