Single Ventricle

Neonatal Management

Emre Belli

The Fontan operation

Surgical repair of tricuspid atresia

Ann Chir Thorac Cardiovasc 1971;10:39-47.

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20 most frequently appearing citations for "advances" in pediatric cardiac surgery*

Support technologies

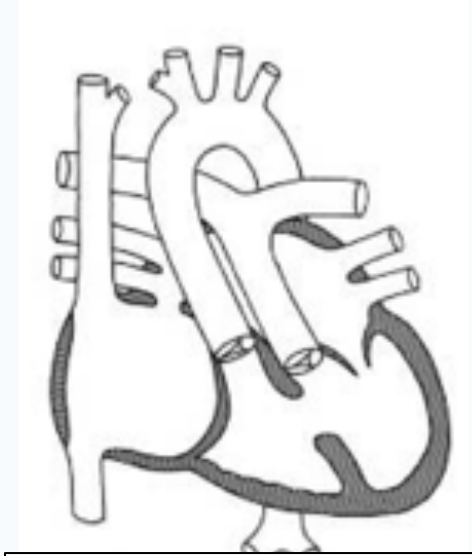
Operative strategies

*Karl TR et al 2015

The UVH or SV

- Nearly 10% of congenital heart defects belong to the group of functionally univentricular hearts.
- Heterogeneous group of cardiac malformations almost always determined by a dominant ventricle of either L or R ventricular morphology
- The natural history of the vast majority is characterized by a fatal course in the neonatal period or in early infancy.
- Only a few patients may survive into adulthood without surgical intervention.

The SV anatomy



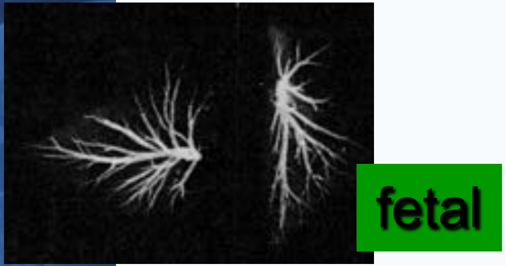
- Group 1:
  - univentricular AV connection or two AVV drain into a dominant ventricle
    - as in patients with double inlet ventricle

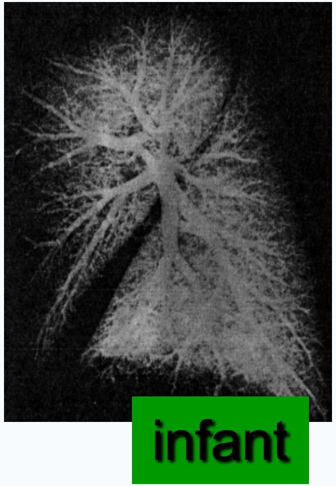
- Group 2:
  - Absence or severe stenosis of R or L AV connection and hypoplasia of corresponding ventricle
    - patients with tricuspid or mitral atresia, HLHS

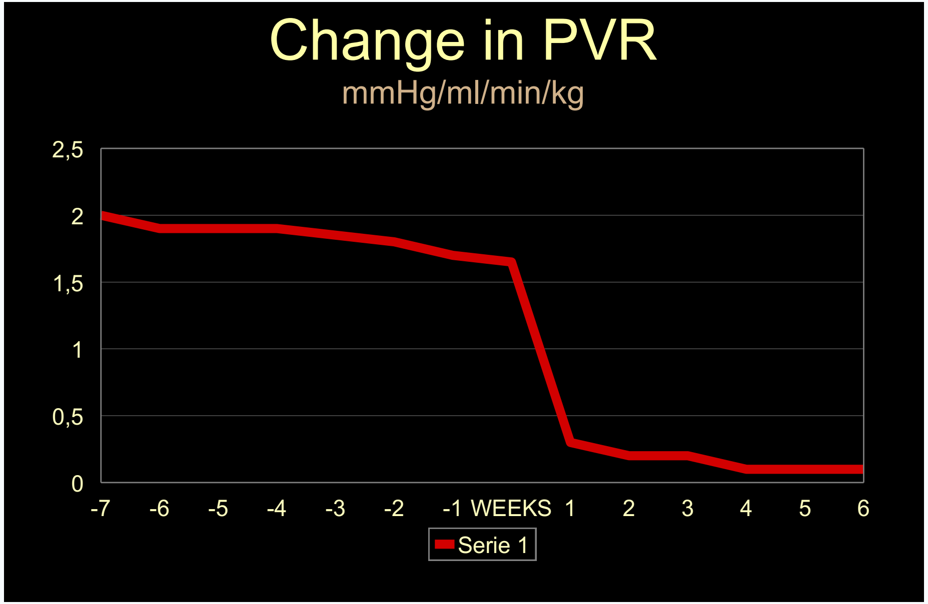
- Group 3:
  - Bilateral AV connection but either marked hypoplasia of the R or L ventricle (associated or not with abnormalities of AV or VA connection) precluding a 2V repair:
    - unbalanced AVSD
    - complex TGA / DORV

SV from anatomy to pathophysiology

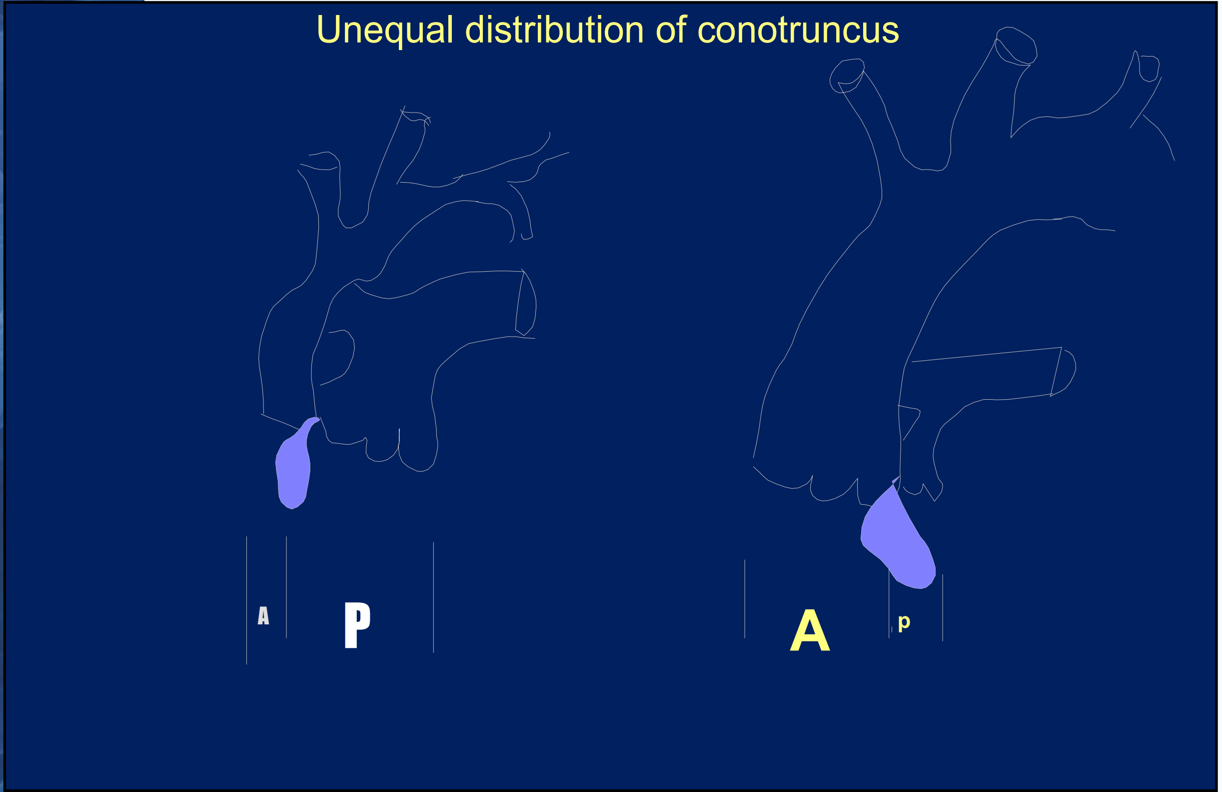
Lung maturation







SV from anatomy to pathophysiology



- Obstruction in systemic or pulmonary circulation
- Compensated by patent arterial duct (foetal circulation)

SV Pathophysiology

Obstruction

- Systemic circulation
- Pulmonary circulation
- Pulmonary venous return

Clinical Picture

- Decreased PBF
- Increased PBF
- Increased PBF & decreased SBF
- Balanced SBF & PBF

SV ... Neonatal diagnosis

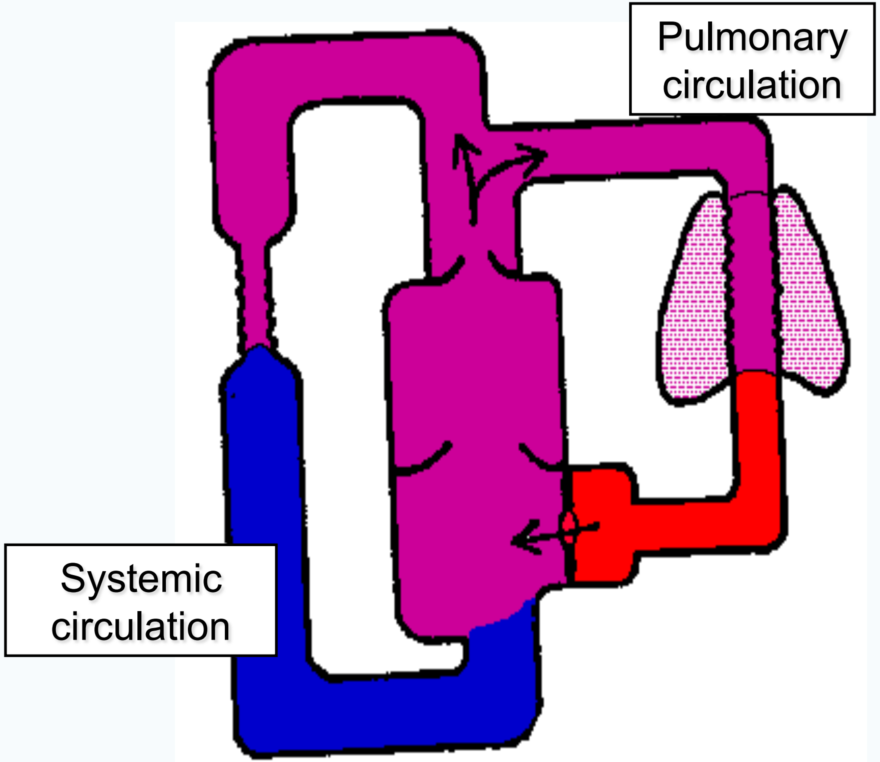
- Pressure gradients unuseful to assess obstruction severity
- Imaging of potential narrowing
  - Aortic arch
  - LVOTO
  - RVOTO
- Pulmonary artery & vein anatomy

- Echocardiography
- Sometimes CT scan
- Angiography rarely need

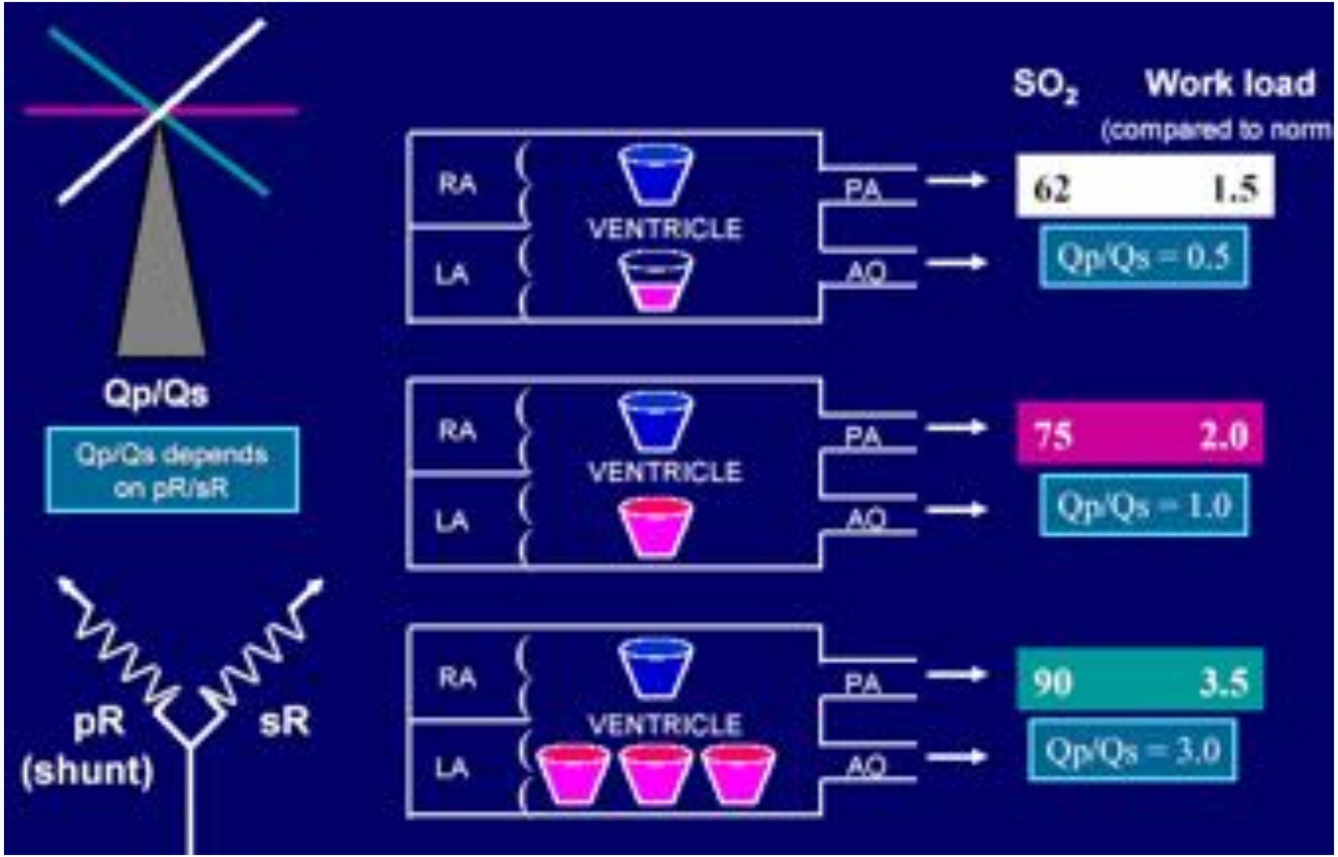
SV... Pathophysiology

Precarious model

- ✓ Single ventricle (RV)
- ✓ Shunt dependent
  - ASD
  - PDA
- ✓ Qtot = QP + QS
- ✓ ↑ ventricular work



SV... Pathophysiology



SV ... Initial medical treatment

- Diuretics – Vasodilators
- PGE
- Controlled ventilation
- Inotropic support

Avoid “Heroic Surgery”

SV initial palliation

- The goal of initial surgical palliation is to provide
  - unobstructed systemic outflow,
  - restricted pulmonary blood flow to maintain normal PAP and minimizing PA distortion
  - unobstructed systemic and pulmonary venous return to the heart

The SV surgical pathway

Single ventricle

Palliation
- S-P Shunt
- PAB
- Norwood - DKS

Bilateral cavopulmonary shunt: BPCS, HemiFontan

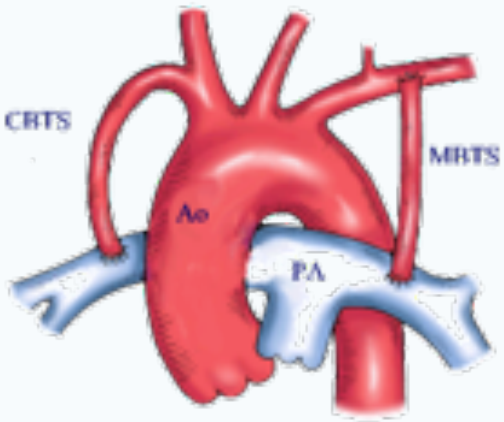
Fontan type op.

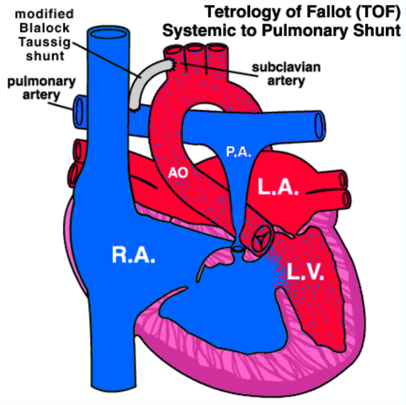
Eur J Cardiothorac Surg 2003;24:716-722

SV & reduced PBF: initial palliation

Systemic to pulmonary artery shunt

- Initial palliations to increase pulmonary blood flow in the presence of severely reduced pulmonary perfusion include aortopulmonary shunt procedures.





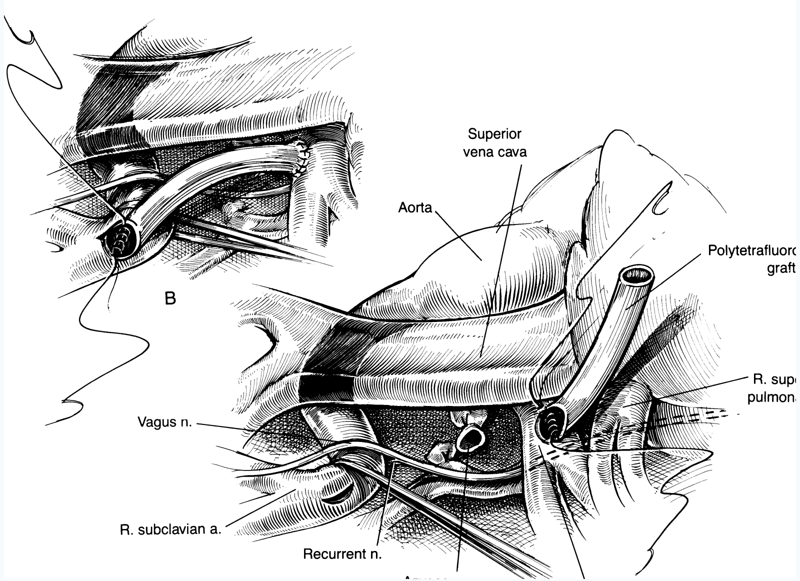
S-P shunt: technical aspect

When ?

Thoracotomy vs sternotomy

RPA vs LPA

Size



Shunt ... results

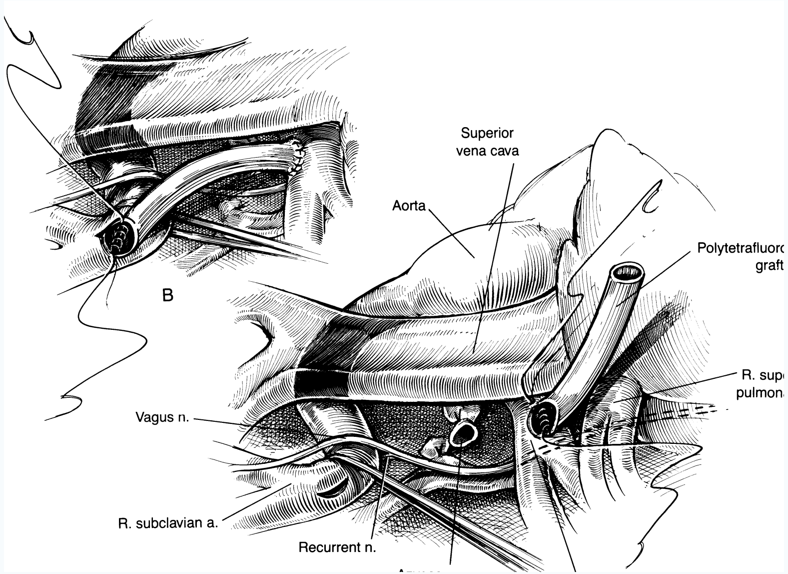
Pulmonary artery

- vessels distortion
- flow distribution

Ventricular function

- volume overload
- AVV regurgitation
- coronary perfusion

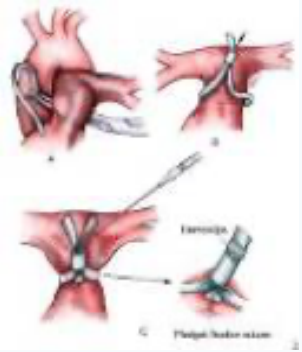
Mortality around 10%



SV & increased PBF: initial palliation

Pulmonary artery banding

- In case of unobstructed pulmonary outflow, excessive pulmonary blood flow might occur as pulmonary resistance falls in the first weeks of life.
- In this situation restriction of pulmonary blood flow can be achieved by surgical creation of a supravalvular stenosis ("pulmonary banding").



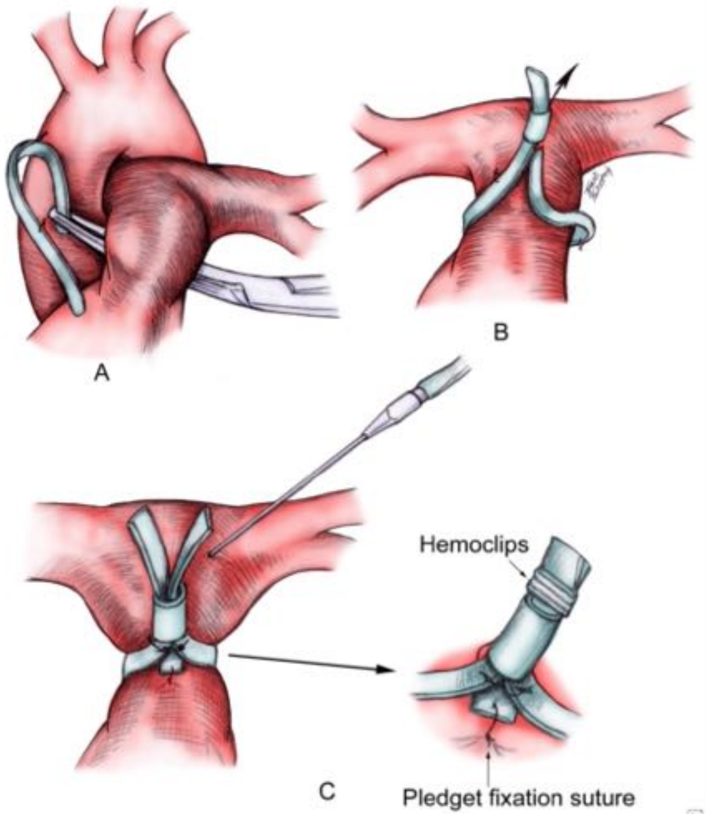
Trusler's formula: 24mm+ 1mm/kg

PAB: technical aspect

When ?

Thoracotomy vs sternotomy

Band calibration



PAB ... results

Pulmonary artery

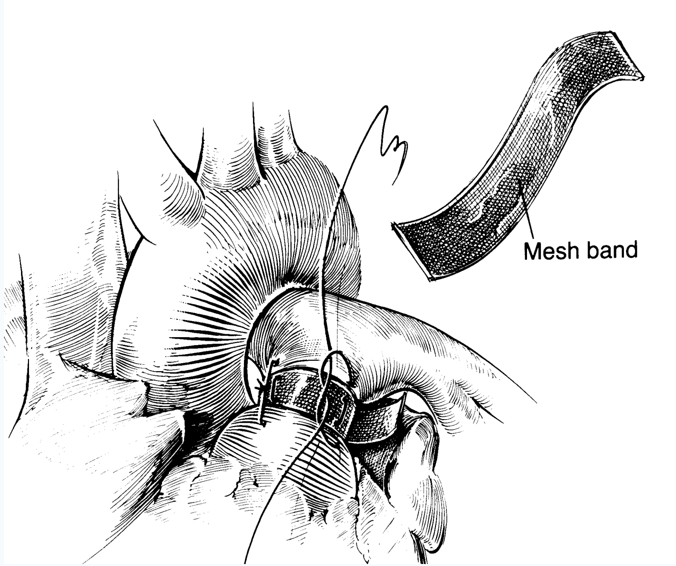
- vessels distortion
- flow distribution

Ventricular function

- pressure overload
- coronary perfusion

Mortality more than 10%

And ...

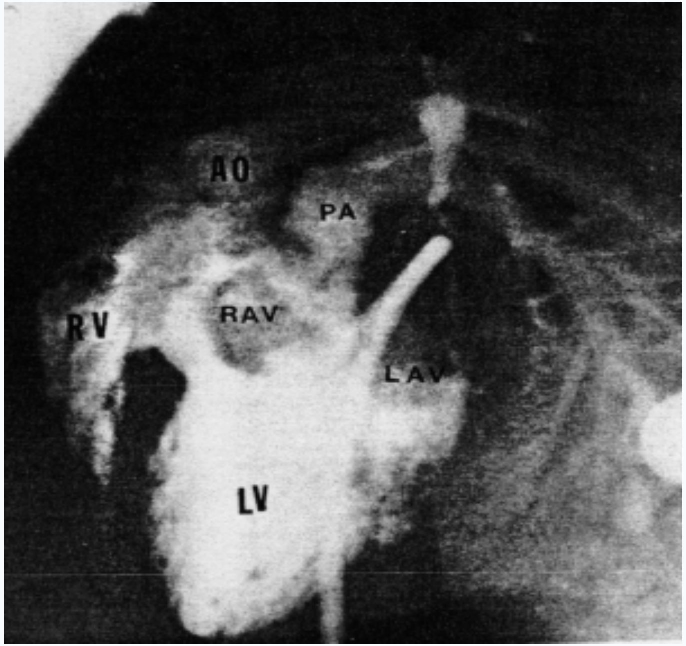


SV and subaortic obstruction

- PAB could unmask SAS

- PAB make progress SAS

- SAS and Arch obstruction

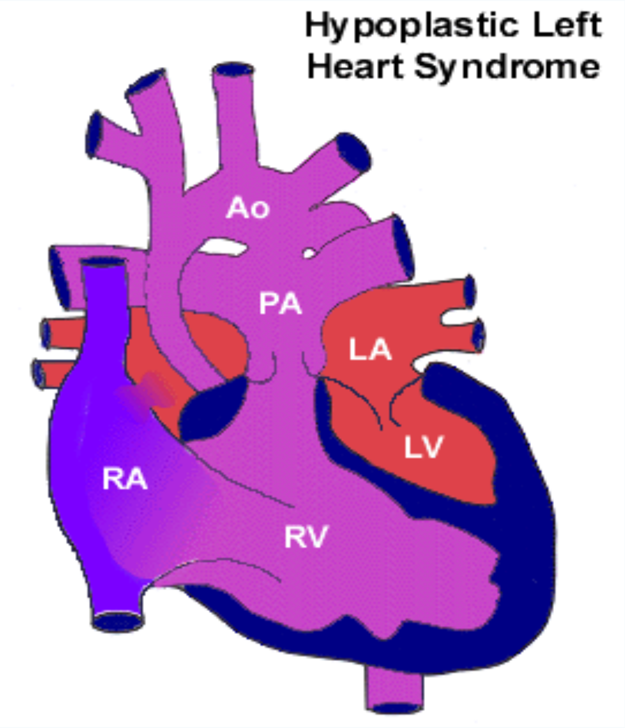


Di Donato et al, JTCVS 1993

HLHS… anatomical features

Definition

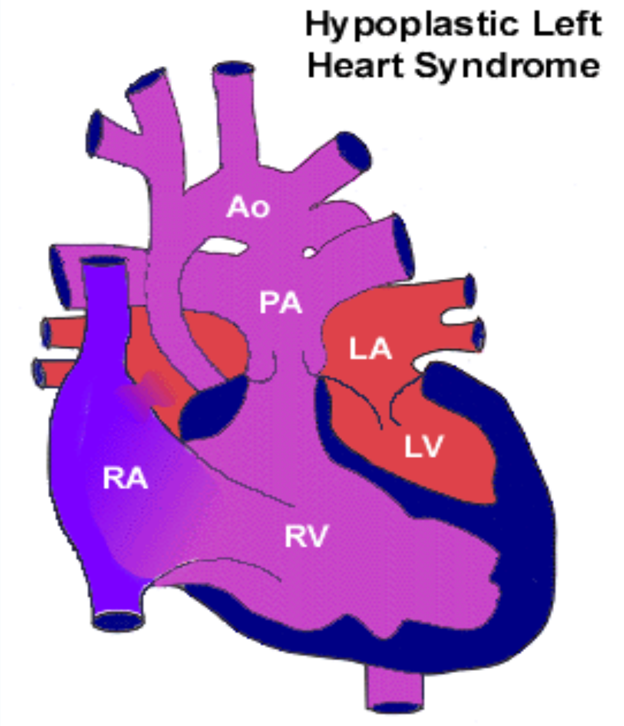
- Hypoplasia LV – Aorta complex
- Systemic flow obstruction
- RV dependent systemic perfusion (trough PDA)



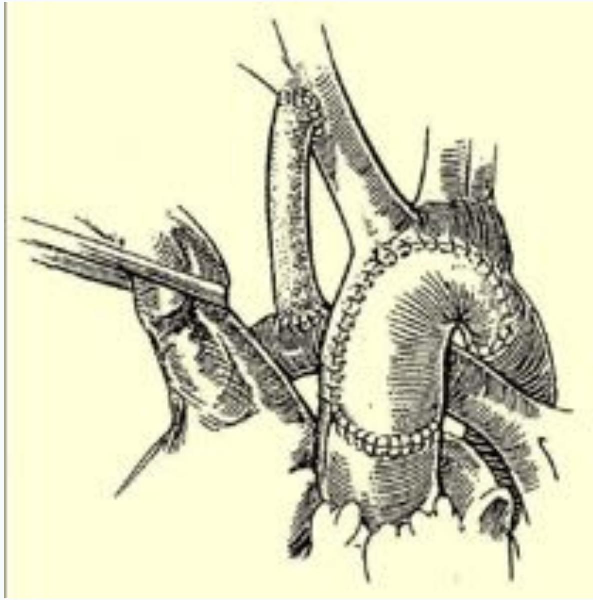
HLHS… anatomical features

Anatomical Variability

- Aortic Atresia (85%) or stenosis (15%)
- Mitral Atresia (66%) or stenosis (33%)
- Umbalanced DORV and/or CAV (10%)



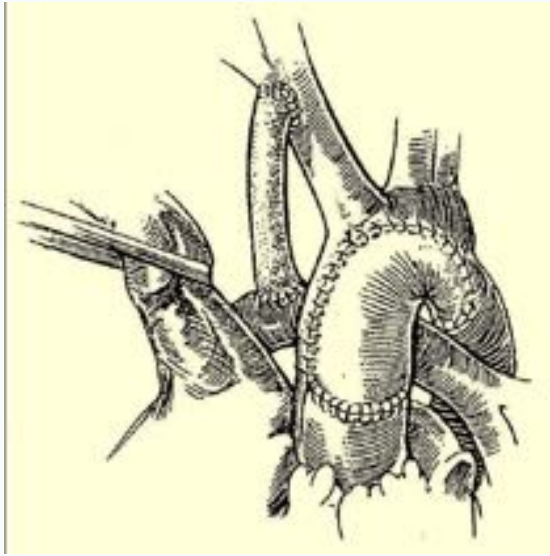
Norwood Stage I



Objectives

1. Suppression of PDA dependent systemic circulation
   – Unobstructed pathway RV → Aorta
2. Regulate PBF
   – Shunt / RVPA conduit
3. Avoid PV hypertension
   – Unrestricted P venous return

Norwood Stage I

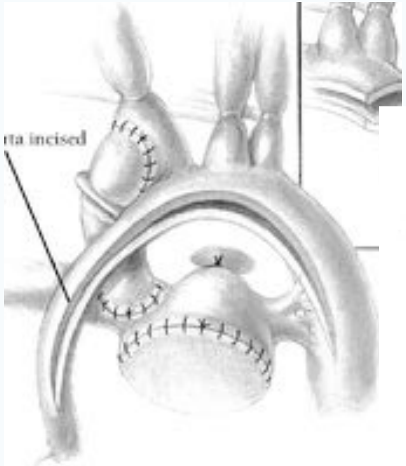


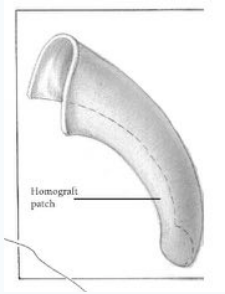
“Surgical step”

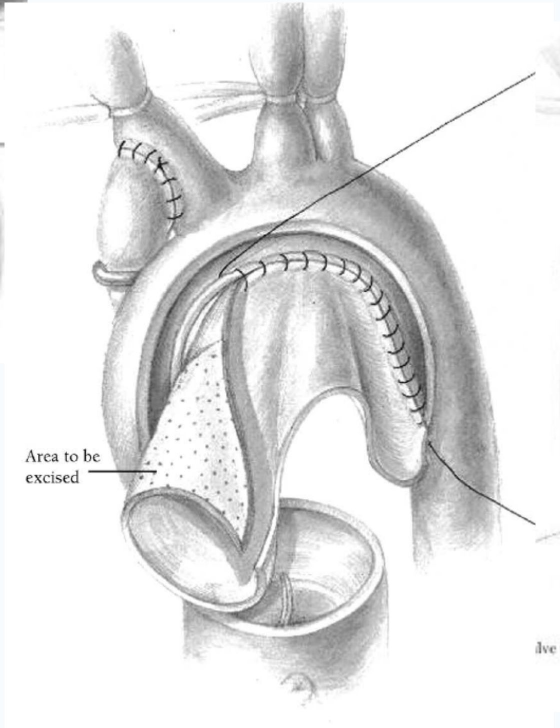
1. CPB strategy
2. ASD enlargement
3. Confluence PA Reconstruction
4. Reconstruction of neo-aorta
5. Establishment of PBF
6. CBP weaning and ICU management

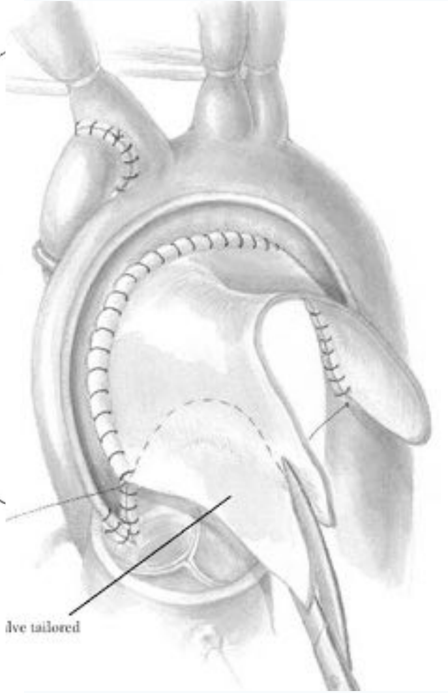
Norwood St I: neo-aorta reconstruction

“Our preferred technique”

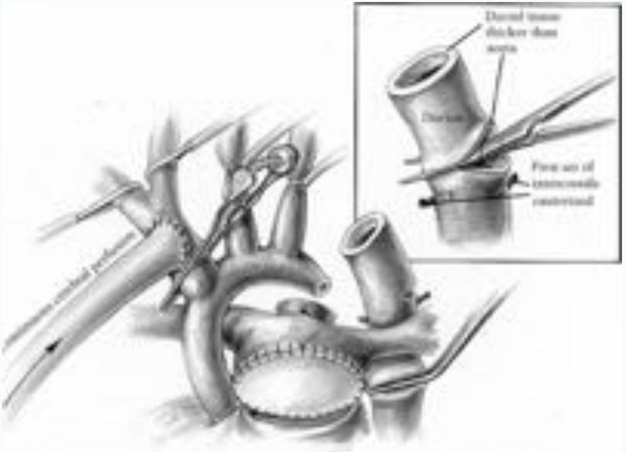




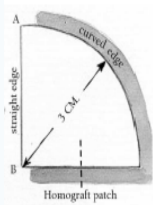


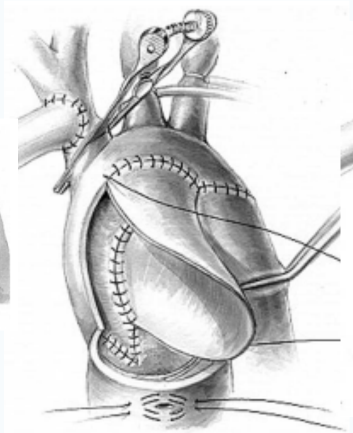


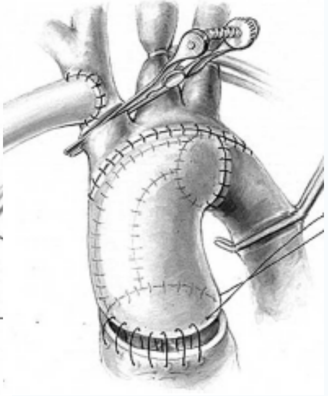
AA reconstruction… modified



Ductal tissue resection

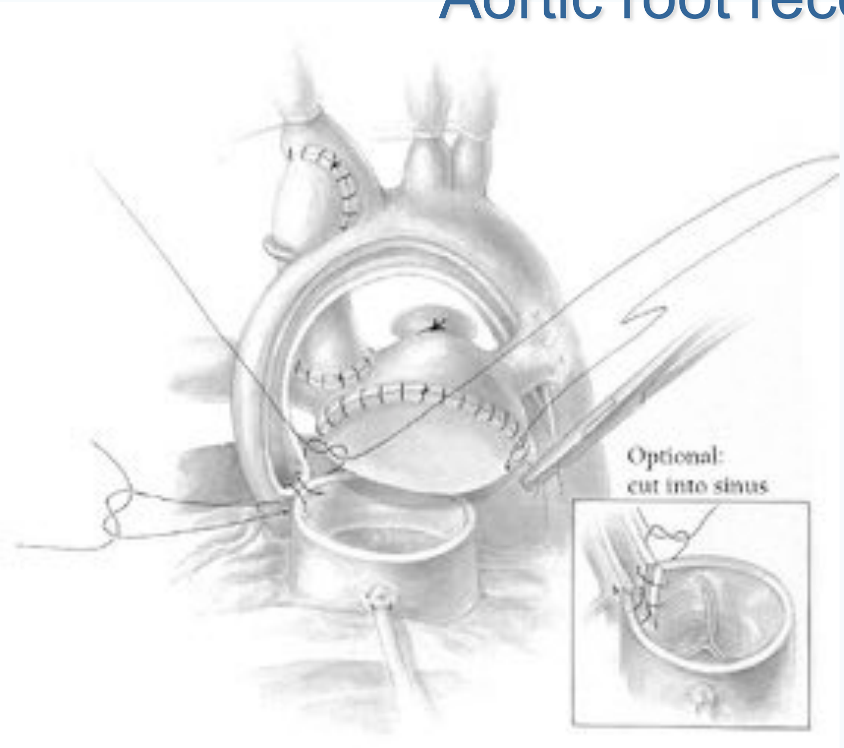


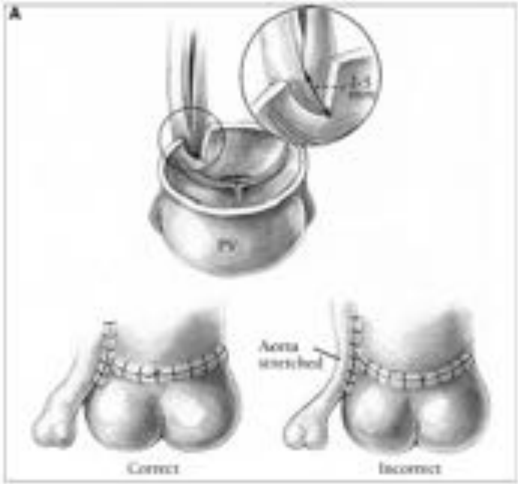




Norwood St I: neo-aorta reconstruction

Aortic root reconstruction





Norwood St I: neo-aorta reconstruction

- Impact on RV function
  - Ischemia
    - Coronary stretching
  - ↑ Ventricular afterload
    - Residual obstruction
  - ↑ Ventricular volume load
    - Increased PBF
    - Neo-ao regurgitation

Norwood St I: neo-aorta reconstruction

Ventricular Function Deteriorates With Recurrent Coarctation in Hypoplastic Left Heart Syndrome

Luis Alesandro Larrazabal, MD, Elif Seda Selamet Tierney, MD, David W. Brown, MD, Kimberlee Gauvreau, ScD, Vladimiro L. Vida, MD, Lisa Bergersen, MD, Frank A. Pigula, MD, Pedro J. del Nido, MD, and Emile A. Bacha, MD

Departments of Cardiac Surgery and Cardiology, Children's Hospital Boston, Harvard Medical School, Boston, Massachusetts

Background. Recurrent coarctation (re-CoA) after stage I palliation in hypoplastic left heart syndrome (HLHS) is deleterious. We studied whether re-CoA had an effect on ventricular systolic function.

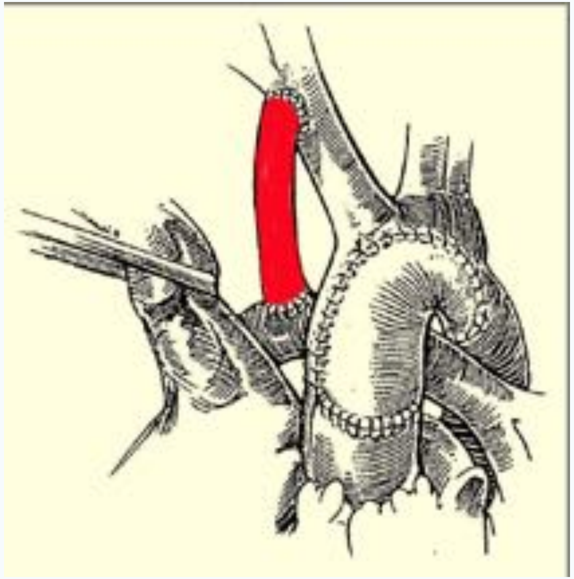
Methods. Retrospectively reviewed were HLHS patients surviving stage I Norwood palliation (stage I) and cavopulmonary shunt (CPS) between January 2004 and February 2007. Echocardiographic right ventricular fractional area change (RV-FAC) was used to evaluate ventricular systolic function after stage I, before CPS, and before Fontan procedures. Cardiac catheterization and magnetic resonance imaging data before CPS were reviewed to assess re-CoA, using a coarctation index (CI = isthmus diameter/descending aortic diameter).

Results. Fifty-one patients were included, and 21 had a CI of less than 0.75 (mean, 0.82 ± 0.19; 21). Twelve patients required arch balloon dilation between CPS and Fontan. The change of RV-FAC for all patients between stage I and CPS was −2.2% ± 9.6%. Pearson correlation coefficient demonstrated a significant correlation between lower CI values and lower RV-FAC at the pre-CPS echocardiogram (r = .35, p = 0.03); and lower CI values and greater decrease in RV-FAC between stage I and pre-CPS evaluation (r = 0.40, p = 0.018). At follow-up pre-Fontan, RV-FAC for patients who underwent balloon dilation for re-CoA recovered to a level that was inferior but not significantly different from that of patients who did not need balloon dilation.

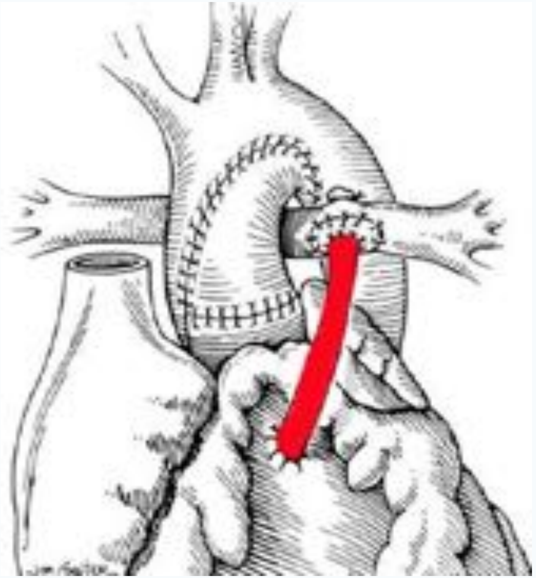
Conclusions. Recurrent aortic arch obstruction after stage I for HLHS is associated with worse RV systolic function at the time of stage II operation. Timely intervention on the re-CoA results in recovery of RV function.

(Ann Thorac Surg 2008;86:869–74)

Norwood St I: PBF establishment

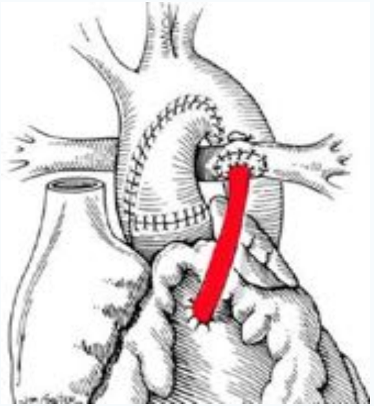


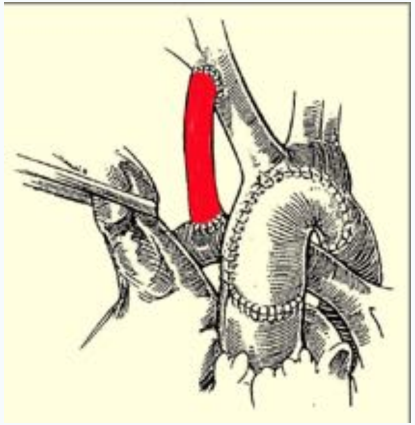
Diastolic steal



Ventriculotomy

Norwood St I: PBF establishment





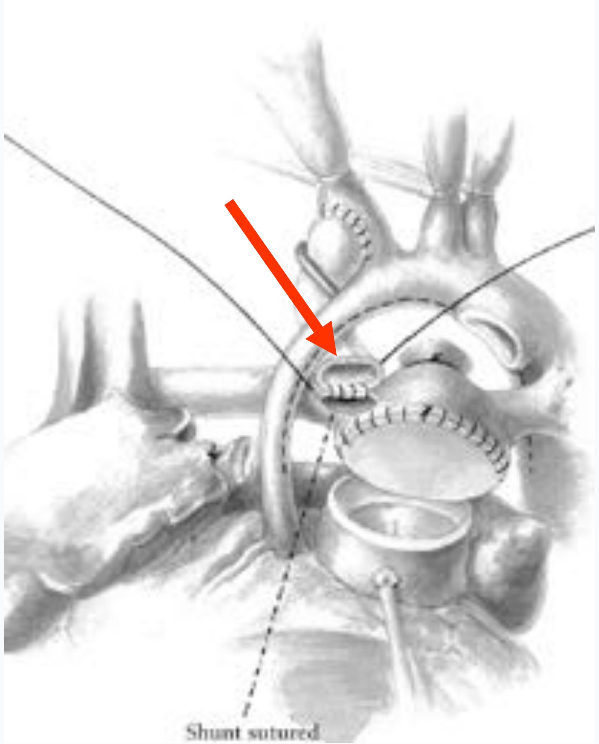
“first choice”

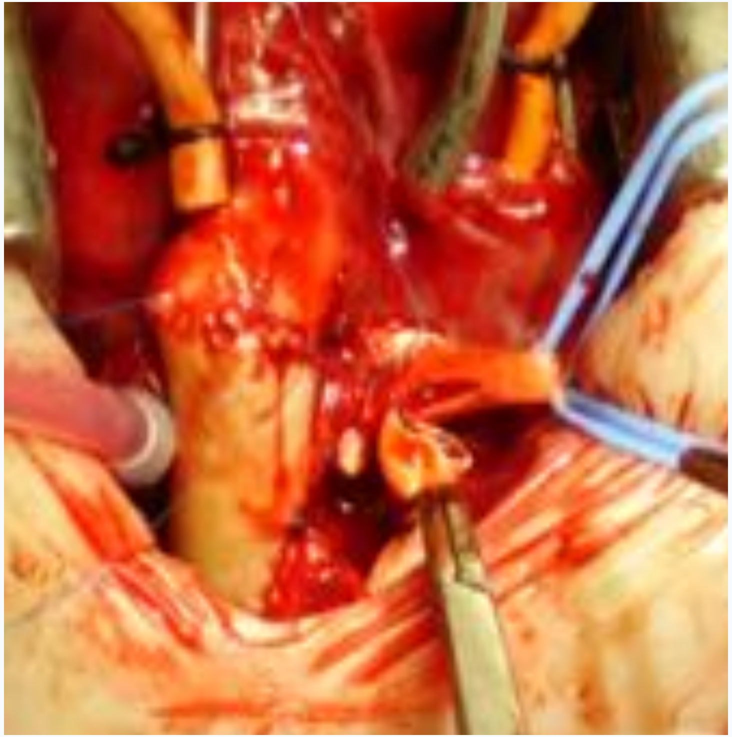
- < 2 Kg 4 mm, < 3,5 kg 5 mm, > 4 kg 6 mm
- “reinforced ring”
- 1V→2V conversion

“special situation”

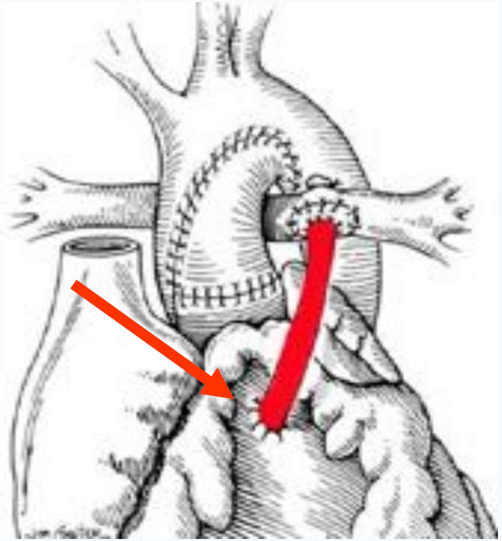
- HLHS equivalent
- 3,5mm < 3,5 Kg
- 4 mm > 3,5 kg

Norwood St I: S-P shunt





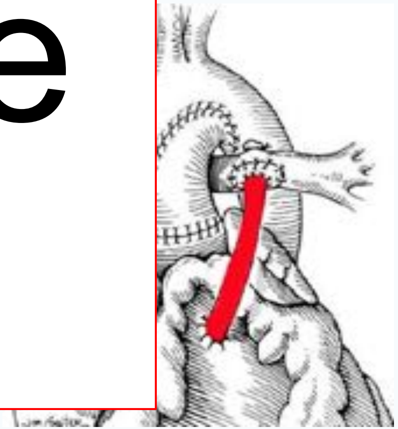
Norwood St I: RV-PA conduit



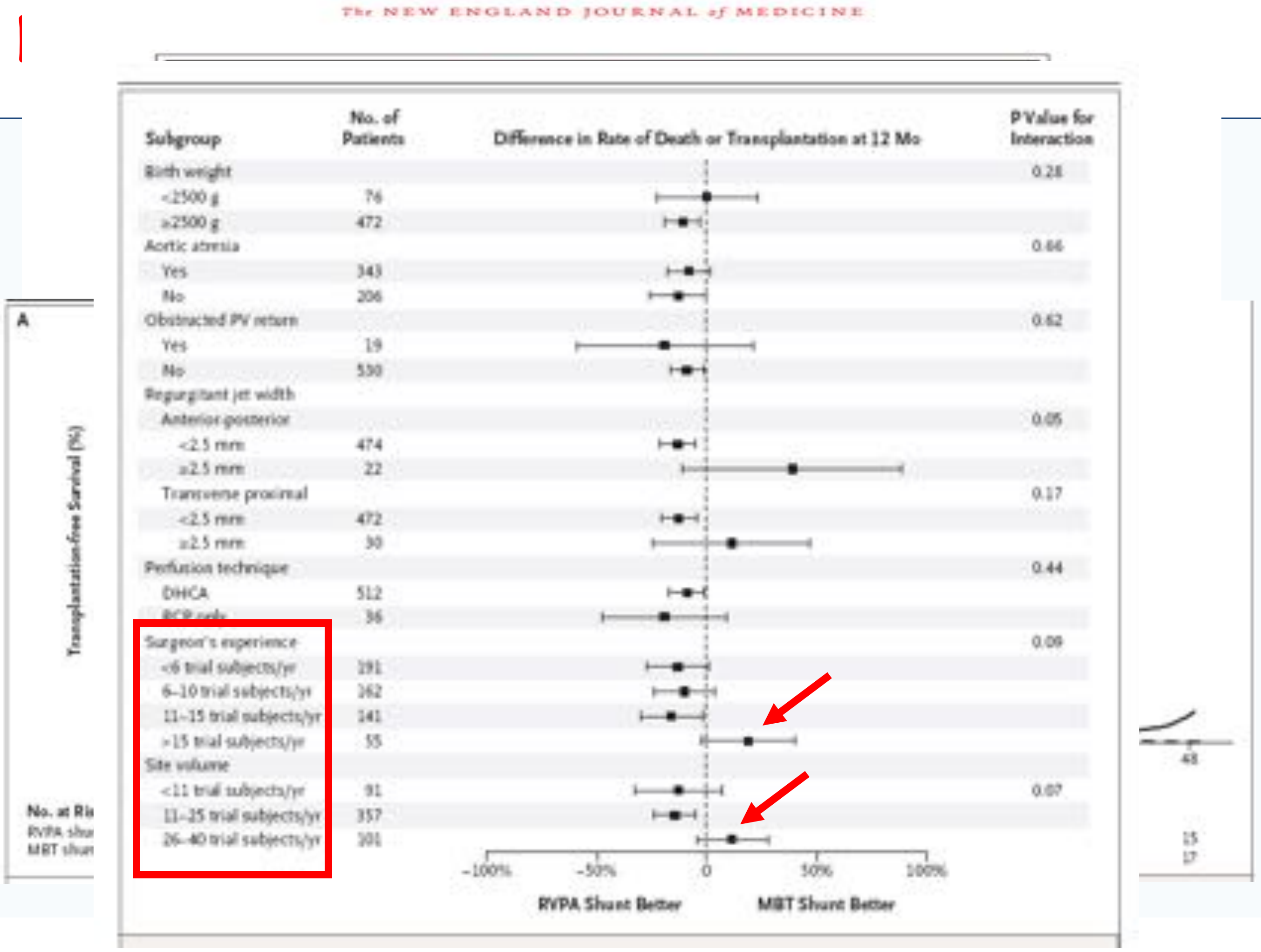
Norwood St I: shunt or RVPA conduit ?

- Early survival

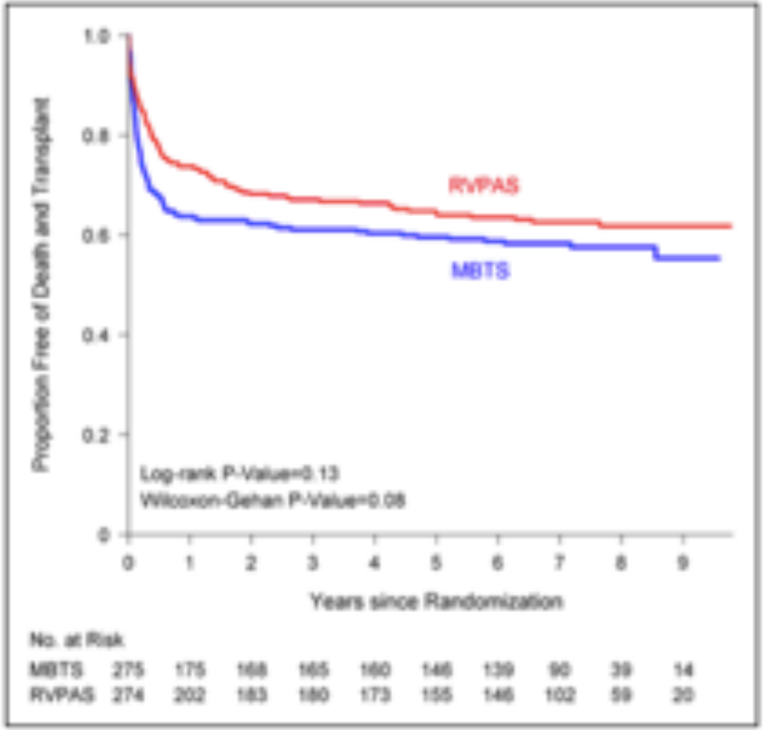
What's the best ?

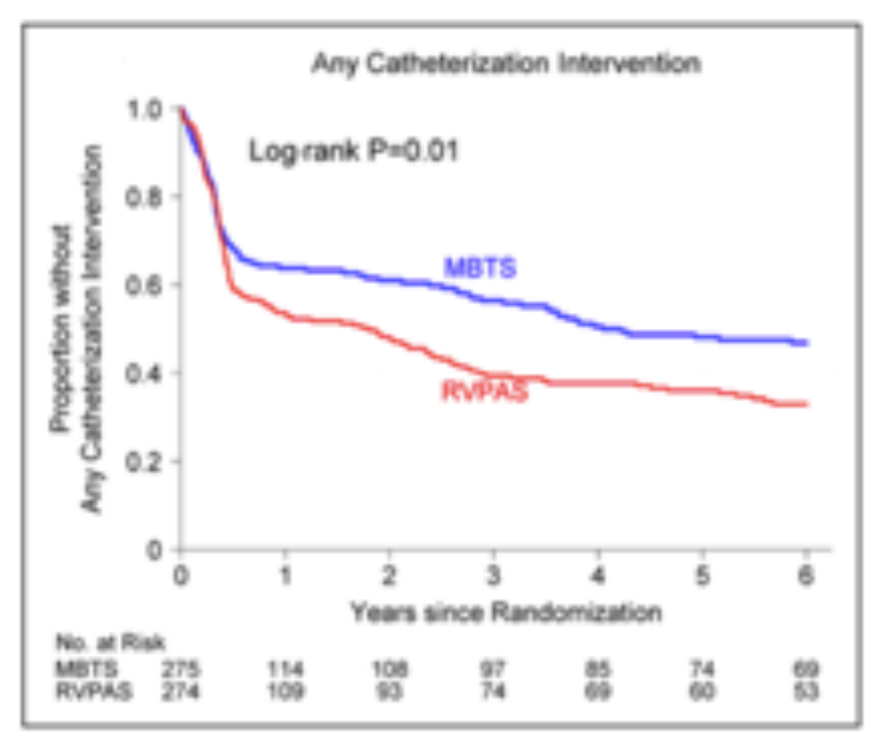


- Interstage mortality
- RV & TV function


The NEW ENGLAND JOURNAL of MEDICINE
Subgroup
No. of Patients
Difference in Rate of Death or Transplantation at 12 Mo
P Value for Interaction
Birth weight
<2500 g
76
≥2500 g
472
0.28
Aortic atresia
Yes
343
No
206
0.66
Obstructed PV return
Yes
19
No
530
0.62
Regurgitant jet width
Anterior-posterior
<2.5 mm
474
≥2.5 mm
22
0.05
Transverse proximal
<2.5 mm
472
≥2.5 mm
30
0.17
Perfusion technique
DHCA
512
RCP only
36
0.44
Surgeon's experience
<6 trial subjects/yr
191
6–10 trial subjects/yr
262
11–15 trial subjects/yr
141
>15 trial subjects/yr
55
0.09
Site volume
<11 trial subjects/yr
91
11–25 trial subjects/yr
357
26–40 trial subjects/yr
101
0.07
−100%
−50%
0
50%
100%
RVPA Shunt Better
MBT Shunt Better
A
Transplantation-free Survival (%)
No. at Risk
RVPA shunt
MBT shunt
48
15
17

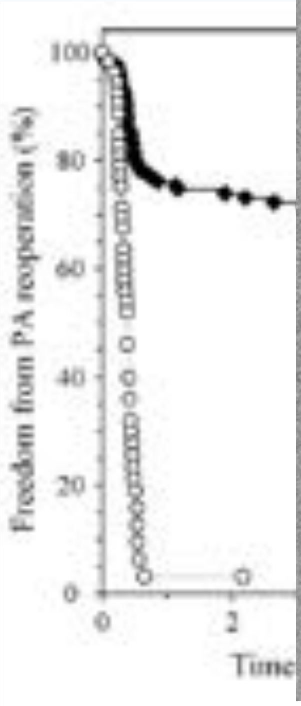
Norwood St I: SVR trial at 6 yrs





The Sano modification

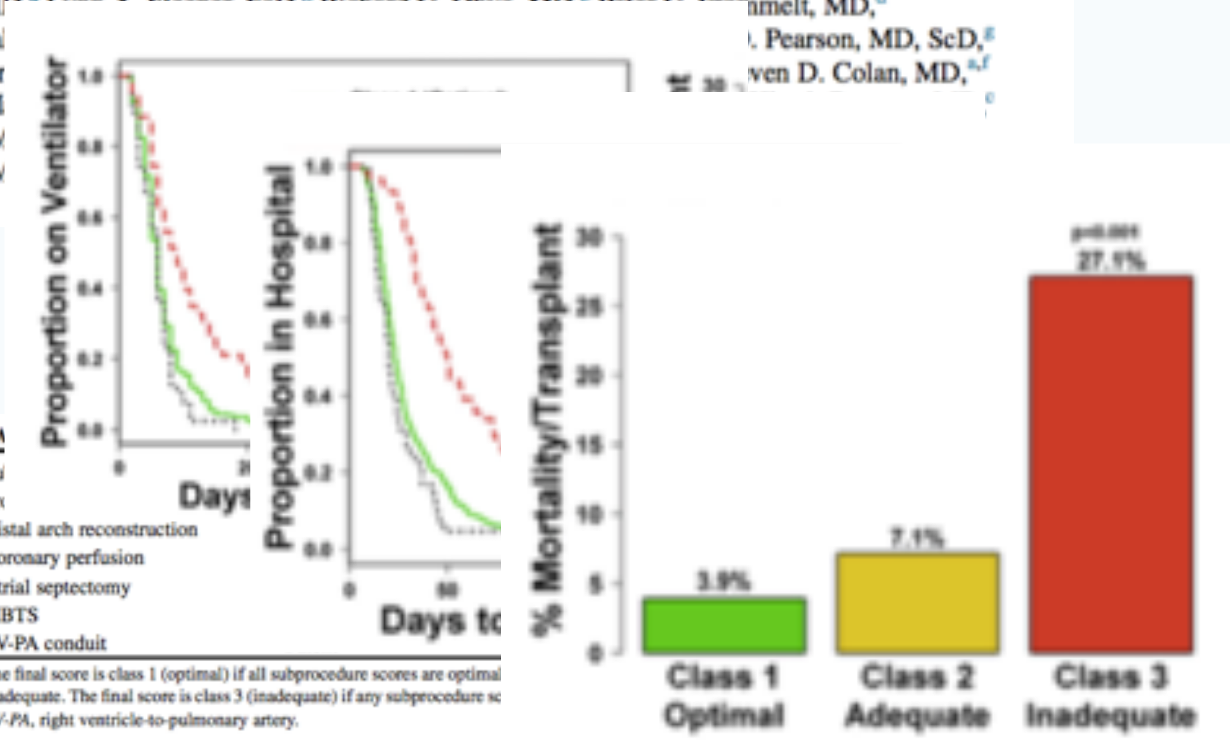
Be aware of the pulmonary artery stenosis!



M. Griselli et al. EJCTS 2006;30:930—935

Technical performance score is associated with outcomes after the Norwood procedure

Meena Nathan, MD ... mmelt, MD,[d]
Christopher A. Ca... . Pearson, MD, ScD,[g]
J. William Gaynor ... ven D. Colan, MD,[a,f]
Carolyn Dunbar-M...
Jeffrey P. Jacobs, M...
Scott M. Bradley, M...
Investigators



TA...
Su...
Pr...
Distal arch reconstruction
Coronary perfusion
Atrial septectomy
MBTS
RV-PA conduit

The final score is class 1 (optimal) if all subprocedure scores are optimal ... inadequate. The final score is class 3 (inadequate) if any subprocedure sc... *RV-PA*, right ventricle-to-pulmonary artery.

Norwood I… results

Risk factors

- Preoperative shock
- Prematurity (<36 w)
- Low birth weight (< 2,5 kg)
- Coexisting morbidity
- Syndrome
- Restrictive ASD
- TV regurgitation
- Anatomical subtypes
  - AA/MS → LV to coronary fistulas
- Low volume institution

San Francisco, 2007

- "… sometimes something new arrives and change everything …"

Circulation

JOURNAL OF THE AMERICAN HEART ASSOCIATION

(*Circulation*. 2002;105:1099-1103.)

Stenting of the Arterial Duct and Banding of the Pulmonary Arteries

Basis for Combined Norwood Stage I and II Repair in Hypoplastic Left Heart

Hakan Akintuerk, MD; Ina Michel-Behnke, MD; Klaus Valeske, MD; Matthias Mueller, MD; Josef Thul, MD; Juergen Bauer, MD; Karl-Juergen Hagel, MD; Joachim Kreuder, MD; Paul Vogt, MD; Dietmar Schranz, MD

Pediatr Cardiol 26:190–199, 2005

Pediatric Cardiology

Lessons Learned from the Development of a New Hybrid Strategy for the Management of Hypoplastic Left Heart Syndrome

M. Galantowicz,[1,2] J.P. Cheatham[1,3]

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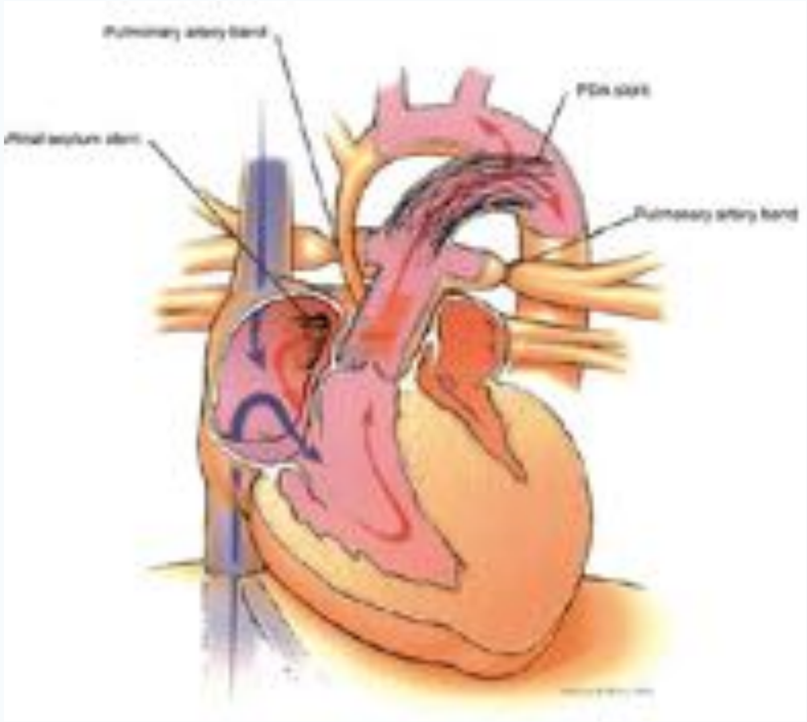
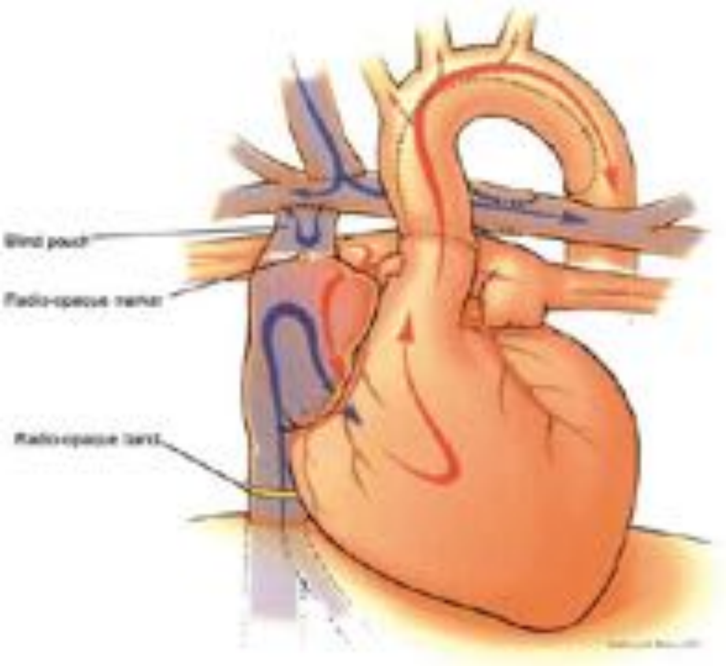
HLHS…hybrid procedure



Hybrid Stage I

- Bilateral PA Banding
- Ductal Stenting (simultaneous /successive)
- Advantage
  - avoidance of complex palliative operation in neonatal period
  - high-risk patients

HLHS…hybrid procedure



Comprehensive Stage II

- Arch & AA reconstruction
- PA reconstruction
- BCPA

- Advantage
  - Venous rather than arterial shunt
- Disadvantage
  - ASD management in interstage
  - Very complex procedure
  - PA hypoplasia & distortion

HLHS… hybrid procedure

“Is superior to Classical /Modified Stage I ?”

- Survival
- Fontan Candidancy
- Neurological Outcome
- Resource utilization

HLHS… hybrid procedure

"Is superior to Classical /Modified Stage I ?"

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Is a hybrid strategy a lower-risk alternative to stage 1 Norwood operation?

Travis J. Wilder, MD,[a] Brian W. McCrindle, MD,[b] Edward J. Hickey, MD,[c] Gerhard Ziemer, MD, PhD,[d] Christo I. Tchervenkov, MD,[e] Marshall L. Jacobs, MD,[f] Peter J. Gruber, MD, PhD,[g] Eugene H. Blackstone, MD,[h] William G. Williams, MD,[a] William M. DeCampli, MD, PhD,[i] Christopher A. Caldarone, MD,[c] and Christian Pizarro, MD,[j] for the Congenital Heart Surgeons' Society

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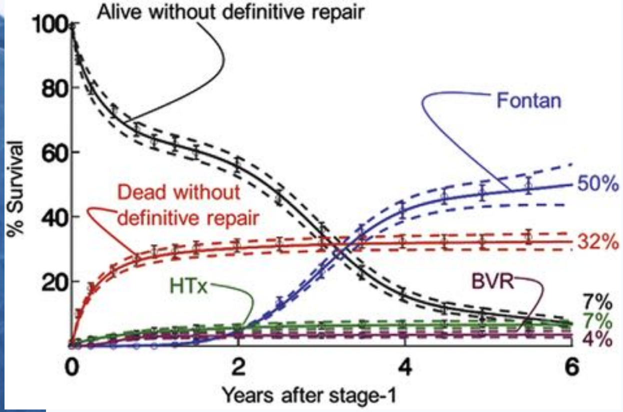
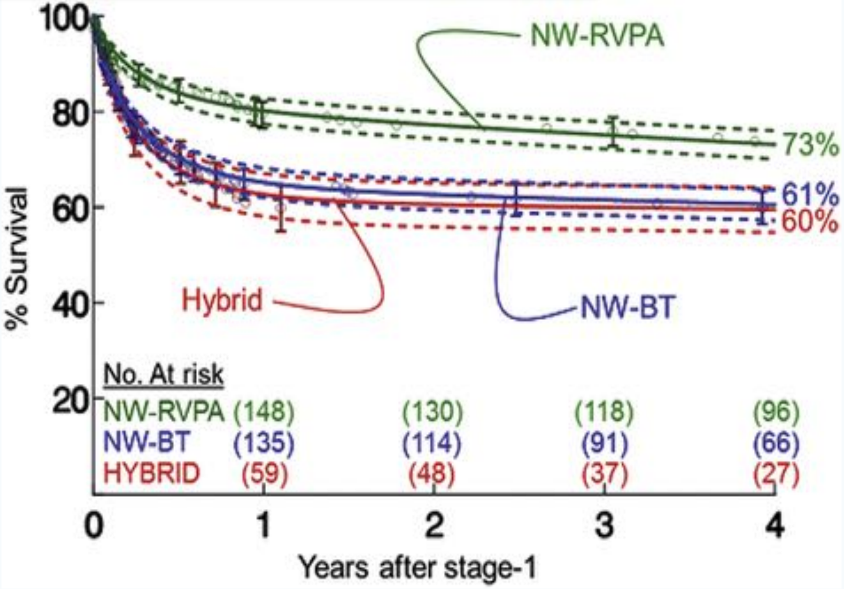
- 232 Norwood-Sano
- 222 Norwood-BTS
- 110 Hybrid

- Overall survival 65%





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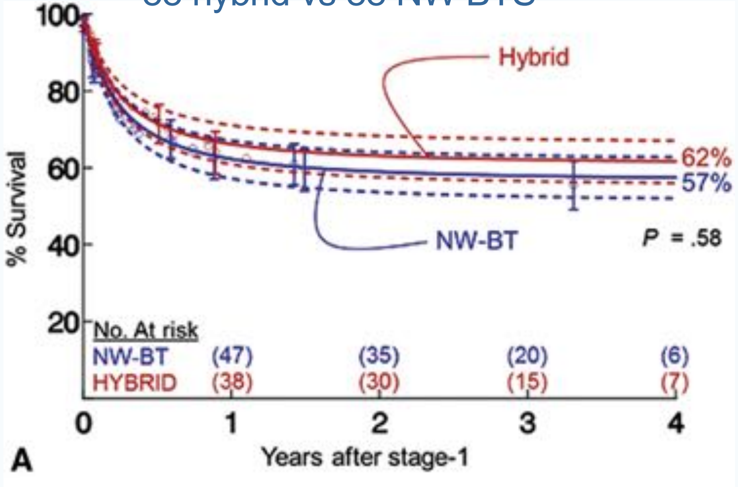
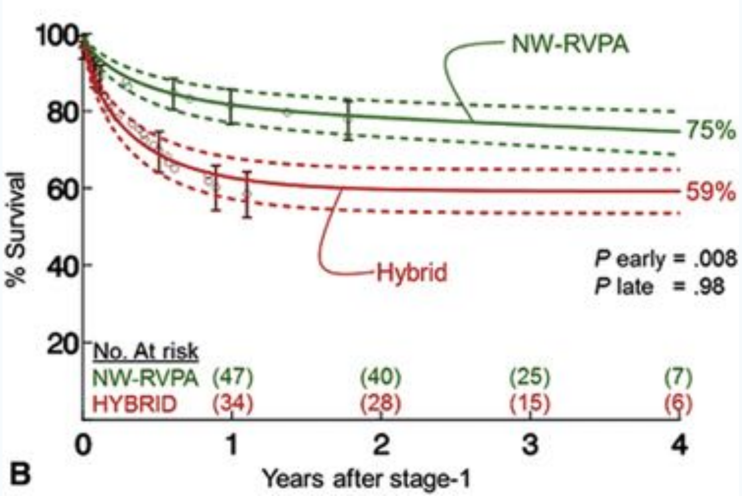
Propensity matched analysis

88 hybrid vs 88 NW BTS



81 hybrid vs 81 NW RVPA



Diastolic run-off

Is a hybrid strategy a lower-risk alternative to stage 1 Norwood operation?

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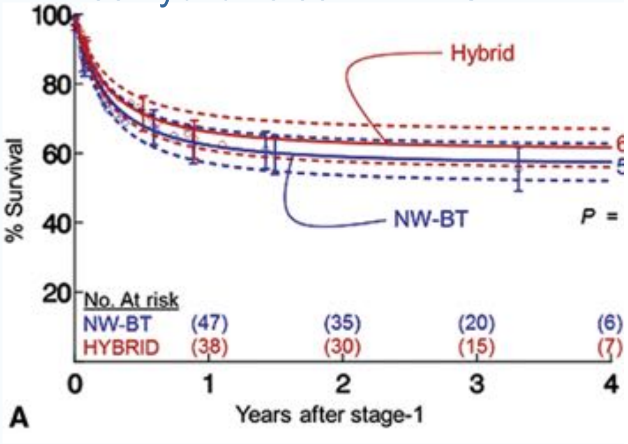

Diastolic run-o

TABLE 2. Incremental risk factors for death

Risk factor	Parameter estimate	P value	Reliability, %
All 564 neonates			
Early-phase risk factors			
NW-RVPA	−0.85	.001	71
Hybrid procedure*	1.6	.025	—
Birth weight (inverse transformation), kg†	7.7	<.001	67
Ascending aorta index, cm/m^2	−0.29	.039	89
Aortic valve atresia	0.45	.044	58
Interaction, procedure and birth weight‡	−5.0	.01	—
Constant phase risk factors			
Mitral valve z-score	−0.09	.099	40
Matched hybrid and NW-BT neonates (n = 176)			
Early phase risk factors			
Hybrid procedure*	−0.63	.53	43
Birth weight (inverse transformation), kg†	3.3	.002	60
Ascending aorta index, cm/m^2	−2.6	.01	47
Matched hybrid and NW-RVPA neonates (n = 162)			
Early-phase risk factors			
NW-RVPA	−2.4	.02	70
Birth weight (inverse transformation), kg†	3.6	.001	66
Smaller LVOT diameter z-score	−1.8	.05	74

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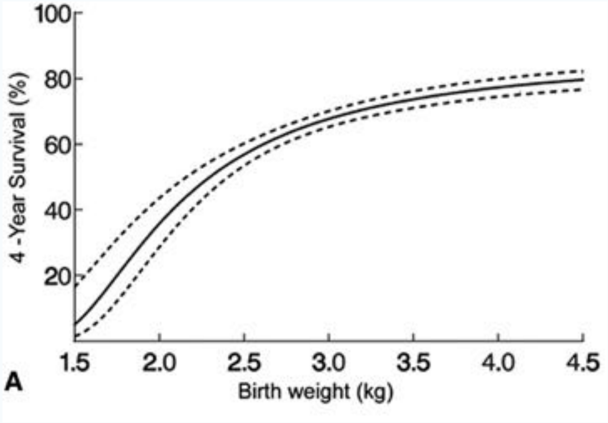
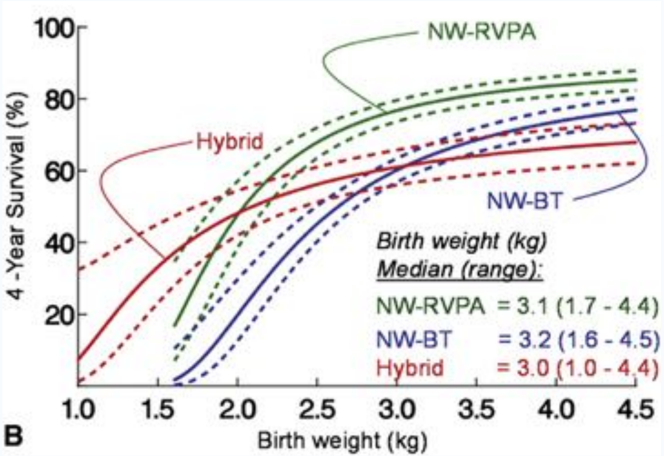
Impact of low-weight



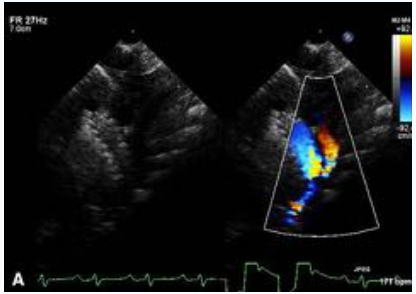


..maybe

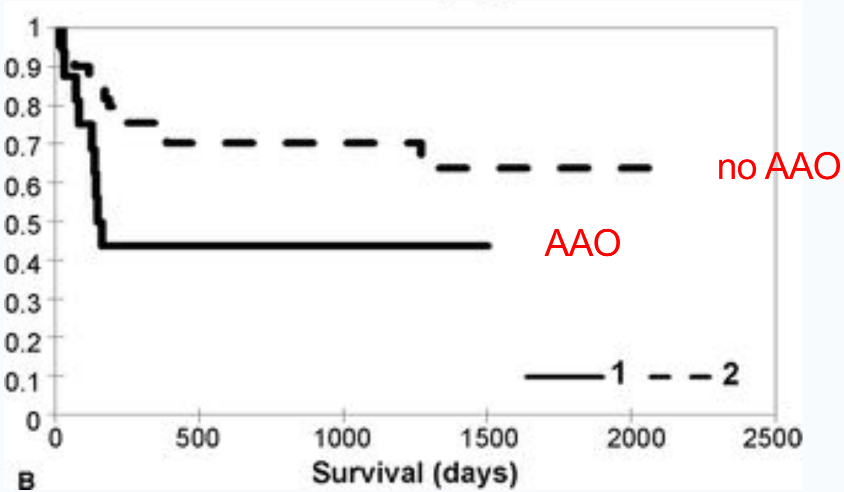
The Retrograde Aortic Arch in the Hybrid Approach to Hypoplastic Left Heart Syndrome

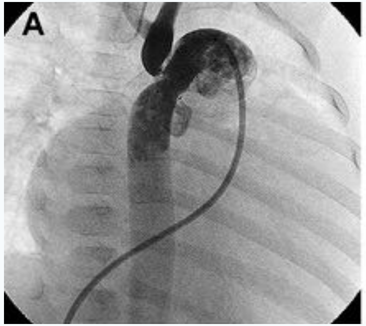
Serban C. Stoica, MD, Alistair B. Philips, MD, Matthew Egan, MD,
Roberta Rodeman, RN, Joanne Chisolm, RN, Sharon Hill, ACNP,
John P. Cheatham, MD, and Mark E. Galantowicz, MD

Nationwide Children's Hospital, Columbus, Ohio



Overall survival





(Ann Thorac Surg 2009;88:1939–47)

HLHS… hybrid procedure

"Is superior to Classical /Modified Stage I ?"

- Survival
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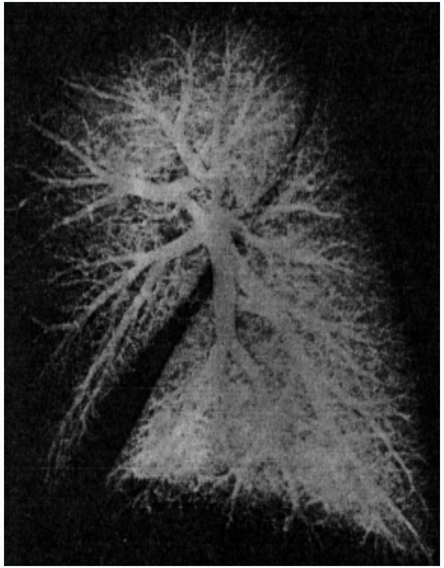
Optimal outcome from a Fontan procedure when

Ventricular function (systolic-diastolic)
and geometry are nearly normal as possible

&

Pulmonary vascular architecture and physiology is normal and mature



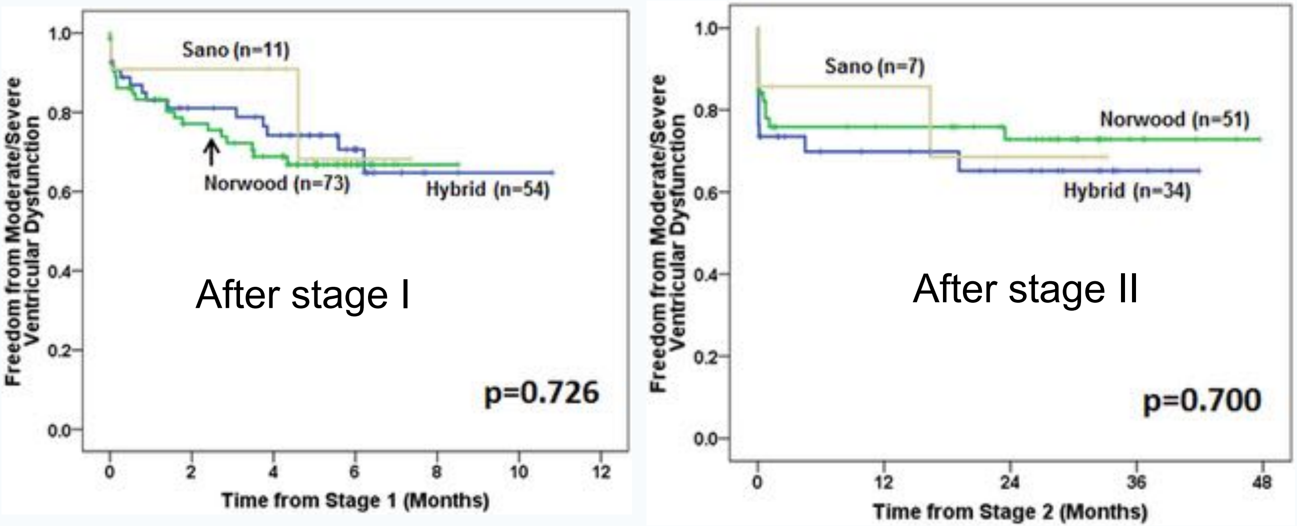
"The two commandments". Hosein RBM et al. Eur J Cardioth Surg 2007; 31: 344-53

HLHS… hybrid & SV function

Surgical Palliation Strategy Does Not Affect Interstage Ventricular Dysfunction or Atrioventricular Valve Regurgitation in Children With Hypoplastic Left Heart Syndrome and Variants

(*Circulation*. 2013;128[suppl 1]:S205-S212.)

Devin Chetan, HBA; Yasuhiro Kotani, MD, PhD; Frederic Jacques, MD, MSc; Jeffrey A. Poynter, MD;
Lee N. Benson, MD; Kyong-Jin Lee, MD; Rajiv R. Chaturvedi, MD, PhD;
Mark K. Friedberg, MD; Glen S. Van Arsdell, MD; Christopher A. Caldarone, MD;
Osami Honjo, MD, PhD



Improved outcomes with the comprehensive stage 2 procedure after an initial hybrid stage 1

Mark Galantowicz, MD,[a,b,d] and Andrew R. Yates, MD[a,c,e]

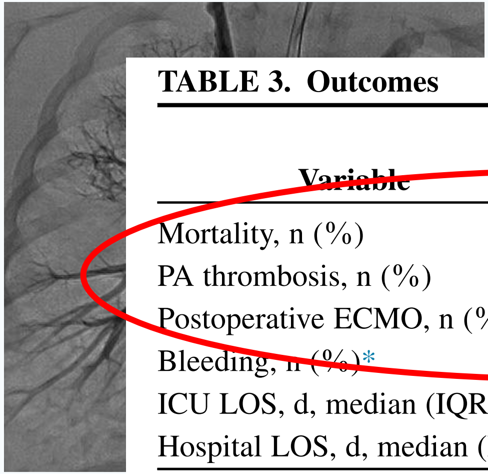
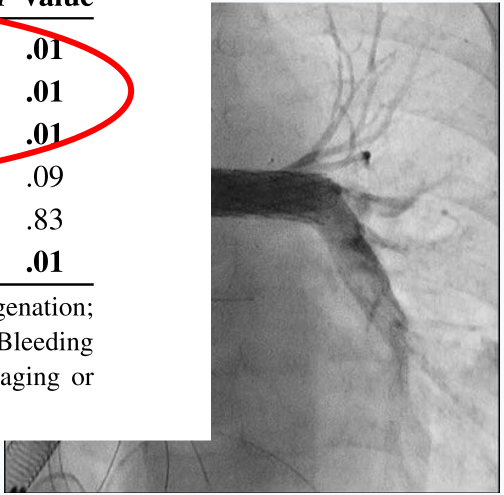


TABLE 3. Outcomes

Variable	Pre-protocol (n = 64)	Post-protocol (n = 55)	*P* value
Mortality, n (%)	12 (19)	2 (4)	**.01**
PA thrombosis, n (%)	7 (11)	0	**.01**
Postoperative ECMO, n (%)	7 (11)	0	**.01**
Bleeding, n (%)*	10 (16)	3 (5)	.09
ICU LOS, d, median (IQR)	4.5 (3-9.75)	5 (3-10)	.83
Hospital LOS, d, median (IQR)	9 (7-16.75)	13 (8-22)	**.01**

Significant *P* values are in bold type. *ECMO*, Extracorporeal membrane oxygenation; *ICU*, intensive care unit; *LOS*, length of stay; *IQR*, interquartile range. *Bleeding complication was defined as evidence of a new hemorrhage on brain imaging or systemic bleeding requiring a transfusion.



High rate of PA stenosis

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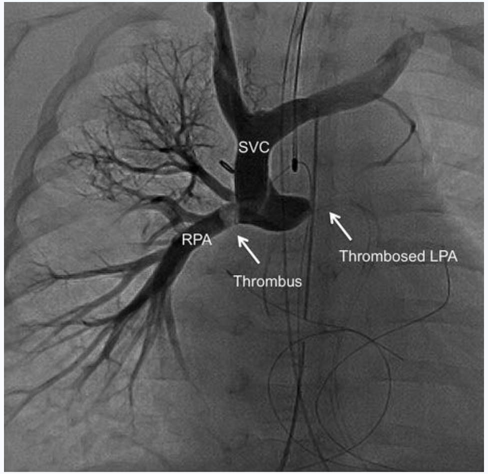
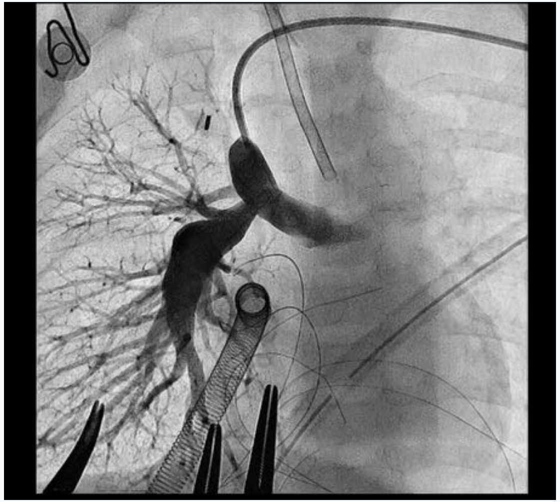
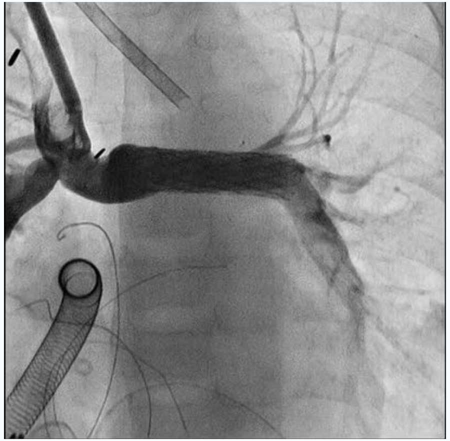
The Journal of Thoracic and Cardiovascular Surgery • February 2016







High rate of PA stenosis

Hybrid Versus Norwood Strategies for Single-Ventricle Palliation

Circulation September 11, 2012

Kenji Baba, MD, PhD; Yasuhiro Kotani, MD, PhD; Devin Chetan, HBA; Rajiv R. Chaturvedi, MD, PhD; Kyong-Jin Lee, MD; Lee N. Benson, MD; Lars Grosse-Wortmann, MD; Glen S. Van Arsdell, MD; Christopher A. Caldarone, MD; Osami Honjo, MD, PhD

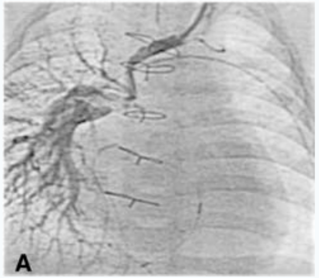


Table 3. Pre-Fontan Evaluation

	Norwood (n=25)	Hybrid (n=14)	*P*
Patient demographics			
Body weight, kg	12.7 (11.9–13.2)	11.6 (11.2–13.2)	0.195
Body surface area, m^2	0.55 (0.52–0.57)	0.53 (0.51–0.55)	0.185
Catheterization/magnetic resonance imaging data			
Arterial saturation, %	88.0 (84.8–90.3)	89.0 (87.0–91.0)	0.236
Ventricular end-diastolic pressure, mm Hg	7.0 (6.0–8.0)	7.5 (7.0–8.0)	0.665
Mean pulmonary artery pressure, mm Hg	10.0 (9.0–12.0)	10.0 (9.0–11.5)	0.605
Common atrial pressure, mm Hg	5.5 (4.3–6.8)	5.0 (4.5–6.0)	0.611
Transpulmonary gradient, mm Hg	5.0 (4.0–6.0)	5.3 (4.3–6.0)	0.981
Pulmonary-to-systemic flow ratio, Qp/Qs	0.81 (0.74–1.02)	1.10 (0.88–1.19)	0.383
Pulmonary vascular resistance, Woods Units	2.36 (1.94–2.70)	2.20 (1.95–2.95)	0.877
Pulmonary arteries			
Right pulmonary artery diameter, mm	9.90 (9.05–11.53)	8.35 (6.80–9.50)	0.003
Indexed right pulmonary artery diameter, mm^2/m^2	18.5 (17.0–20.5)	15.0 (13.5–16.7)	0.008
Left pulmonary artery diameter, mm	7.40 (6.45–8.20)	6.20 (5.43–7.50)	0.071
Indexed left pulmonary artery diameter, mm^2/m^2	13.3 (11.7–15.2)	13.1 (10.7–13.9)	0.273
Nakata Index, mm^2/m^2	216 (187–277)	184 (127–213)	0.015
Right-to-left ratio	1.43 (1.24–1.63)	1.15 (1.10–1.65)	0.622
Lower lobes			
Right lower lobe diameter, mm	7.45 (5.98–8.23)	6.10 (5.68–6.85)	0.093
Left lower lobe diameter, mm	6.20 (5.23–6.60)	5.25 (4.20–6.10)	0.087
Lower lobe index, mm^2/m^2	134 (96–156)	109 (75–131)	0.113

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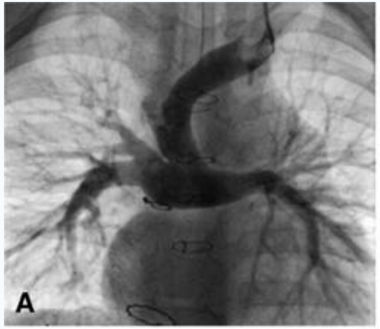

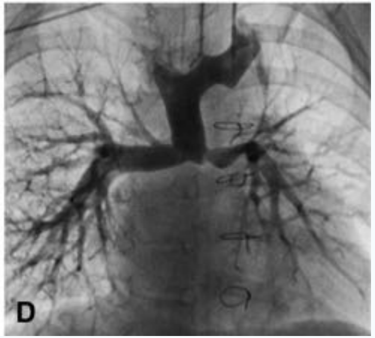

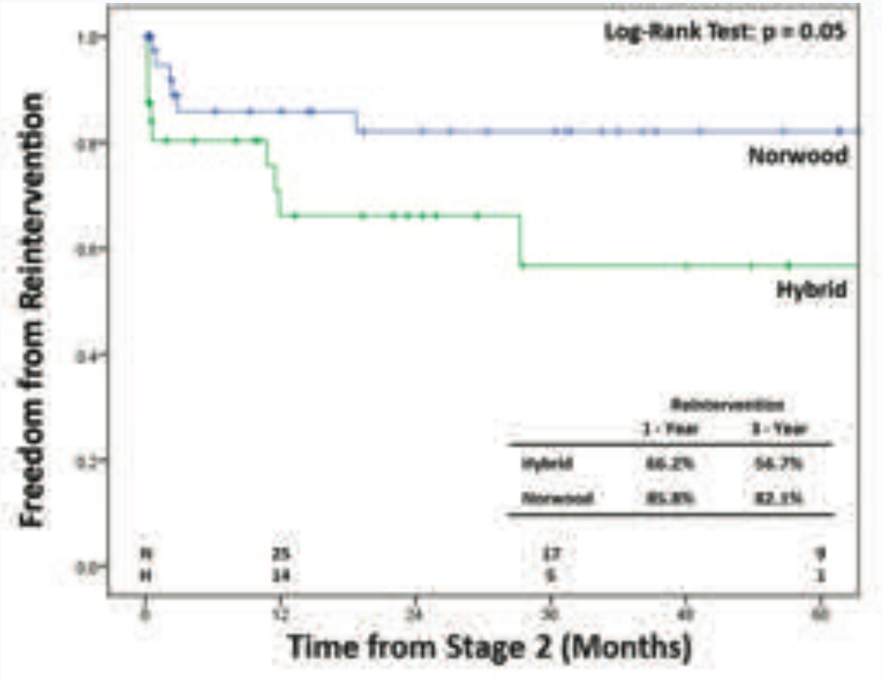

HLHS… hybrid procedure

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HLHS….reason for Hybrid

"In favor of Hybrid Stage 1 as the initial palliation for HLHS" Galantowicz M, Ped Cardiac Surg Ann 2013

1. Promote evolution in HLHS management
2. Changing management concept
3. Resource utilization
4. Bridge to rescue
5. Bridge to HTX
6. Bridge to 2V repair
7. Bridge to Norwood Stage 1
8. Effective for high risk
9. Improve early results
10. Improve late results & Fontan candidancy

HLHS… hybrid procedure

"Is superior to Classical /Modified Stage I ?"

No