PEDIATRIC CARDIOLOGY

Palliation of Complex Cardiac Anomalies With Subaortic Obstruction: New Operative Approach

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The modified Fontan operation for complex cardiac anomalies associated with subaortic obstruction entails a high surgical risk. It is likely that ventricular hypertrophy secondary to chronic pressure overload plays a significant role. This problem was approached with a new type of palliative operation comprising both a proximal pulmonary artery to ascending aorta anastomosis and a bidirectional cavopulmonary anastomosis.

This operation was performed in six children ranging in age from 26 to 63 months. There was one intraoperative death due to hemorrhage. In one patient, a pulmonary to aorta conduit caused compression of the right coronary artery; the problem was solved by lengthening the conduit with a second period of cardiopulmonary bypass.

The five survivors experienced an uneventful postoperative course. Repeat cardiac catheterization in these five patients showed low pressure in the cavopulmonary system (mean 10 mm Hg), absence of a gradient at rest between the systemic ventricle and aorta and fair arterial oxygenation (mean 82%). A technetium-99m perfusion lung scan visualized a slight prevalence of pulmonary blood flow ipsilateral to the shunt in three cases, whereas in one case preferential flow to the right lung was associated with a narrowing at the site of the cavopulmonary anastomosis. Mild hypoperfusion of the anterior pulmonary segments was observed in two cases.

Both pressure and volume overload are abolished with this procedure and a satisfactory oxygenation is provided. Low venous pressure in the coronary, hepatic and renal areas as well as the short bypass time may explain the smoothness of the postoperative course in our patients. It is conceivable that oxygenation can be improved by a modified Fontan operation at a lower operative risk than is obtained with a single stage procedure because of regression of ventricular hypertrophy. Long-term follow-up indicates the value of this operation as a form of definitive palliation.

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Complex cardiac anomalies, currently amenable to repair by a modified Fontan operation (1,2), can be complicated by the presence of subaortic obstruction, either spontaneous (3,4) or more frequently as a consequence of pulmonary artery banding (4–7). Alternatives to pulmonary artery banding have been devised. Freedom (7), Jonas (8), Rothman (9) and their coworkers advocate a Norwood-type palliative procedure in early infancy for all patients with univentricular heart or tricuspid atresia with a subaortic outlet chamber. Early orthoterminal repair, if calculated pulmonary vascular resistance is low, is theoretically an alternative choice (10).

When subaortic stenosis has developed, with or without a

pulmonary artery band in place, surgical options vary. Relief or bypass of the subaortic obstruction together with an atriopulmonary connection has been described (11–16). Results were inconsistent, but early mortality was generally high. Early take-down of the atriopulmonary anastomosis was necessary in some cases (17). Lin et al. (18) obtained excellent results with a main pulmonary trunk to ascending aorta end to side anastomosis and atriopulmonary connection in patients with univentricular heart and subaortic stenosis after pulmonary artery banding.

Barber et al. (19), having experienced a high mortality rate in patients <10 years of age with a univentricular heart and obstructed outlet foramen, favor a staged approach, consisting of preliminary resection of the subaortic obstruction followed by a modified Fontan operation.

Isolated resection of subaortic stenosis was successful in six of nine patients in the series of Newfeld and Nikaidoh (20), but heart block developed in two survivors. Direct resection of subaortic stenosis, together with or before an

Table 1. Summary of Patient Data

Patient	Age (mo)	Weight (kg)	Diagnosis	Previous Operations	Outcome
1	26	9.8	Double inlet LV, l-TGA	PAB (4 mo)	Operative death
2	28.5	10.5	Complete AVSD with RV dominance, DORV, restrictive VSD	PAB (2.5 mo)	Survivor
3	45	13	Situs inversus, DOLV, uncommitted VSD, l-MGA	PAB (6.5 mo) left BT (20.5 mo)	Survivor
4	31	10.5	Double inlet LV, l-TGA, hypoplastic left AV valve	PAB + atrial septectomy (4 mo)	Survivor
5	34	11.8	Double inlet LV, l-TGA	PAB (2 mo)	Survivor
6	63	14.5	Criss-cross heart, l-TGA, VSD, hypoplastic RV, LSVC, LJAA	PAB (27 mo)	Survivor

AV = atrioventricular; AVSD = atrioventricular septal defect; DOLV = double outlet left ventricle; DORV = double outlet right ventricle; LJAA = left juxtaposition of the atrial appendages; l-MGA = l-malposition of the great arteries; LSVC = left superior vena cava; l-TGA = l-transposition of the great arteries; LV = left ventricle; PAB = pulmonary artery banding; RV = right ventricle; VSD = ventricular septal defect.

atriopulmonary connection, is also advocated by Fontan (10). We have approached this problem in six children with a new type of palliation: the operation combines a main pulmonary artery to ascending aorta anastomosis with a bidirectional cavopulmonary anastomosis.

Methods

Study of patients (Table 1). Six children were operated on between July 1986 and June 1987. Median age at surgery was 31 months and median weight 10.5 kg. Diagnoses included both univentricular (Patients 1, 4 and 5) and biventricular anomalies (Patients 2, 3 and 6). Common clinical findings were cardiac failure, growth retardation and cyanosis. Patient 4 also suffered from multiple episodes of syncope. Two-dimensional echocardiography showed dynamic subaortic obstruction and ventricular hypertrophy in all.

Pulmonary artery banding had been performed 22 to 39 months (mean 29) before the operation in all cases. During the interval from pulmonary artery banding to operation, frequent clinical assessments and at least two cardiac catheterizations were performed in each patient. The latter included, in five cases, a standard isoproterenol infusion (1.6 μg/ml) to obtain a heart rate of 180 to 200 beats/min. This procedure unmasked a severe subaortic obstruction in two cases (Patients 1 and 4) and moderate obstruction in two (Patients 5 and 6); in Patient 2 a significant pressure gradient was found at rest. Angiography (Fig. 1) strongly suggested subaortic obstruction in Patient 3. although a gradient could not be measured.

Hemodynamic data are summarized in Table 2. None of these patients was considered suitable for anatomic repair. A Fontan procedure associated with relief or bypass of the subaortic obstruction was deemed excessively hazardous. A review of 10 recent surgical series (9,11,12,14–19,21) revealed a cumulative early mortality rate of 39% (Table 3).

Surgical technique (Fig. 2). Through a standard median

sternotomy, a moderately hypothermic cardiopulmonary bypass was established by cannulating the distal ascending aorta and the venae cavae separately. Cold cristalloid cardioplegia was used for myocardial preservation. Previous shunts were ligated and divided. Removal of the pulmonary artery band was followed by transection of the main pulmonary trunk and pericardial patch closure of the pulmonary artery confluence. The confluence arterioplasty was incomplete in Patient 4 (Fig. 2a inset), as a residual opening was left on the right side.

With aortic cross-clamping (mean duration 34 min, range 23 to 57), an end to side anastomosis was performed between the proximal main pulmonary artery and the ascending aorta. This was direct in Patient 2 and with interposition of a Dacron conduit (diameter 16 to 20 mm) in the other five patients. After ligation of the azygos vein, a bidirectional, end to side cavopulmonary anastomosis was established between the superior vena cava and the cranial aspect of the undivided right pulmonary artery (four patients), left pulmonary artery (one patient) or the residual opening of the pulmonary artery confluence (one patient). The superior vena cava-right atrial junction was closed by direct suture.

Results

Operative mortality. There was one operative death (early mortality rate 17%). In Patient 1 the procedure consisted initially of bypass of the subaortic obstruction, atrial septectomy and systemic to pulmonary shunt. Attempts to wean the patient from cardiopulmonary bypass failed because of inadequate pulmonary blood flow from an intrapericardial Blalock-Taussig shunt. A bidirectional cavopulmonary anastomosis was constructed and cardiac function improved, permitting cardiopulmonary bypass to be terminated. However, uncontrollable bleeding from the aortopulmonary anastomosis eventually caused the patient's death. Lung histologic study showed no vascular changes.
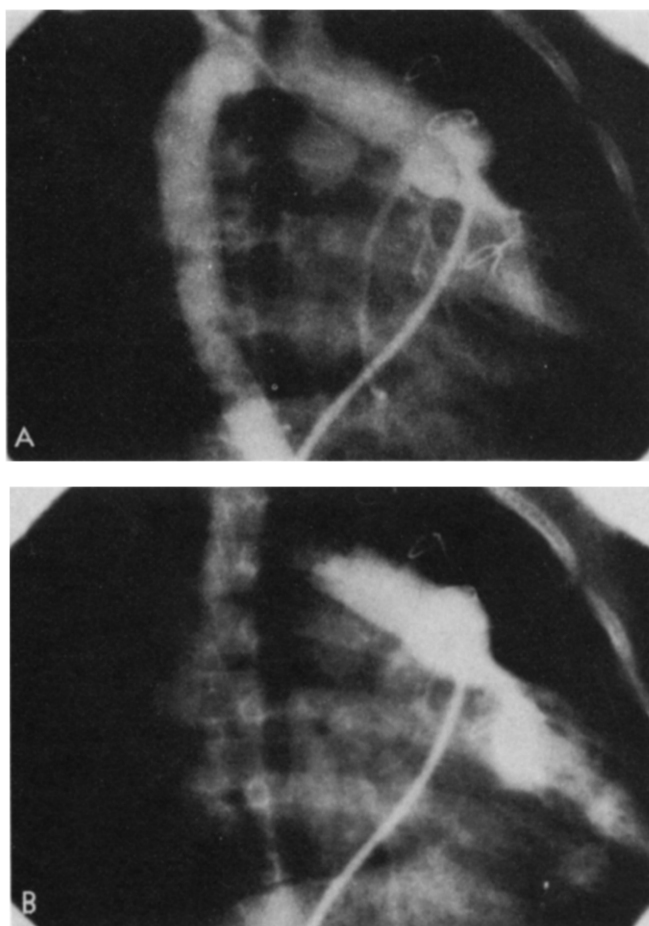
Figure 1. Patient 3. Left ventricular contrast injection in the right anterior oblique view demonstrating the obstructive subaortic infundibulum in a patient with double outlet left ventricle, uncommitted ventricular septal defect and corrected malposition of the great arteries. A, Systole; B, diastole.

Postoperative course. Four patients (Cases 2, 3, 4 and 6) were easily weaned from cardiopulmonary bypass and experienced an uneventful postoperative course. They re-

Table 3. Modified Fontan Operation for Univentricular Heart With Subaortic Obstruction: Previous Reports

First Author and Reference	Year	Patients (no.)	
		Total	Survivors
Yacoub (11)	1976	1	1
Moreno-Cabral (15)	1980	2	2
Doty (12)	1981	2	2
Penkoske (21)	1984	7	4*
Barber (19)	1984	13	7†
Waldman (14)	1984	1	1
Lin (18)	1986	6	6
DeLeon (17)	1986	3	2‡
Stark (16)	1986	7	2
Rothman (9)	1987	2	0
Total		44	27
			(61%)

*In a subsequent report from the same institution (7), there were no long-term Fontan survivors (number of patients unstated) except two patients in whom an arterial switch operation was associated with the Fontan operation; †types of procedure to relieve the subaortic obstruction unstated; ‡these patients survived with early take-down of the Fontan operation and were left with a pulmonary artery to ascending aorta anastomosis and a classical Glenn shunt.

ceived mild inotropic support and were extubated 24 to 48 h after the procedure. Pleural effusion, ascites and peripheral edema were not observed. These patients were discharged 6 to 14 days postoperatively.

The intraoperative course of Patient 5 was complicated by compression of the right coronary artery by the pulmonary to aorta conduit; the bypass was reinstituted and the conduit appropriately lengthened. The postoperative period was characterized by moderately depressed ventricular function and prolonged need of inotropic support. The patient was discharged 2 weeks after surgery in good clinical condition.

All five survivors had sinus rhythm. Examination of the fundus oculi did not reveal increased intracranial pressure in any patient.

Table 2. Hemodynamic Investigation in Six Cases

	Preoperative Data					Postoperative Data				
	Basal Condition			After Isoproterenol Test		Basal Condition			After Isoproterenol Test	
Patient	Syst Sat. (%)	MV Press (mm Hg)	MV-Ao Grad (mm Hg)	MV Press (mm Hg)	MV-Ao Grad (mm Hg)	Syst Sat (%)	MV Press (mm Hg)	MV-Ao Grad (mm Hg)	MV Press (mm Hg)	MV-Ao Grad (mm Hg)
1	92	110/0 to 6	5	140/0 to 10	60	—	—	—	—	—
2	84	155/0 to 11	60	—	—	81	80/0 to 3	0	105/0 to 6	15
3	81	115/0 to 9	0	135/0 to 9	5	83	90/0 to 3	0	70/0 to 2	5
4	82	110/0 to 4	10	160/0 to 10	70	79	100/0 to 7	0	125/2 to 6	25
5	75	90/0 to 7	0	110/0 to 7	30	85	85/0 to 3	0	90/0 to 5	15
6	80	80/0 to 4	0	100/0 to 7	30	83	115/0 to 4	5	120/0 to 10	10

Ao = aorta; Grad = gradient (systolic); MV = main ventricular chamber; Press = pressure; Sat = oxygen saturation; Syst = systemic.
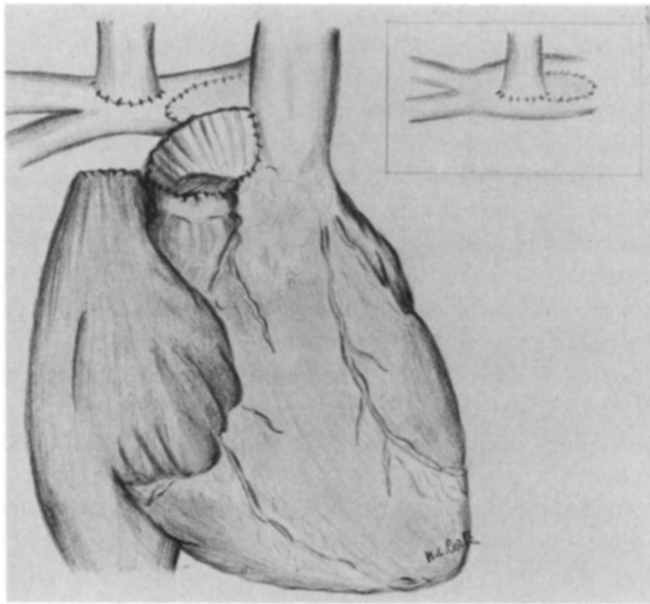
Figure 2. Surgical technique. See text for explanation. Inset shows the incomplete confluence arterioplasty in Patient 4.
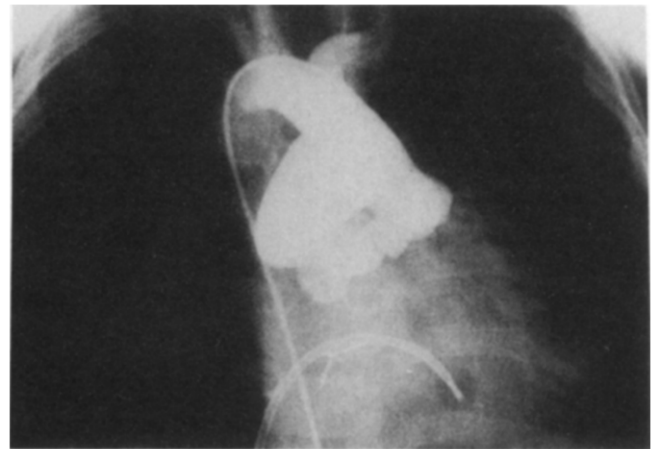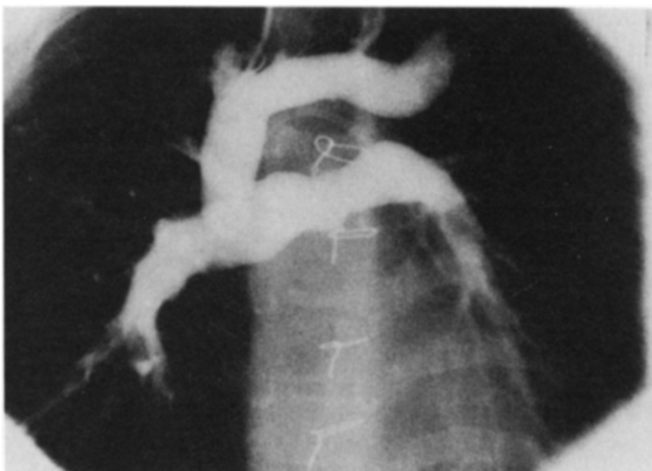

Figure 4. Patient 3. Postoperative contrast injection in the aortic root (posteroanterior view) showing the wide pulmonary-aorta anastomosis and competent semilunar valves.

Follow-up. The five survivors were in good condition at the last follow-up assessment, 7 to 18 months (median 9) postoperatively. One patient requires moderate diuretic treatment and one requires digitalis and diuretic treatment. Hemoglobin level ranged from 14.6 to 18.1 g/dl (median 15.5). All patients are able to attend school or preschool groups.

Postoperative hemodynamic study. Invasive restudy was carried out in all five patients at a mean interval of 57 days from surgery (range 12 days to 9 months). The arterial oxygen saturation averaged 82% (range 79 to 85%) and the

Figure 3. Patient 3. Postoperative contrast angiography of superior vena cava in the posteroanterior view showing the bidirectional cavopulmonary anastomosis.

pressure in the cavopulmonary system averaged 10 mm Hg (range 5 to 16). The rest gradient between the main ventricular chamber and the aorta was abolished; a mild gradient was disclosed by the isoproterenol test. A reduction of main chamber end-diastolic pressure was demonstrated in Patients 2, 3 and 5, and a mild increase in Patient 4; it was unchanged in Patient 6. On angiography, the cavopulmonary (Fig. 3) and the pulmonary artery to ascending aorta (Fig. 4) anastomoses were patent, and both semilunar valves were competent.

Technetium-99m perfusion lung scan in four patients visualized a moderate predominance of pulmonary blood flow ipsilateral to the shunt side: 63% versus 37% in Patient 2, 57% versus 43% in Patient 3, 56% versus 44% in Patient 6 and 78% versus 22% in Patient 4. In Patient 4 angiography disclosed narrowing of the pulmonary confluence at the anastomotic site (Fig. 5). In this patient, a residual opening of the pulmonary confluence had been used for the anastomosis. In two patients, there was mild hypoperfusion of the anterior pulmonary segments (scan performed in the supine position).

Discussion

Role of subaortic obstruction in the selection of operative procedure. Several drawbacks diminish the importance of pulmonary artery banding as a palliative method in infants for whom a Fontan procedure is anticipated (7,22). The band may cause distortion of the pulmonary arteries (23), thereby increasing the risk at orthoterminal repair (24). Subaortic obstruction may occur as early as weeks after banding (5–7,24). In these patients, the "main" ventricular chamber is subjected to both pressure and volume overload; ventricular hypertrophy and dilation often follow (5,6,25,26).

Figure 5. Patient 4. Contrast angiography of superior vena cava (posteroanterior view) showing the bidirectional cavopulmonary anastomosis with the narrowed pulmonary confluence near the anastomotic site.

The grading of "obstruction" remains controversial. Although surgery has often been unsuccessful in patients with a severe pressure gradient ($\geq$75 mm Hg) (9,18,20), the criteria for defining "subaortic obstruction" in the single ventricle domain have not been established (4,9,14,18,20). We endorsed the suggestions of Freedom et al. (4,6,7) and accepted a broad range of hemodynamic and angiographic criteria to define significant obstruction; we also routinely adopted a provocative test during the preoperative evaluation of patients "at risk" for this complication.

Ventricular hypertrophy is a recognized risk factor in patients undergoing the Fontan operation (27). Reduced ventricular compliance (19) and increased sensitivity to ischemic damage (26,27) have been implicated. Results of the application of the Fontan principle to this context have been disappointing (Table 3). We elected a palliative approach, endorsing the concepts of Barber et al. (19) and Newfeld et al. (20), with bypass of subaortic obstruction plus a cavopulmonary shunt.

Bypass of the subaortic obstruction. The proximal pulmonary artery to ascending aorta anastomosis is an established method of bypassing a subaortic obstruction in a variety of situations (8,11,28,29). Its efficacy in relieving the rest "main" ventricle-aortic gradient is substantiated by our postoperative data. This extracardiac technique compares favorably with the direct resection of subaortic obstructing muscle. Drawbacks of the latter technique are incomplete relief of the obstruction and injury to the conduction system, the coronary arteries and the atrioventricular valve tensor apparatus (18,20).

One concern related to use of the pulmonary artery to ascending aorta anastomosis is the possibility of late development of pulmonary and aortic valve regurgitation. Our postoperative angiographic data and experience with the

Damus-Kaye-Stansel operation (28,29) suggest that the pulmonary valve can adapt to the new conditions and function as the systemic semilunar valve. Aortic regurgitation seems to occur predominantly in aortic valves that are immobile throughout the cardiac cycle (30) (di Carlo DC, Annichiarico M, Giannico S, Ballerini L. Development of aortic regurgitation after conduit surgery for relief of subaortic obstruction. Unpublished observations).

One can speculate that ventricular hypertrophy may regress and compliance improve after the relief of subaortic obstruction (19). Recent data from children operated on for congenital aortic stenosis suggest that there may be an increase in left ventricular ejection fraction and end-diastolic and end-systolic indexes (31). Pantley et al. (32) and Schwarz et al. (33) reported that, after aortic valve replacement for aortic stenosis in adults, left ventricular mass and volume regressed, although not to normal levels. Mirsky et al. (34) found a significant decrease in left ventricular end-diastolic pressure in similar patients. In a short follow-up period, we have observed a remarkable reduction of the end-diastolic pressure in three of the five restudied survivors in our series.

Bidirectional cavopulmonary anastomosis. The use of the main pulmonary artery to bypass the subaortic obstruction imposes the creation of an alternative source of pulmonary blood flow. The calibration of pulmonary blood flow is often difficult (18), as demonstrated by Patient 1 in this series. A systemic to pulmonary artery shunt in patients with univentricular heart may provoke congestive heart failure (35,36) and increase the risk of a subsequent Fontan operation (37). The classic cavopulmonary anastomosis (38) is a safe palliative operation with proven long-term efficacy (39,40). Its use as a first stage procedure in patients who are less than ideal candidates for a Fontan operation has been recommended (41,42). A few patients have survived with a Glenn shunt after the take-down of a failed Fontan operation (17).

The bidirectional cavopulmonary anastomosis, first reported on by Azzolina et al. (43), has been revived (38,44,45). This shunt supplies one-third of the systemic venous return to both lungs; the inferior vena cava is left to drain in the pulmonary atrium and its mean pressure is similar to the end-diastolic pressure of the systemic/main ventricle. The venous pressure in the coronary, hepatic and renal areas is consequently low. Common complications of the Fontan operation such as edema and ascites did not occur postoperatively in our patients.

Repeat cardiac catheterization demonstrated that oxygenation was unchanged from the preoperative status in our patients; this level was achieved without volume overload of the systemic or single ventricle. The cavopulmonary system was at low pressure (mean 10 mm Hg) in all cases, suggesting normal diastolic function of the main/single ventricular chamber.

The major long-term hazard of the classic cavopulmo-

nary shunt is the development of pulmonary arteriovenous fistulas (38,46,47). Multiple factors seem to be involved in this phenomenon: preferential distribution of blood flow to the lower pulmonary lobes, extended time from surgery, long-standing hypoxemia and polycythemia (47). Lung scintigraphy in four of our survivors showed that the caval flow was rather uniformly distributed to the lungs, but preferential gravity-induced flow to the posterior segments was also noted in two cases. This occurrence, commonly seen after the classic cavopulmonary shunt, was recently noted in patients with long follow-up after a Fontan procedure. It is not known whether the bidirectional cavopulmonary shunt will behave like a classic cavopulmonary shunt (Glenn) or like a total atriopulmonary shunt (Fontan operation), which has not to date been associated with the development of pulmonary arteriovenous fistulas.

Conclusions. Several surgical options are available for complex cardiac malformations with subaortic stenosis. Because both palliative and corrective procedures entail a high operative risk, the search for alternatives should continue. The combination of proximal pulmonary artery to ascending aorta anastomosis with a bidirectional cavopulmonary anastomosis aims to 1) relieve the ventricular volume and pressure overload, and 2) provide a satisfactory level of oxygenation. In this procedure, unlike the Fontan type of repair, vital organs, such as liver, kidneys and heart, are separated from the hypertensive venous system. This separation might be the reason for the smooth postoperative course of our early survivors. The shortness of bypass time certainly played a positive role in early success. The completion of the Fontan repair, including atrial septation and ventricular exclusion, would have significantly prolonged the duration of cardiopulmonary bypass and of myocardial ischemia.

Close monitoring of patients who have undergone pulmonary artery banding for complex cardiac anomalies is recommended (5–8) and we endorse this concept. The occurrence of subaortic stenosis, with a gradient at rest or on catecholamine testing should dictate the need for early intervention. Angiographic evidence of subaortic narrowing should be treated according to the same criteria. The progress of these patients will be watched carefully. In light of the good results of the classic cavopulmonary anastomosis, a long period of satisfactory palliation can be anticipated with the described procedure. Without volume or pressure overload, the main ventricle should work at its best, the limiting factor being a moderate arterial desaturation.

Time will indicate whether the benefit of this palliative intervention will extend indefinitely, enabling the patient to avoid further surgery. If this is not the case, the possibility of improving arterial oxygenation by atrial septation and atriopulmonary connection is not impaired. Regression of ventricular hypertrophy could produce a better surgical candidate for the Fontan operation, so that the cumulative risk

with a staged procedure might prove lower than that with the one stage operation.

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