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Ventriculo-arterial discordance: switching the morphologically left ventricle into the systemic circulation after 3 months of age[1]

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Abstract

Objective: To retrospectively examine a 4 year policy of restoring the morphologically left ventricle to the systemic circuit in patients presenting after 3 months of age with ventriculo-arterial discordance with or without associated atrio-ventricular discordance. This policy was stimulated by the known tendency of the morphologically right ventricle to develop dysfunction sooner or later when left in the systemic circuit. Such a policy dictates a more complex surgical approach and, at this point, it remains controversial whether or not the increased surgical complexity is warranted. **Methods**: From July 1, 1993 to March 31, 1997, a total of 29 patients were entered into a protocol for placement of the morphologically left ventricle into the systemic circuit. Three groups of patients were identified. Group I; congenitally corrected transposition in 14 patients – were treated with either a Senning plus arterial switch operation or Senning plus Rastelli procedure. Group II; failed atrial switch procedure in 12 patients of which nine proceeded to arterial switch operation with Senning or Mustard takedown and atrial reseptation. Group III; D-transposition of the great vessels presenting more than 1 year after birth in three patients who underwent arterial switch operation alone. A deconditioned morphologically left ventricle required reconditioning by means of preparatory pulmonary artery banding in 17 of 29 patients. In the patients requiring pulmonary artery banding, an average of 2.1 pulmonary artery bandings was required to prepare the morphologically left ventricle for a systemic pressure workload. **Results**: In those patients with a deconditioned morphologically left ventricle requiring preparatory pulmonary artery banding, the mean ratio between the left ventricular and right ventricular systolic pressure increased from 0.48 to 0.95. The left ventricular mass increased from 46.6 to 81.8 g/m^2 in five patients subjected to serial MRI measurement. Three patients failed the preparatory pulmonary artery banding and did not proceed to anatomical correction. Two subsequently died at a later time. In the patients proceeding to complete anatomical correction: group I – there were no early or late deaths. Two patients required pacemaker implantation post-operatively. Group II – there were two in-hospital deaths, one early due to intrapulmonary hemorrhage and one late, secondary to postoperative left ventricular failure with a stormy post-operative course requiring successful ECMO placement and weaning. These patients were 18 and 25 years old, respectively. One patient proceeded to cardiac transplantation 3 months after surgery due to ongoing morphologically left and right ventricular dysfunction. Group III – all patients continue to do well. **Conclusions**: Late anatomic correction of ventriculo-arterial discordance with or without atrio-ventricular discordance can be performed at a relatively low risk. Reconditioning of the morphologically left ventricle can be achieved by sequential pulmonary banding but is not without risk. Failure to achieve adequate reconditioning of the morphologically left ventricle by pulmonary artery banding in the older patient probably increases the risk of non-survival and may be offset by timely transplantation. Longer follow-up and an assessment of the functional status of these patients is required to assess whether or not this complex surgical approach is indeed warranted. © 1998 Elsevier Science B.V. All rights reserved

Keywords: Ventriculo-arterial discordance; Arterial switch; Left ventricular retraining

1. Introduction

We retrospectively examined a 4-year policy of restoring the morphologically left ventricle (MLV) to the systemic circulation in patients after 3 months of age with ventriculo-arterial discordance with or without associated atrio-ventricular discordance. This policy was stimulated by the known tendency of the morphologically right ventricle (MRV) to develop dysfunction sooner or later when left in the systemic circuit. Such a policy dictates a more complex

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surgical approach and, at this point in time, it remains controversial whether or not the increased surgical complexity is warranted.

Three groups of patients in whom this policy was applied have been identified: congenitally corrected transposition (ccTGA), failed atrial switch procedures (either Senning or Mustard) as the primary treatment for D-transposition (D-TGA) and patients with D-TGA who presented late (after 1 year of age), who may still be candidates for the arterial switch operation (ASO). One important issue is whether or not the MLV is capable of functioning in the systemic circuit after having been deconditioned in the pulmonary circuit for a period of time. Reconditioning the MLV by means of pulmonary artery banding has provided a method for successful transfer of the MLV into the systemic circuit [10,11].

ccTGA is often associated with other defects such as ventricular septal defects, pulmonary stenosis, tricuspid valve abnormalities and conduction disturbances [14]. Classical repair of ccTGA, by leaving the right ventricle on the systemic side, results in a high incidence of tricuspid valve regurgitation and right ventricular failure developing in the years after repair [1–4]. Even in the absence of other structural defects, there remains a significant incidence of right ventricular dysfunction in older patients [5,6].

Likewise, after the Mustard or Senning operation for D-TGA, important failure of the systemic MRV is seen in at least 10% of patients late after surgery [7,8] and furthermore, many ‘asymptomatic’ patients exhibit dysfunction of the systemic MRV during exercise [9].

This unreliability of the MRV in the systemic circuit has led to our policy of using the MLV as the systemic pump whenever possible. This includes primary or secondary repair of ccTGA, older patients with D-TGA presenting late, as well as patients with right ventricular failure following an atrial switch operation. A review of this policy is presented.

2. Materials and methods

2.1. Patient population

From July 1, 1993 to March 31, 1997, a review of 29 patients (18 male and 11 female) was undertaken. Three groups of patients were identified; group I: ccTGA, group II: failed atrial switch procedures for the primary treatment of D-TGA, group III: D-TGA presenting after 1 year of age.

Table 1

Demographics of group I patients: congenitally corrected transposition ($n = 14$)

SLL	13
IDD	1
Ventricular septal defect	12
Pulmonary stenosis	8
Ebsteinoid malformation of tricuspid valve	5
Coarctation of aorta	1
Tricuspid regurgitation	6
Interrupted IVC	1

Table 2

Demographics of group II patients: failed atrial switch ($n = 12$)

Mustard procedure	7
Senning procedure	5
Prior ventricular septal defect closure	1
Subpulmonary resection	1
Surgery for baffle obstruction	2

2.1.1. Group I: ccTGA

Fourteen patients with a mean age of 2.6 years (range 3 months to 8.5 years). The associated defects are shown in Table 1. Previous surgery included systemic to pulmonary artery shunting in seven patients of which two proceeded to bidirectional cavopulmonary anastomoses. One patient had repair of an aortic coarctation. Three patients with large VSDs had prior PAB and four patients with unobstructed pulmonary blood flow, with or without previous surgery, had deconditioned MLVs. Presentation was for a failing MRV in the systemic circuit in three patients and presentation for complete repair alone in 11 patients.

2.1.2. Group II: failed atrial switch operation (Mustard or Senning)

12 patients with a mean age of 10.1 years (range 16 months to 25.4 years). One patient had systemic MLV pressure from left ventricular outflow tract obstruction (LVOTO).The demographics of this group are shown in Table 2. All patients in this group presented for surgery with failure of the MRV and significant functional compromise. One patient developed hepatic cirrhosis after transfusion related hepatitis and, although she was a transplant candidate, was refused transplantation elsewhere on the grounds of active hepatitis.

2.1.3. Group III: D-TGA

Comprised three patients with a mean age of 5.3 years (range 16 to months–8.5 years). Two patients had a previous atrial balloon septostomy and one patient with a large non-restrictive atrial septal defect had no prior interventions.

2.2. Surgical policies and procedures

Patients were subjected either to anatomical correction and placement of the MLV into the systemic circuit or, preparatory reconditioning of the MLV by means of pulmonary artery banding for work within the systemic circuit. The suitability of these patients for final repair involved the exclusion of factors which might preclude a good surgical outcome. These would be irreversible, structural dysfunction of the MLV due to previous insult, more than mild mitral regurgitation, pulmonary valve abnormalities precluding use of the pulmonary valve in the ASO, subpulmonary stenosis not amenable to surgical relief and a coronary artery pattern precluding ASO. We did not encounter any of these contraindications in any of our patients.

The adequacy of conditioning of the MLV for transfer into the systemic circuit was based on the echocardiographic, cardiac catheterization and more recently, in the last five patients, MRI assessment of the MLV. As a principle, we have accepted an MLV pressure to MRV pressure ratio of greater than 0.75 as being indicative of a conditioned ventricle [10]. More recently, we have been able to identify the presence of a left ventricular mass of more than 80 g/m^2 as being indicative of adequate MLV conditioning [16].This was not used in the decision-making for these patients.

If the MLV was pre-conditioned by the presence of a ventricular septal defect and/or pulmonary stenosis, or pulmonary hypertension, anatomical transfer of the MLV into the systemic circuit was immediately undertaken. This was done in 11 patients.

2.3. Pulmonary artery banding

Preparation of the MLV for a systemic workload was undertaken by increasing the afterload progressively, by means of a pulmonary artery band [10]. Depending upon the clinical response, as well as the echocardiographic, cardiac catheterization and MRI data, subsequent tightening was undertaken if appropriate. In D-TGA patients with no prior atrial switch, a modified Blalock–Taussig shunt was added to ensure adequate pulmonary blood flow. At initial placement of the pulmonary artery band, tightening was guided intra-operatively by measurements of systemic arterial pressure, MLV systolic and end diastolic pressures and systemic venous pressure. Intra-operative transesophageal echocardiography is indispensable for following the changes in ventricular size and contractile function at the time of band placement. The degree of shift of the interventricular septum towards the MRV, as well as the decrease in tricuspid valve regurgitation that accompanies the increase in pulmonary artery impedance, are important clinical markers. Excessive pulmonary artery band tightening leads to reduced MLV function and edema of the myocardium, as documented by MRI. Any early or late MLV dysfunction should lead to consideration of PAB loosening. The post-operative protocol therefore includes a minimum of 24 h of paralysis with ventilatory support and dobutamine support for at least five days. If the clinical and echocardiographic parameters deteriorate, the pulmonary artery band is immediately loosened. Dobutamine is weaned based upon echocardiographic documentation of sustained good MLV function. Five patients in the latter part of this study were also followed by an MRI which allowed non-invasive measurement of changes in ejection fraction and ventricular mass. Additionally, T2 weighted MRI scan (16) has enabled us to document the presence of acute myocardial edema in response to what we believe is excessive increase in afterload on the MLV. This myocardial edema does disappear as myocardial function improves. As described by the senior author, it is often necessary to tighten the pulmonary artery band one week after placement. This is due to the creation of folds within the main pulmonary artery following the first pulmonary artery banding. Over a period of time these folds flatten out, and the initial increase in afterload obtained at the first pulmonary artery banding is significantly lowered. The need for multiple band adjustments, particularly with low MLV starting pressures, is evidenced by a mean of 2.1 bandings per patient. Following the initial pulmonary artery banding, these patients are closely monitored both clinically, and by serial echocardiographic measurements, in order to determine whether or not the MLV is capable of sustaining the acute hemodynamic change. Likewise, echocardiographic documentation helps with the decision for intermediate term band retightening or late band retightening. Prior to undertaking any further band retightening, patients are subjected to cardiac catheterization for accurate measurement of the MLV/RV pressure ratio.

2.4. Final repair

The ‘double switch’ by our definition includes a Senning atrial switch procedure combined with either an ASO [12,13] or a Rastelli procedure, and does not include conversion of an atrial switch to an arterial switch. Reversal of an atrial switch is mostly done by utilizing the atrial tissue originally used to create the Senning baffle as previously described [15] or by utilizing the original Mustard baffle if this remains pliable. Only one patient required atrial septation with the placement of a Gore-Tex patch. In simple D-TGA, an ASO with shunt takedown and pulmonary artery reconstruction is performed. Crystalloid cardioplegia is administered in a prograde fashion unless ventricular reserve is marginal and cardioplegia delivery questionable and, under these circumstances, use would be made of blood cardioplegia with Aspartate and Glutamate enrichment and retrograde and prograde administration of cardioplegia. Detailed techniques for the surgical conversion of these patients have previously been described by the senior author [10,13,15,16].

2.5. Statistical analysis

A paired *t*-test was used to compare ventricular measurements. Fisher’s exact test was used to compare survival between two age groups. A *P*-value of 0.05 was considered to be significant.

3. Results

The clinical outcomes of 29 patients at presentation to the Cleveland Clinic Foundation are shown in Fig. 1.

3.1. Preparatory pulmonary artery banding to recondition the MLV

Twelve patients – 10 ccTGA, one TGA and one TGA with prior atrial switch and LVOTO had nearly systemic pressure in the MLV. These patients proceeded immediately to complete anatomical correction. In this group of patients, the mean ratio of left to right ventricular systolic pressure was 0.98 (range 0.80–1.06). Of the remaining 17 patients with a deconditioned MLV, four were ccTGA, 11 were failed atrial switches and two were unoperated D-TGA. These patients had a mean ratio of left to right ventricular pressure of 0.48 (range 0.30–0.76) and were subjected to pulmonary artery banding and MLV reconditioning.

17 out of 29 patients who were subjected to pulmonary artery banding procedures underwent 1–5 pulmonary artery banding procedures (mean 2.1 procedures per patient). This protocol, as described, consisted of pulmonary artery banding followed by adjustment after one week (tightening in eight and loosening in one) as appropriate, and, if necessary, further tightening after 6 months (six patients) with an average of 7.1 months between tightening. The mean time elapsing from first banding to final repair was 11.2 months with a range of 24 days to 19 months. The MLV to MRV pressure ratio increased from 0.048 to 0.95 in the banded patients. In five patients, comparative measurements by MRI were undertaken, the left ventricular mass increased from 46.6 g/m^2 (range 32–59 g/m^2) to 81.8 g/m^2 (range 54–100 g/m^2) ($P = 0.04$), whereas the ejection fraction showed a decrease from 66.4% (range 53–76%) to 50.8% (range 28–74%), P-value 0.22, which at this stage was not noted to be statistically significant.

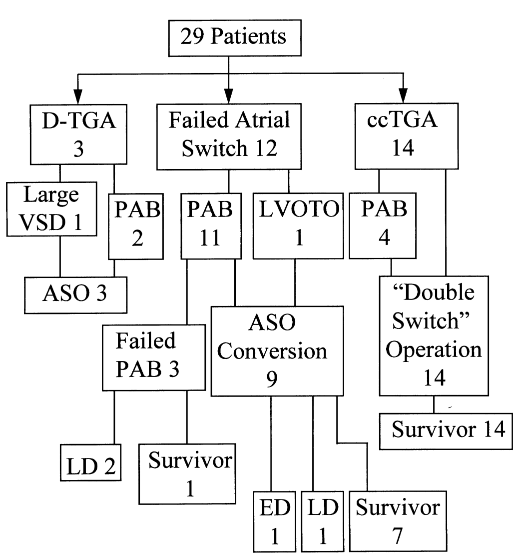

Fig. 1. Clinical outcomes of 29 patients at presentation to the Cleveland Clinic Foundation; PAP, pulmonary artery banding; LD, late death; ED, early death; LVOTO, left ventricular outflow tract obstruction. Double switch operation is ASO with Senning or Senning with Rastelli.

Three patients did not tolerate the pulmonary artery banding procedure and two were subsequently debanded. One patient with previously diagnosed baffle obstruction, who was subjected to pulmonary artery band placement following balloon dilatation of the obstruction, was debanded within 24 h of band placement. This patient never fully recovered from acute biventricular failure and subsequently developed multiple organ failure with end organ compromise and death. The other debanded patient, also with baffle obstruction died 3 months later with progressive MRV dysfunction. The third patient developed late MLV dysfunction and remains quite well with less MRV dysfunction and less tricuspid regurgitation with a moderately tight band still in place.

3.2. Final repair

Anatomical correction with placement of the MLV into the systemic circuit was performed in a total of 26 patients. Group I ccTGA patients were subjected to a 'double switch procedure' consisting of a Senning procedure and an ASO in seven patients or a Senning procedure with a Rastelli operation [7]. Associated surgery included VSD closure (five patients), takedown of systemic to pulmonary or cavopulmonary shunts (seven patients), left ventricular outflow tract resection (one patient) and tricuspid valve repair (one patient). There was no early mortality in this group. Two patients with peri-operative atrioventricular conduction disturbances both required pacemaker implantation after surgery. In three cases, 'mini' peritoneal dialysis from 1-5 days was used. Fifty percent of these patients developed pleural effusions requiring drainage contributing to a mean postoperative stay of 10.0 days (range 5–24 days). Group II failed atrial switch patients; nine were subjected to final repair. Associated surgery included left ventricular outflow tract resection (two patients) and pulmonary valve repair (two patients). In this group, there was one early mortality in a 25-year-old female with hepatic cirrhosis and intrapulmonary vascular malformations who suffered a fatal intrapulmonary hemorrhage. An 18-year-old male who had a left pulmonary artery occlusion due to early erroneous ligation of the left pulmonary at another institution developed severe left ventricular failure after intra-operative problems complicating transfer of the right coronary artery. He was successfully supported and weaned from ECMO. He remained, however, in renal and respiratory failure and, after three months, he suffered a fatal ventricular fibrillation episode in another institution. In this group of patients with failed atrial switch, no deaths occurred in patients younger than 15 years and of the two patients older than 15 years (18 and 25 years), submitted to final anatomic repair, no survivors were obtained ($P = 0.003$). Morbidity included exacerbation of pre-existing sick sinus syndrome requiring pacemaker implantation in one patient, adjustment of atrial septation in one patient and closure of an undiagnosed ventricular septal defect in one patient. Mean hospital stay was 11.5 days (range 6–45 days). Group III patients with D-TGA but no prior switch operation had an uncomplicated ASO and closure of atrial septal defect. No mortality or major complications occurred in this group. With a mean follow-up of 14 months (range 1–38 months), no late mortality has occurred.

4. Discussion

Switching the MLV to the systemic circuit is only feasible if the MLV is capable of generating systemic pressures, i.e. it is conditioned. This ability is preserved early in life as demonstrated by the good results of the neonatal arterial switch operation and is easily recovered, even in early infancy as is evidenced by the rapid two-stage arterial switch operation. In the deconditioned MLV, retraining of this ventricle is possible throughout childhood. Even though only one death in this series was clearly related to left ventricular failure, the fate of the two young adults in this study might suggest that, for older age patients, reconditioning is more difficult. A ratio of left to right ventricular systolic pressure of 0.75 or greater, in the presence of well preserved MLV function, is considered to be adequate for anatomical correction. The ideal left ventricular mass is, as yet, not defined. In the present study, we documented a significant increase in both LV to RV pressure ratios and left ventricular muscle mass in a small number of these patients ($n = 5$). The left ventricular ejection fraction showed a decrease during reconditioning, but the significance of this is not yet fully understood. The altered geometry of the ventricle with increased afterload might make a direct comparison invalid, but it also might reflect true dysfunction due to inadequacy of the ventricle. A study [16] suggests that a low initial left to right ventricular pressure ratio and too large a muscle mass after training is associated with left ventricular dysfunction. It is probable that not all ventricles are retrainable. As documented, three patients, of whom two ultimately died, failed attempted reconditioning.

In the unprepared ventricles, a mean of 2.1 banding procedures was required. A period of 11.2 months, on average, was necessary to reach the desired pressure ratio. This places a considerable burden on the patient but it must be compared with transplantation which is the only long-term alternative for the patient with irreversible systemic ventricular failure. Ideally, a MLV should function for a lifetime but even if the retrained ventricle should later fail or the retraining prove impossible, a bridge to transplantation has been achieved. In patients who fail pulmonary artery banding, consideration should be given to earlier transplantation.

The good early results of the ‘double switch procedure’ in our series compare favorably with the well documented operative mortality, and high rate of early re-intervention for tricuspid insufficiency and persisting left ventricular outflow tract obstruction after ‘classical’ repair of ccTGA. [2–4].With non-resectable left ventricular outflow tract obstruction, a Rastelli type of operation is performed. This has the advantage of giving access to the interventricular septum from the MRV, hence minimizing the risk of damage to the conduction system. Only two patients, both with previous conduction problems, required pacemaker implantation. The Senning procedure carries a risk of its own for venous obstruction or atrial arrhythmias late after surgery. We have not observed this, but our follow-up is short. The high rate of ‘mini’ peritoneal dialysis in our series is a reflection of our low threshold of institution of this therapy. The reason for the high rate of pleural effusion as evidenced in the Group I ccTGA, is not entirely clear, however, it did prolong hospital stay. Group II (failed atrial switch) were the patients at highest risk in this policy. This group of patients is more uniformly compromised pre-operatively with a heart transplant often being the only other alternative.

5. Conclusion

Late anatomic correction of ventriculo-arterial discordance, with or without atrio-ventricular discordance can be performed at a relatively low risk. Reconditioning of the MLV can be achieved by sequential pulmonary banding but is not without risk. Failure to achieve adequate reconditioning of the MLV by pulmonary artery banding in the older patient probably increases the risk of non-survival and may be offset by timely cardiac transplantation. A longer follow-up and assessment of the functional status of these patients is required to assess whether or not this complex surgical approach is appropriate.

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Appendix A. Conference discussion

Dr J. Tingleff (*Copenhagen, Denmark*): I will start on retraining of left ventricular function. This patient group has shown that on echocardiography one point you should look upon is morphology of the left ventricle. If you look on echo, a normal right ventricle is banana-shaped. In your study have you looked at how the morphology and how the outlook on echo was in these patients? And, in particular, the two failures you have, did you see that the right ventricle was not going over to more of a banana shape and the left ventricle was not a circular one as you normally see on echoes?

Dr Helvind: Well, during the banding we aim for a shift of the septum to a midline position. Since you have the same pressure in the two ventricles when you have finished the training, you will expect the septum to be in the middle, which we also saw on the MRI. So the banana shape of the, by that time, pulmonic right ventricle is not expected to be seen until after the switch. Of the two patients who died, one had an ischemic event during surgery and a global dysfunction of the left ventricle, the other had actually a good function of the left ventricle after the switch operation, and I don't think the shape of the right ventricle had any bearing on the outcome.

In another study we have compared MRI with echo, and even though we use echo a lot to follow the day-to-day development, especially in the early part of the training period, it is difficult to determine wall thickness or left ventricular mass accurately with echo. It is much more accurate with MRI

Dr F. Lacour-Gayet (*Paris, France*): In Paris, at Marie Lannelongue, we share with you the same difficulties in trying to retrain left ventricles beyond a certain age. My first question is: is there in your experience, an upper age limit for retraining left ventricle? My second question is to ask you more details on the method of retraining: what are your landmarks for the first PA banding? How do you handle the associated tricuspid regurgitation and the sort of double ventricular failure that is generated? In our hands, these procedures have been quite difficult.

Dr Helvind: When it comes to the age, there are only two patients here who are in adult age and the other patients are younger than 12 years, which means that there is a gap there. You could speculate that as long as the child is growing, there is a better substrate for developing the left ventricle. But really we cannot tell from this. We are, as I said, cautious when we advise about double switch versus transplantation in adult patients.

When it comes to the banding, first of all we look at the maximal pressure that the ventricle can generate. We do that both in the cath lab prior to the switch and in the OR by occluding the pulmonary artery. If you can generate a high pressure in the left ventricle, you can go a bit further in your tightening. Otherwise, we tighten the band to the point where you see the shift in the septum, so it's in mid-position, and then we wait for a while, and if all of the hemodynamics are well, we'll tighten it a little bit further and wait for a while, and often we will have to go a step backwards because of failure of the systemic circulation. Post-operatively, as I said, we support the patients both with paralysis and ventilation first and later with dobutamine for quite a long time. The tricuspid regurgitation is not as big a problem as you would think because the shift of the septum gives a better geometry of the right ventricle and the tricuspid regurgitation actually is reduced during the banding period. It was not so clearly shown in this study. That is why I have not presented the figures. But clinically it is our impression that it gets less and the patients actually tolerate this long period of banding quite well because their tricuspid regurgitation decreases, and presumably they get a better function of the right ventricle because of the support of the septum. The reduction in tricuspid regurgitation has been described by Frank Hanley's group. Of our patients, only one needed tricuspid valve plasty at the time of final repair in the group of congenitally corrected transposition.