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Univentricular heart with low pulmonary vascular resistances: septation vs atriopulmonary anastomosys

The choice of the surgical procedure for the univentricular heart is a real challenge. Three different techniques are most used: 1) atriopulmonary anastomosis with or without a valvar prothesis between the atrium and pulmonary artery; 2) intraventricular seplation; and 3) partial correction of the pulmonary stenosis.

Since 1974, twelve patients were divided in three groups and operated on with cardio-pulmonary bypass.

Group I The three cases operated in 1974 by partial correction of the pulmonary stenosis, since septation was not possible. Two patients are asymptomatic and the other has mild cyanosis and moderate functional incapacity.

Group 11 - The three patients were operated by ventricular septation, where one died in the early post-operative period, another died suddenly three years later, while the sole survivor is very well on medication.

Group III - Six patients were submitted to atriopulmonary anastomosis, one died in the immediate post-operative period. In the follow-up, ergometric testing, pulmonary scin-tigraphy and cardiologic evaluation were performed. All results were normal, including functional capacity.

In conclusion, the atriopulmonary anastomosis is an adequate, low-risk procedure. Septation is a better solution but only when the A-V leaflets are normal, whereas otherwise the procedure incurs in greater risk.

Septation was been advocated by Malm [1] in 1973 as the most logical treatment for single ventricle, and used for those patients with appropiate anatomical features. Nevertheless, the technique is still associated with a high mortality and morbidity [2].

In 1974 we began our experience in septation of single ventricle 34. In 1976 Yacoub 5 reported for the first time atriopulmonary anastomosis (inserting two caval homovalves) for surgical treatment of univentricular hearts following Fontan's principle for tricuspid atresia.

With a different hemodynamic conception (without using caval valves) and supported by our previous experience [7-12] started in 1971 with tricuspid atresia (fig. 1), we began seven years later to perform atriopulmonary anastomosis in patients with single ventricle and low pulmonary blood flow.

The aim of this study is to review the long term follow up data obtained from all the pw tients with this condition sujected to open heart surgery since 1974 and to discuss the appropiate surgical management of univentricular hearts.

Material

Since 1974, 12 patients with univentricular hearts and low pulmonary vascular resistances (excluding classic tricuspid atresia) have been subjected to open heart surgery.

Group 1: Palliation: (table I). In three cases operated upon in 1974 the condition was misdiagnosed, and recognized at the time of the right cardiotomy. The septation was considered to be hazardous because of unfourable anatomical conditions. Therefore palliative relief of the subpulmonic stenosis in the first and third patients and a partial debanding in the second one, were accom'plished wihout mortality. After six years follow up, all of them are alive. Two of them are asymptomatic and lead-ing a normal life, and one has a grade 2 incapacity with moderate cyanosis.

From Hospital de Niños and Clínica Bazterrica, Buenos Aires, Argentina. Read by Dr. A. Schlichter at the 1st World Congress of Pediatric Cardiology. June 1980, London.

All 3 patients are in sinus rhythm. If operated nowadays these patients would be candidates for an atriopulmonary anastomosis.

Group 2: Septation (table II). Three other patients were septated, because the anatomic conditions were considered to be favourable.

The first one died 48 hours after surgery because of low cardiac output, due to balooning of a large teflon patch into the right ventricle.

A second patient presented AV dissociation during the first week postoperatively. She was discharged from hospital in sinus rithm, and remained asimptomatic for three years, until her sudden death.

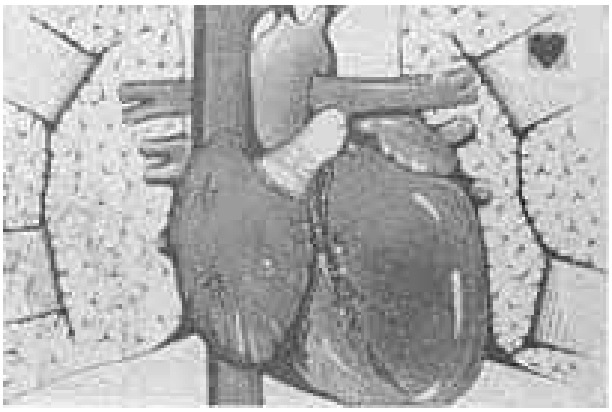


Fig. 1 - Atriopulmonary anastomosis performed in 1971 for tripcuspid atresia without using caval valves.

Table I - Open Heart Palliation.
Unfavourable intracardiac anatomic conditions

Patient	Years	Operation	Results
KH	4	Subpulmonary resection RV small if septated	Good functional capacity Acyanotic Moderate cardiomegaly PAP 30 mmHg
MU	7	Partial debanding Unfav. Pap. muscle Distribution	Asymptomatic Acyanotic Excercise test 91,7%
VJ	26	Partial subpulmonary Resection Unfav. outlet chamber	Gd. 11 incapacity Moderate cyanosis

All patients were sujected to open surgery in 1974 missdiagnosed

Table II – Septation.
Double inlet hearts, no outlet chamber.

Age	Operation	Results
4	Septation Subpulmonary resect.	Inmed. postop. death
9	Septation Subpulmonary resect.	Transient A-V block Late sudden death (3 years after surgery)
15	Septation Homograft reconst.	Excelent, Asimptomat. Normal Funct. Capacity Follow up 6 years

Table III - Atriopulmonary anastomosis, surgical procedure.

A-P continuity	1	22 mm Dacron Tube
	5	Direct anastomosis (1 anterior Pericardial patch)
A.S.D. Closure	4	Direct closure (running sut)
	2	Pericardial patch
Right A-V valve	2	Pericardial patch
	1	Direct closure

Hipoplastic right A-V valve diagnosed by two dimen. Echo. Closed at. Reop. 6 days later.

A third patient with a double inlet univentricular heart with anterior aorta, was septated, and a fresh homograph right outflow tract reconstruction accomplished.

After six years follow up, her clinical condition is excellent, she is asymptomatic, and with normal functional capacity.

Group 3: Atriopulmonary anastomosis (table III) After 1978, six atriopulmonary anastomosis were performed, with one early post-operative death.

The anatomic features (fig. 2) of these patients were: dextrocardia in four (three of them had absent AV connection), the other one had a hipoplastic right AV valve. Out of the six, five had a left anterior aorta the remainining one having a normal arterial relationship. (Holmes Heart).

Their ages ranged from two to eleven years, at the time of surgery.

Cardiopulmonary bypass was established, inserting caval canulae directly into the veins, preserving the right atrial wall and the sinus node. Only one atriotomy was performed carefully choosing the site of the incision (Fig. 3A) always considering the relation with the pulmonary artery and the future anastomosis. Care must be taken with the sinus node artery. At this point it is easier to see it using cardioplegic solution. The ASD and the right AV valve were closed through the same incision.

The ASD was closed with a pericardial patch in two, and with a double running suture in the others. The AV valve was obliterated (fig. 3D) with a pericardial patch sutured to the base of the valve leaflets in the first two patients, and with a double running suture in the others.

Atriopulmonary anastomosis was achieved with a direct anastomosis in the last five patients, and by means of a 22 mm. non valved dacron tube in the only one with normal arterial relationship.

We have never inserted caval valves in any atriopulmonary anastomosis, since our first

case in tricuspid atresia in 1971.

Neither did we use valved conduits to connect the right atrium with the pulmonary artery in this series.

The only death was in the fifth patient on the sixt postoperative day, after a long standing low cardiac output syndrome, with renal failure.

The sixth patient in this series was noted to be cyanosed in the immediate postoperative course. A previously not diagnosed hipoplastic right AV valve was disclosed by a contrast two dimensional echo. The patient was reoperated upon, and a 6 mm hipoplasic valve closed with a direct suture.

Results

The follow up period (table IV) of the 5 survivors ranges from 11 to 27 months (De-cember 1980).

The patient with the Holmes Heart (case 1) in whom a 22 mm dacron tube was inserted, has had tachicardia with AV dissociation and atrial flutter. These arrhytmias were confirmed by 24 hours Holter monitoring. During the atrial flutter, marked liver enlargement and dyspnea were noted. This was the only patient to have pleural effusion and mild ascites during the early postoperative course. He is now in sinus rhythm and asymptomatic. The four patients in whom a direct anastomosis between he right atrium and the pulmonary artery was performed, (our actual technique of choice) have been completely asymptomatic since their discharge from hospital.

In the late follow up, the liver was palpable between 0.5 and 2 cm, from the right costal margin. It is interesting to note that the liver had become progressively smaller with the passing of time, although diuretic therapy had been discontinued in all of them. The urinanalisis was normal,

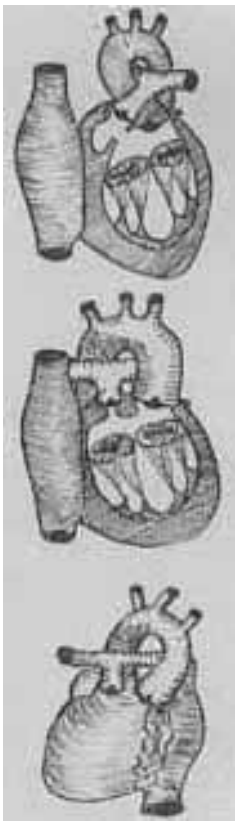


Fig. 2 - Anatomical fetures of patients submited to atriopulmonary anastomosis: A: case 1 (Holmes Heart) Double inlet ventricle with normorelated great arteries. B: case 2, Double inlet ventricular heart with left anterior aorta. C: cases 3,4,5,6, Left anterior aorta in dextrocardia with absent right AV connection in 3,4,5 and a hipoplastic AV connection in case 6.

Table IV - Atriopulmonary anastomosis – Follow up.

Follow up	Arrhythmias	Symptoms	Liver	Urine
22 months	Atrial flutter Tachicard. with AV disoc. Actually sinus R.	Pleural efus. Dyspnea Asymptomatic	 4-5 cm 2,0 cm	 Normal (no albumin)
21 months	Junctional tachicard. Actually sinus P.	None	0,5 cm	Normal (no albumin)
12 months	None	None	1,0 cm	Normal (no albumin)
7 months	None	None	1,0 cm	Normal (no albumin)
5 months	None	None	2,0 cm	Normal (no albumin)

and no albumin could be detected in any of the five patients.

The lung scanning (fig. 4) performed in upright position, showed a preferencial perfusion of the medular portion of the lungs whereas in horizontal Dosition in this same patient it showed a slightly increased perfusion of the bases of the lungs.

The exercise test performed under Bruce protocol (table V) carried out 3 to 16 months after surgery, showed a slightly reduced capacity to exercise in four cases, and normality in one. To determine the normal, 100 non cardiac patient of different ages were studied under the same protocol (table VI and VII).

Four of he five survivors were recatheterized (table VIII): mean right atrial pressure ranged between 10 to 12 mmHg, in the 3 patients with a large direct atriopulmonary anastomosis, and 20 mmHg, in the only one with and anterior dacron tube. The left atrial pressure was between 6 and 8 mmHg.

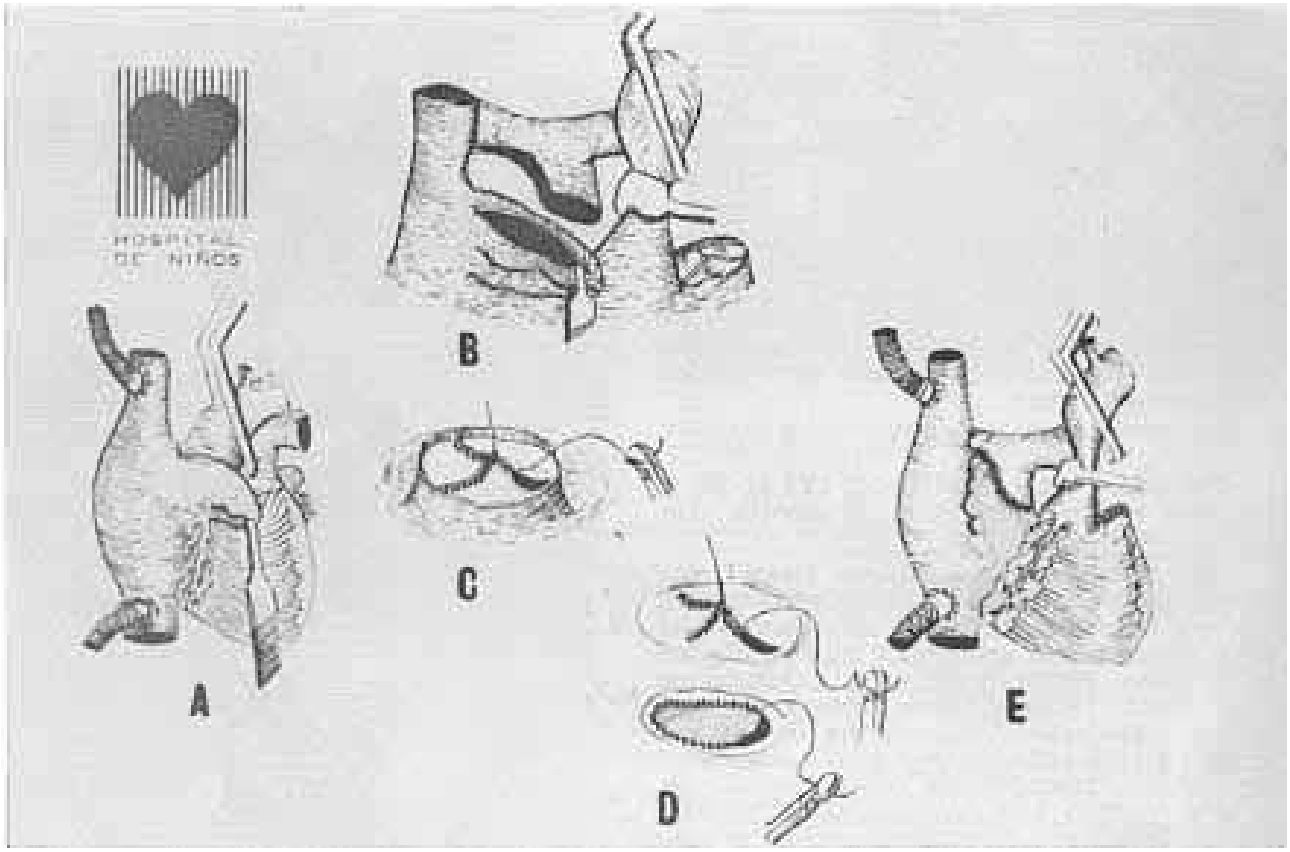
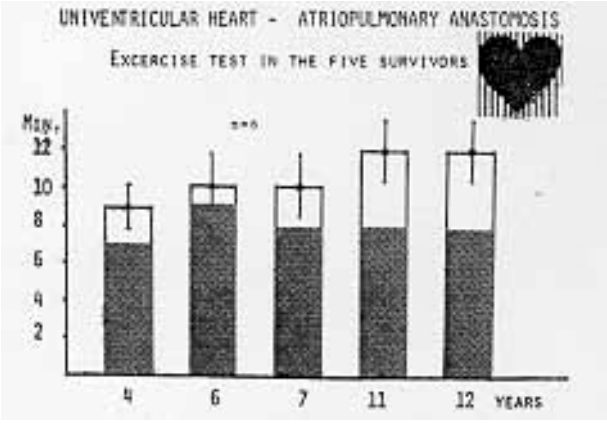


Fig. 3 - Atriopulmonary anastomosis, surgical technique. A: Univentricular heart with normally related great arteries. Both caval veins were canulated outside the right atrium. The right atrial appendage was exposed in order to show the site of the atrial incision. B: The main pulmonary artery was transected just above the leaflets, and mobilized behind the aorta. The opening of the pulmonary artery was extended from the main pulmonary artery into the right branch. An incision in the right atrium was performed in closest relationship with the opening of the pulmonary artery. C: The pulmonary valve leaflets were closed with a double running suture. D: The tricuspid valve leaflets can be closed by a double running suture or with a pericardial patch suture directly to the leaflets 2 mm. away from the annulus. E: The atriopulmonary anstomosis was performed. The proximal stump of the pulmonary artery is already closed.



Patient 2 is absoluetly normal, and the other four have slightly reduced capacity to exercise, but are leading a O Normal standard deviation.

The left ventricular and end diastolic pressure was normal in all cases. The pressure curves were morphologicaly similar in the right atrium, pulmonary artery, and both vena cava.

Systemic saturation was normal. Angiography performed in the right atrium showed good filling of the main pulmonary artery and branches (fig. 5, 6, 7).

Table VI - Normal excercise test.
Normal exercise test in children
Duration in minutes*

	Age	Surface	Results
Grupo I	3-5y	0,72m	9 ± 1,15
Grupo II	6-8y	0,91m	10,1 ± 1,77
Grupo III	9-12y	1,22m	12,08 ± 1,59
Grupo IV	13-15y	1,30m	13,24 ± 2,48

*n – 100 non cardiac patients.
100 non cardiac patients were studdied under Bruce Protocol, in ord to establish the normal duration of the test according to age.

Discussion

Comparison between the three procedures shows bipartition of the ventricular chamber as the most corrective operation. A pulmonary circulation with pulsatile flow, makes this procedure attractive. But favourable anatomical conditions are essential for a successful septation. Two normal AV valves without straddling, and adequate distribution of the papillary muscles for bipartition are necessary. Moreover, this technique still carries a high surgical mortality (40-50%) [2-13].

Table VII - Excercise test in normal, group comparison.
Group comparison
Levels of significance

Group I Vs Group II p< 0,05
Group II Vs Group III p < 0,001
Group III Vs Group IV p > 0,05
Group I Vs Group III p< 0,001

*Buenos Aires Childrens Hospital
Four ages group were selected, and results compared in order to know their level of significance.

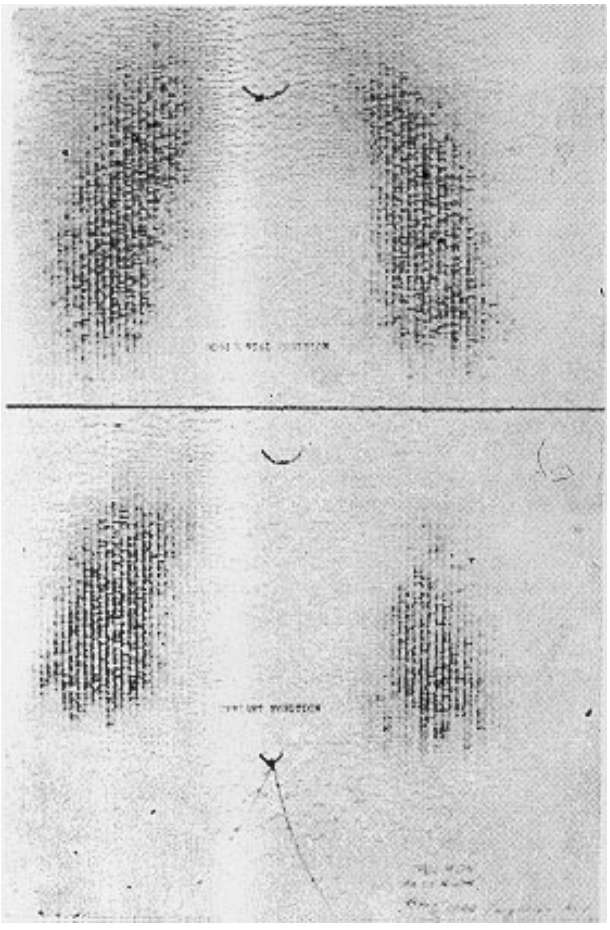


Fig. 4 - A: Lung scanning performed in upright position showed a preferential perfussion of the medular portion of both lungs. B: The same patient studied in prone position displayed a slight increase of vascularity in bases.

It has the hazard of lesion of the conduction system and the lack of septal contraction with the possibility of balooning into the right ventricle [13]. Only carefully selected patients should be septated and the final decision must be taken in the operation room.

The long term evolution of the patients subjected to septation presented by other surgical groups 13 shows important incapacity due to disfunction of the partitioned univentricular chamber. On the other hand, these patients have the possibility of undergoing an atriopulmonary anastomosis with low surgical risk, having a good functional result in the longterm follow up.

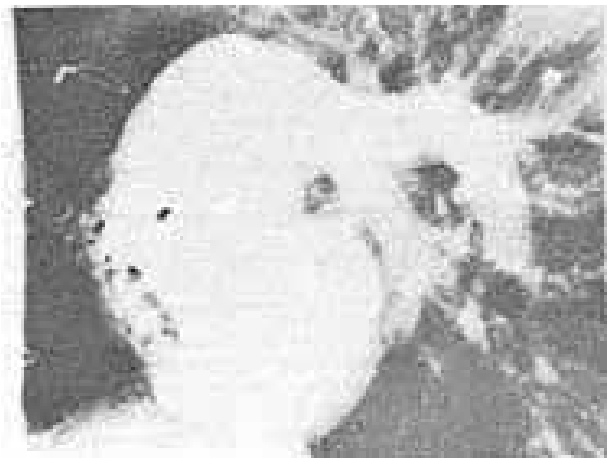


Fig. 5 - Case 2. Right atrial angiography demonstrates good filling of the pulmonary artery and its branches.

Fig. 6 - Case 3. Right atrial injection during ventricular systole. In this case the pulmonary artery was not transected. After suturing the leaflets, and end-to-side atriopulmonary anastomosis was performed. The sutured pulmonary valve leaflets are seen balonning into the main pulmonary artery without blanching. (Arrow).

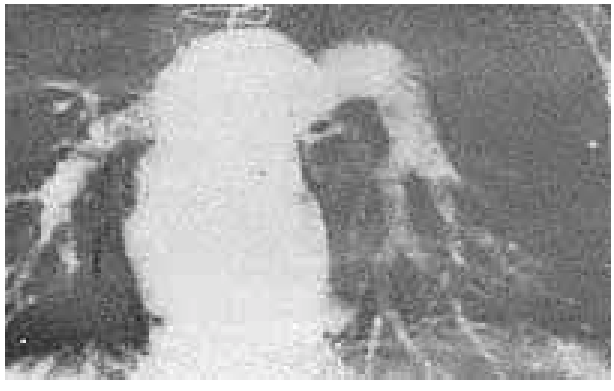


Fig. 7 - Case 3. Right atrial injection in ventricular diastole. The pulmonary sinus of valjalva are opacified (Arrow).

For atriopulmonary anastomosis, selection of the proper place for the single atriotomy is important, as it should be used to close the ASD, the right AV valve when patent, and to connect the right atrium with the pulmonary artery. A direct anastomosis should be performed when possible. It should be as large as possible to be non restrictive to venous flow. If necessary an anterior pericardial patch may be used to obtain an acceptable large anastomosis. In the presence of an adequate surgical indication a right atrial pressure higher than 20 mmHg,

Tabela VIII - Postoperative hemodynamic data in four survivors after atriopulmonary anastomosis.

Case	R.A.	P.A.	L.A.	L.V.E.D.	Ao-Sat
C.P.	20	18	6	6	96,5%
S.V.	10	10	6	6	97,0%
D.N.	10	10	6	6	96,8%
S.A.	12	12	8	8	96,0%

Mean right atrial pressure was below 12 mmHg, in the last 3 cases in whom a large direct atriopulmonary anastomosis was obtained; it was 20 mmHg. in the first patient, in whom a 22 mm. anterior dacron tube was implanted. This is the only case in whom a pressure gradient was registered. The main left atrial pressure through the left ventricle) was always normal and equal to the end dyastolic pressure of the ventricle. Aortic saturation was normal in all cases. Last patient has not yet been cathet.

should suggest a restrictive anastomotic orifice (only if fluid overloading has been ruled out). The most favorable anatomic situation is present when the main pulmonary artery is in a close relationship with the right atrial appendage, (L malposition of the aorta). However, in patients with normally related or transposed great arteries, the main pulmonary artery must be transected, allowing the mobilization of the distal end behind the aorta. (fig. 3B and E).

In order to obtain the largest anastomotic orifice, we extend the opening of the pulmonary artery trunk towards the right main pulmonary branch. Previous assessment of the caliber of both pulmonary arteries branches becomes mandatory to avoid any restriction to flow.

Closure of the AV valves may be done with a pericardial patch sutured to the base of the valvular leaflets or with a double running suture in the coaptation border of the leaflets (Fig. 4). Up to the date we have had no disruptions or leaks of this suture as it was previously reported by the Mayo Clinic group [14].

We have tried not to use any foreign material inside the right atrium in order to avoid intimal aposition of fibrinous tissue [9-12] and future obstruction as was reported 15. Moreover, we believe that such valves are potentially dangerous. The insertion of these, prolongs and complicates surgery, and its long term outcome seem to be deterioration.

In order to achieve a correct hemodynamic functioning of an atriopulmonary system it is absolutely esential to have sinus rythm and low left atrial pressure with gradient between right atrium and left atrium [12]. Obviously the left heart must be normal.

The lack of sinus rythm will increase left atrial pressure and therefore patients with arrhythmias will present cardiac failure with pleural effusion[12]. This is not related to the lack of contraction of the right atrium, since it will never function as a pump. We have had this experience in case 1, and in another case of tricuspid atresia with a nodal rythm and a cannon wave which increased the left atrial pressure. Both patients had pleural effusion which disappeared when the sinus rythm was restored.

Pulmonary phasic flow was shown at the postoperative catheterization although no caval valves were used. The right atrial contraction contributes to the hemodynamic of this system simply by producing a phasic pressure wave. It is remarkable to note that all patients with a huge direct atripulmonary anastomosis have a right atrial pressure of 12 mmHg or lower (table 8) but the only patient in whom a dacron tube was used the pressure is 20 mmHg. Currently, we would not use an anterior dacron tube in patients with normally related great arteries as we did in case 1 (fig. 3).

An important question is if these patients are going to develop intrapulmonary shunts in the future, as the cavo pulmonary shunt do [16-19] The phasic flow may preclude the development of intrapulmonary shunts.

Our experience after an eight years follow up period, in 15 patients with atriopulmonary anastomosis in tricuspid atresia, supports the belief that intrapulmonary shunting will not develop in this hemodynamic situation.

In the last five patients, we have used albumin during the first four days of the post-operative course (1 g/Kg/day). No important pleural effusions have been seen since.

The exercise tests have shown a good functional capacity.

This point together with a low surgical risk, render this operation a good palliative treatment for such a complex malformation. This operation should be indicated to those patients older than 5 years of age, with increased cyanosis and functional incapacity [12]. Pulmonary vascular resistances should be lower than 4 units [20]. Left end diastolic pressure should be less than 12 mmHg. The only exception should be patients with over shunting and left diastolic pressure above this limit but with less than 4 units of pulmonary vascular resistances.

Pulmonary arteries should be normal not having any restriction to flow.

In conclusion for this group of patients open heart palliation should be avoided. When septation is not feasible, atriopulmonary anastomosis seems to be the procedure of choice, offering a low surgical risk and a good long term hemodynamic situation.

Addendum: After completion of the manuscript two other patients with univentricular heart and low pulmonary vascular resistances were operated upon.

A 14 years old girl with a double inlet univentricular heart with anterior aorta was subjected to septation and a porcine heterographt reconstruction of the right outflow tract was performed. Her postoperative course was uneventful and she is in sinus rhythm and asymptomatic.

A 10 years old girl with dextrocardia, hipoplastic right AV valve and normal arterial relationship was operated, and an atriopulmonary anastomosis acomplished.

The hipoplastic right AV valve and the atrial septal defect were closed with a direct suture.

Atriopulmonary continuity was obtained, transecting the main pulmonary artery (fig. 3E) oversewing the proximal opening, and the dis-

tal end was passed behind the aorta. Therefore the pulmonary artery resembled the medial position it has in corrected transposition of the great arteries. The postoperative course was uneventful. The patient is acianotic, in sinus rythm, asymptomatic without medication. This is our technique of choice in patients with normally related great arteries.

Resumo

Um verdadeiro desafio cirúrgico é a decisão da técnica a ser empregada na correção do ventrículo único; a escolha prende-se, fundamentalmente, a três técnicas diferentes: 1) anastomose atriopulmonar com ou sem o emprego de valva entre o átrio e a artéria pulmonar; 2) septação intraventricular; 3) remoção parcial da estenose pulmonar,

Para avaliar o melhor critério, foram estudados 12 pacientes operados com o auxílio de CEC, a partir de 1974, divididos em 3 grupos:

Grupo I - Em 3 casos, operados em 1974, a anomalia foi um achado cirúrgico, sendo realizada a liberação parcial da obstrução pulmonar por apresentar anatomia desfavorável à septação. Não houve mortalidade imediata; a evolução tardia foi boa, encontrando-se 2 pacientes assintomáticos e o outro com cianose leve e incapacidade funcional moderada.

Grupo II - Em 3 pacientes, realizou-se a septação ventricular, sendo que 1 paciente morreu no pós-operatório imediato, outro subitamente, 3 anos após, e o sobrevivente mantém excelente capacidade funcional, sem o auxílio de medicação.

Grupo III - Em 6 pacientes, foi feita a anastomose atriopulmonar, com 1 óbito imediato. No pós-operatório tardio, foram feitas ergometria, cintilografia pulmonar e evolução clínica cardiológica. As provas de esforço demonstraram capacidade funcional dentro dos limites da normalidade. Os dados do cateterismo mostraram ausência de curto-circuitos, pressão M de átrio direito entre 10 e 20 mm Hg, do átrio esquerdo entre 6 e 8 mmHg e pd$_2$ ventricular normal em todos os casos; 6 fígado foi palpado entre 1 e 2 cm da reborda costal, não existindo albuminúria em nenhum caso.

Conclusão: a anastomose atriopulmonar é uma boa possibilidade cirúrgica, acarretando baixo risco. A septação é a melhor situação hemodinâmica, mas somente se toma possível existindo Valvas atriaventriculares normais, sendo seu risco cirúrgico maior.

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