Fetal and Hybrid Procedures in Congenital Heart Diseases

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32 Prolonged Right-Ventricle-to-Left-Ventricle Support (Hybrid or Surgical) to Delay Decision-Making in Borderline Left Ventricles

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32.1 Introduction

The management of neonates with hypoplastic left heart syndrome (HLHS) has markedly evolved since the 1950s [1]. Treatment strategies have developed to the extent that now the majority of these children survive until adulthood and beyond. Development of hybrid palliation avoided early circulatory arrest and cardiopulmonary bypass in a neonate, was even possible in low birth weight infants, and combined all the physiological goals requiring only a single surgical sternotomy [2].

Two ends of a spectrum can now clearly be distinguished and managed accordingly. At the one end are neonates with obvious hypoplastic left ventricles (LV) that cannot support the systemic circulation now or later, qualifying only for univentricular repair (UR). At the other end are newborns with a relatively small left heart and a coarctation of the aorta (CoA) that will clearly be able to maintain a biventricular circulation. In between these two extremes are a group at the lower limits of LV functional ability that may or may not sustain a systemic circulation early after birth with a decreasing pulmonary vascular resistance. This ill-defined group, which consists of an LV that is neither rudimentary nor near normal, is generally referred to as "borderline" left ventricles. It includes a heterogeneous group of anatomical and functional LV impairment combined with varying degrees of aortic stenosis

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(AS), coarctation of the aorta (CoA), and other forms of the hypoplastic left heart complex such as aortic arch hypoplasia and mitral valve and left ventricle abnormalities alone or in combination. This nebulous group lies in a gray area where no clear definitions or guidelines exist; the clinician is frequently left undecided as to what constitutes the best management. Even more challenging, the decision has to be made soon after birth in patients who might have the potential of catch-up growth. Delaying the decision at this stage will allow some ventricles to develop and expose small LVs which will never cope with a biventricular circulation.

32.2 Hemodynamic Considerations

32.2.1 Fetal Circulation

Normal physiology During fetal life, the right ventricle is responsible for two-thirds of the combined cardiac output allowing parallel development of the fragile pulmonary vascular bed and the complex structured left ventricle. For the left ventricle, 30 % of the preload comes from left-to-right shunting via the foramen ovale (FO) and about 8 % from the pulmonary veins. The main stimulus for growth appears to be the shear stresses as a result of blood flow over the vascular endothelium [3].

Small left hearts If the FO is restrictive, a decrease down to 25 % of normal LV preload may occur. This will decrease LV growth due to a decrease in flow-related shear stresses. In the case of left ventricular outflow and/or aortic obstruction, pressure rises in the developing left ventricle. This pressure overload leads to myocardial hypertrophy resulting in reduction of left heart volume and compliance and, in extreme cases, endocardial fibroelastosis. Mitral valve abnormalities may also compromise LV flows and impact on LV growth. Fetal somatic development is rarely affected because of compensatory increased right heart output.

Fetal intervention Fetal intervention by means of balloon angioplasty may assist in LV growth in antenatally diagnosed borderline left ventricles [4].

32.2.2 Newborn

As long as the ductus arteriosus remains patent, the majority of infants with a borderline left heart will not immediately become severely symptomatic. Other postnatal circulatory changes will also benefit these hearts.

Normal physiology Preload to the left ventricle will increase immediately after birth due to a significant increase in pulmonary flow to the lungs and thus increase pulmonary venous return; this will lead to the so-called unfolding of the left ventricle. This phenomenon can also be observed in infants where the interventricular

septum was pushed to the left due to increased right heart pressures or flow antenatally. This unfolding on the LV was also been seen by clinicians within hours and days following prostaglandin administration. Adequate or sufficient catch-up development of the left ventricle may, however, take weeks to months to occur.

Small left hearts Afterload reduction by means of aortic balloon angioplasty in the case of aortic stenosis (AS) will decrease pressure overload on the ventricle resulting in reduction of hypertrophy with resultant improvement in LV filling, compliance, and growth. Left ventricular outflow tract obstruction (LVOTO) and the majority of mitral valve abnormalities are not amenable to percutaneous treatment and will be discussed in the next chapter.

Growth potential The summation of all of the physiological and treatment effects may lead to catch-up growth of the small LV over weeks to months analogous to LV growth observed in severely hypertrophied ventricles of infants of diabetic mothers [5] .

32.3 Criteria for the Diagnosis of Borderline Left Ventricle

A closer look at current decision-making algorithms is validated. These were developed in the previous era where limited options were available and any decision needed to be made within days: Norwood for HLHS or biventricular repair for critical CoA to be performed within days after birth. A biventricular circulation is clearly the preferred option if this can be obtained in a safe and predictable manner. Several parameters have to be taken into account including anatomic (e.g., aortic valve, outflow tract, mitral valve and left ventricular dimensions, and volume) and functional (e.g., ejection fraction, diastolic function, etc.) evaluations using echocardiographic and radiological information:

- Rhodes score (1991) [6] includes variables of indexed aortic root and mitral valve dimensions as well as left ventricle dimensions. Mainly applicable to infants with aortic stenosis.
- Colan score (2006) [7] uses body surface area and valve z-score. Mainly applicable to infants with aortic stenosis.
- Congenital Heart Surgeons Society score (2007) [8]: Apart from the above measurements, it also includes variables of the aortic arch. It can be accessed at www.chs.org.

Several publications have shown these scoring systems not to be universally applicable to all infants with borderline left hearts, still leave a large gray zone, and have shown that different scores predict different outcomes of strategies if calculated for the same patient [9–11]. As a result different calculations, including LV volume assessments, have been used, and newer echocardiographic scoring systems are being developed which combine anatomical and flow characteristics

of the left heart or risk of mortality scores [12, 13] . However, to date, no single measurement or combination of measurements is able to unequivocally predict best treatment option for an individual patient. The reason why predictors differ reflects the true nature of a borderline left heart and the shortcomings of current predictors [14].

No clear consensus exists regarding which selection criteria should be used to decide on a UR of BV strategy in infants with a borderline LV. Essentially the question that needs answer is what constitutes the absolute minimum dimensions to allow for BV repair. Even more important some patients clearly show the potential for significant catch-up growth, if sufficient time is allowed. This is important, since going down the wrong pathway can lead to avoidable early and late mortality and morbidity [9, 15–17].

Hybrid strategies (Fig. 32.2) The development of hybrid palliation in recent years and the technical simplicity of this option opened up new avenues of treatment. In the scenario of borderline left hearts, it becomes an attractive option for the following reasons:

- Provide support of the LV by using the RV as temporary assistance
- Can be performed without cardiopulmonary bypass
- Give weeks to months for LV catch-up growth
- Give time and opportunity to treat reversible lesions, e.g., AS, CoA, and left ventricular hypertrophy (LVH)

This could potentially allow for a more weighted decision regarding a univentricular or biventricular strategy and convert more patients safely to a biventricular circulation.

32.4 Right Ventricle-Left Ventricle Assist Strategy

32.4.1 Overview

Our current preferred strategy for infants with borderline left hearts is termed right ventricle-left ventricle assist (RLa) and is demonstrated in Fig. 32.1.

32.4.2 Therapeutic Options

Prostaglandin The immediate decision regarding what to do can initially be delayed by the intravenous administration of prostaglandins (Alprostadil) to maintain ductal patency. This will allow the clinician 1–2 weeks for thorough serial assessment of the left-sided structures and give time for the left ventricle to “unfold” and grow as a result of the augmented preload.

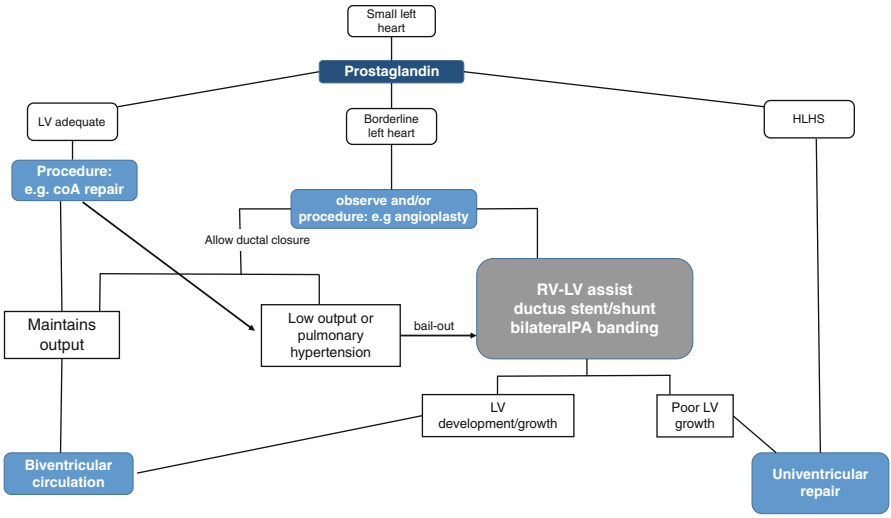

Fig. 32.1 Management strategy

In the latter case, we typically follow with a procedure to unburden the left ventricle. In the case of aortic stenosis, this will consist of aortic valve balloon angioplasty or in the case of aortic arch obstruction coarctectomy or stenting. If the left heart is able to maintain the systemic output, biventricular circulation is aggressively supported.

Right ventricle-left ventricle assist (RLa) This usually consists of bilateral pulmonary artery banding and ductal stenting [18, 19] (Fig. 32.2). Although an elective combined surgical-interventional approach is preferred, a similar result can be accomplished as bail-out procedure by surgery only after an adequate coarctectomy but circulatory failure (see later).

Growth potential If after initial prostaglandin administration after some days the left ventricle remains small but has growth potential (no mitral stenosis, endocardial fibroelastosis) or we are undecided, a hybrid palliation remains our treatment of choice. If the left ventricle after weeks to months shows evidence of development, this option of RV-LV circulatory assist can serve as a bridge to future biventricular circulation [20–22].

No growth potential In the absence of demonstrable or expected left ventricular catch-up growth, the univentricular route with Norwood procedure or hybrid procedure can be chosen.

Surgical RLa In rare cases where the neonatal evaluation and scoring system suggest a good biventricular outcome, but where the postoperative course shows the left heart is not (yet) able to maintain cardiac output, a surgical RLa may be a life saving

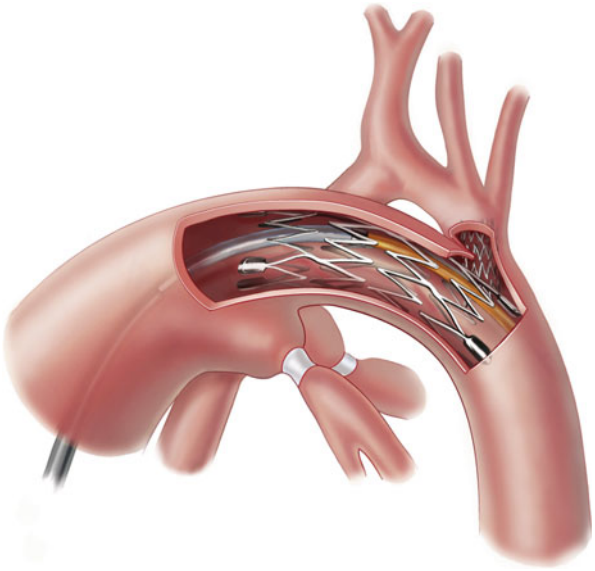

Fig. 32.2 Hybrid right ventricle-left ventricle assist: bilateral pulmonary artery bands and stent in ductus arteriosus also demonstrated. Note: a small stent in aortic isthmus – coarctation; if indicated, the coarctation stent is easier implanted prior to the ductal stent

bail-out procedure. Circulation failure is usually not due to inability of the LV to generate forward flow but as a result of backward failure with the development of retrograde pulmonary hypertension. Pulmonary hypertension may develop early or late, and the resulting interventricular septal shift impinges on the small LV. The surgeon performs bilateral pulmonary artery bandings and a Gore-Tex tube from the pulmonary trunk to thoracic aorta (Fig. 32.3, "reversed" shunt) [23, 24].

32.4.3 Outcomes

32.4.3.1 Results of Strategy

We have performed 18 RV-LV circulatory assist (RLa) procedures for borderline left hearts since 2005. Eleven infants had RLa as the initial procedure as a bridge to biventricular circulation, and two of these proceeded to a univentricular circulation. The majority of these infants (9/11) evolved to a biventricular circulation with a median of 4.4 months (range: 1.5 – 45.8) after the RLa procedure. In the other group ($n=7$) where an initial attempt to relieve LV obstruction was carried out, three ($n=3$) infants of whom one demised evolved to a univentricular circulation. Fifty seven percent ($n=4$) returned to a biventricular circulation with a median of 4 months (range 3.0 – 6.1) after RLa procedure, and one is currently still in follow-up. Characteristics of the groups can be viewed in Table 32.1.

Patients 10 and 11 both initially had a coarctectomy and transection of the ductus arteriosus. After initial stable hemodynamics, they both developed within hours

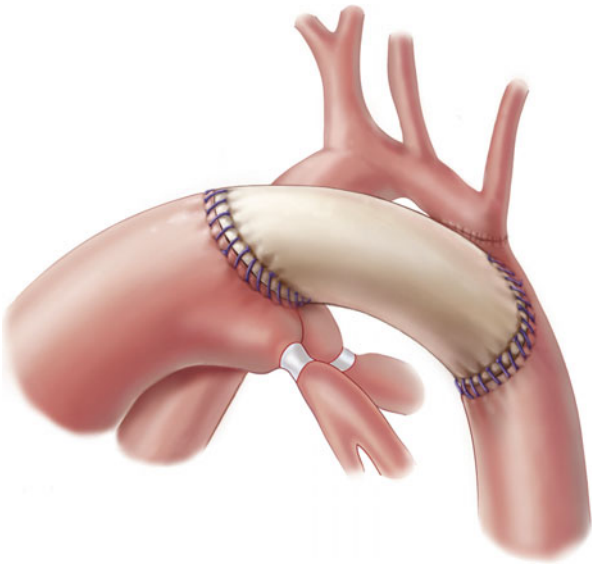

Fig. 32.3 Surgical right ventricular-left ventricular assist procedure "reversed shunt." Bilateral pulmonary artery bands and Gore-Tex pulmonary artery to systemic shunt. Note also that aortic arch was previously reconstructed

suprasystemic retrograde acute pulmonary hypertension with severe RV dilation and compression of the LV. As a result, the small left ventricle could not sustain the systemic output. A "reversed shunt" RLa procedure was performed: a 6 mm Gore-Tex® graft was constructed between the main pulmonary artery and thoracic aorta together with 3.5 mm bilateral pulmonary artery bands (Fig. 32.3). One patient evolved to biventricular repair 4 months later, the other patient to Fontan circulation; both children are doing well.

Patient 13, with a hypoplastic aortic arch complex, proceeded to early RLa. Extensive arch reconstruction (Fig. 32.4b) was carried out 6 months later together with creation of a restrictive interatrial communication to partially decompress the left atrium.

Patient 16 was diagnosed after 6 weeks with Kabuki syndrome, and after consultation with the parents, it was decided that the RLa procedure would be the final palliation. At the last follow-up, the child was 5.5 years old and doing well. This case highlights the fact that RLa procedures can provide long-term palliation with reasonable quality of life in selected cases.

32.4.3.2 Treating Associated Conditions

We have a low tolerance for intervention in these infants following RLa procedures since associated conditions may impede development of the left heart. Aortic valvular stenosis can be safely managed by simple balloon angioplasty. Residual coarctation should be detected early, and balloon angioplasty has been successfully carried out during follow-up.

Table 32.1 Patient characteristics

No	Main lesion	Initial procedure	Age at RLa (day)	Technique	Final procedure	Age (year)
1	AS		9	Percutaneous surgery	Ross	6.5
2	AS		6	Percutaneous surgery	Fontan	
3	AS	BA	28	Percutaneous surgery	PDA closed, bands dilated percutaneously	0.9
4	AS	BA	15	Percutaneous surgery	Arch reconstruction	0.3
5	AS	BA	13	Percutaneous surgery	Fontan	
6	AS	BA	15	Percutaneous surgery	BDG, demise	
7	CoA		8	Percutaneous surgery	Arch reconstruction, subAS resection	1.4
8	CoA		11	Percutaneous surgery	Coarctectomy	0.5
9	CoA		7	Percutaneous surgery	Follow-up	
10	CoA	Coarctectomy	14	Surgical	Arch reconstruction	4
11	CoA	Coarctectomy	14	Surgical	Fontan	
12	CoA-ha		7	Percutaneous surgery	Ventricular septal defect (VSD) closure, debanding	1.6
13	CoA-ha		3	Percutaneous surgery	Arch reconstruction, restrictive fenestration IAS	0.6
14	CoA-ha		3	Percutaneous surgery	Arch reconstruction	0.3
15	CoA-ha	stent CoA	15	Percutaneous surgery	Arch reconstruction	0.6
16	HLHC		8	Percutaneous surgery	Palliative (5.1 year)	
17	HLHC		8	Percutaneous surgery	Kawashima	
18	HLHC		27	Percutaneous surgery	BDG	

AS aortic valve stenosis, *BA* balloon angioplasty, *CoA* coarctation of the aorta, *CoA-ha* coarctation-hypoplastic arch, *HLHC* hypoplastic left heart complex, *BA* balloon valve angioplasty, percutaneous, percutaneous ductus arteriosus stent, and bilateral pulmonary artery banding; surgical, surgical systemic to pulmonary artery shunt and bilateral pulmonary artery banding, *PDA* patent ductus arteriosus, *IAS* interatrial septum, *BDG* bidirectional Glenn

Since ductal stents may show peal or ductal ingrowth over time, these can be balloon dilated or re-stented. Using dilatable bands for the left and right pulmonary arteries is advisable since these can be tailored to adjust flow in relation to somatic growth by balloon angioplasty [25].

Patient 3, with severe AS, had aortic balloon valvuloplasty. The left ventricle was borderline, and an RLa was performed three weeks later. The LV showed continuous improvement in structure and function. The ductus arteriosus stent was later closed by a duct occluder and pulmonary artery bands dilated (Fig 32.4a). This patient was completely rehabilitated by percutaneous means [26].

32.4.3.3 Options for Repair

Fontan Patients proceeding to univentricular circulation typically had extensive aortic arch repair and a Damus-Kaye-Stansel procedure. We often had to treat residual stenosis especially of the left pulmonary artery as a result of banding by means of balloon angioplasty and stenting.

Biventricular circulation This could be accomplished using surgery or by means of percutaneous intervention (see above). Surgery usually consisted of extensive aortic arch reconstruction, clipping the ductus arteriosus, and debanding the pulmonary arteries (Fig 32.4b). Some children required patch plasty at the site of

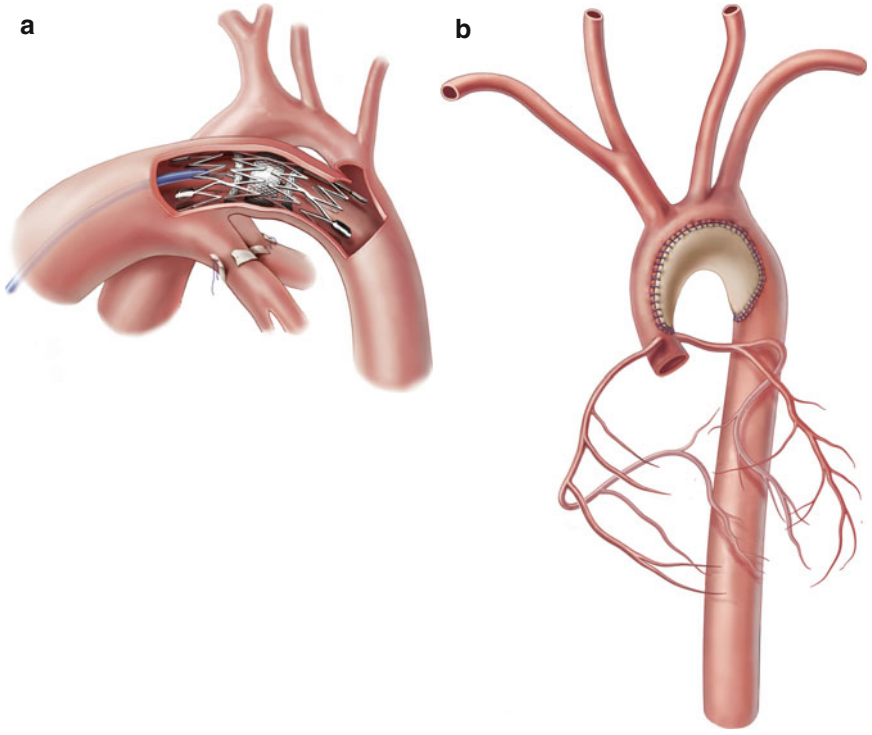

Fig. 32.4 Abolishment of RLa: (**a**) if the aortic arch has sufficiently grown and does not contain a circular stent, the RLa can be occluded percutaneously after balloon release of the PA strips. (**b**) If the aortic arch still shows significant hypoplasia or a circular stent, surgical aortic arch reconstruction is performed

pulmonary artery banding. Several studies have documented good biventricular outcomes in infants with small left hearts [27–31].

32.4.3.4 Impediments to Left Ventricular Growth After RLa Procedures

Mitral Valve

In our experience, certain congenital abnormalities of the mitral valve are an important limiting factor for left ventricular growth in infants with borderline left hearts. Similar to the experience of Del Nido et al., annular hypoplasia, thickened leaflets, commissural fusion, and chordal and papillary muscle abnormalities are common morphological abnormalities seen in these infants and should carefully be assessed and addressed if possible [32].

Endocardial fibroelastosis (EFE) EFE hampers both systolic and diastolic functions of the small left ventricle, and high grades of EFE have been demonstrated to be risk factors for mortality after biventricular repair [33, 34]. Both of our patients with EFE required univentricular circulation.

Atrial septal defect A mildly restrictive shunt through the atrial septal defect (ASD) still allows LV development and further catch-up growth but avoids left atrial hypertension. In selected patients with a small left heart, a mild left-to-right shunt with limited LA hypertension is better tolerated than no shunt but severe retrograde pulmonary hypertension.

32.5 Advantages of Right Ventricle-Left Ventricle Assist Procedure

The main advantages of following this "hybrid approach" for borderline left ventricles are:

- Buy time.
- Allows potential for catch-up growth of LV if possible; most successful in patients where a fetal restrictive foramen ovale was the root cause of the small left heart.
- Can be performed in small and low birthweight infants.
- Allows time for ventricular and somatic growth.
- This strategy can be used as medium term palliation.

Conclusion

The management of infants with borderline left hearts remains a challenge. Current algorithms lacks in ability to discriminate whether biventricular circulation is possible in time if catch-up growth occurs. Right ventricle assist of left ventricle (hybrid) procedures offer the clinician the ability to buy some time to better discriminate which ventricles have adequate growth potential.

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