SURGICAL MANAGEMENT OF ADULTS WITH CYANOTIC CONGENITAL HEART DISEASE

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Ninety seven patients over 16 years of age with cyanotic congenital heart defects underwent corrective surgery at the Brompton Hospital in London, between June 1971 and June 1983. Tetralogy of Fallot was found in 40 patients, pulmonary stenosis in 30, double outlet right ventricle in 12, transposition of the great arteries in nine, tricuspid atresia in three and pulmonary atresia in three. Cyanosis and effort dyspnoea were the most common preoperative clinical findings. Atrial septal defects, ventricular septal defects and pulmonary stenosis were the most frequently associated anomalies. Thirty-one of the patients had undergone palliative cardiac surgery previously. The overall hospital mortality in this series was 12.4 per cent and varied according to cardiac pathology. Low cardiac output contributed to 58.3 per cent of the deaths, particularly in the group of patients in whom the cardiac ischaemic time exceeded 60 minutes ($p < 0.001$). The ischaemic time and lowest myocardial temperature reached were good predictors of outcome when analysed by discrininant analysis. Actuarial analysis shows a survival probability of 93% and 73% for pulmonary stenosis and Fallot's tetralogy, at a mean follow-up period, of 5 years. This study indicates that surgical correction cyanotic congenital heart defects may be accomplished in adults with a low mortality.

It is now uncommon for a patient with cyanotic congenital heart disease to reach adult life without having corrective surgery [1,2]. Notwithstanding this, some patients will still present later in life, usually with well compensated or palliated defects, often having refused surgery earlier or having been lost to follow-up [3-5].

The purpose of this report is to examine the clinical course of 97 adult patients with cyanotic congenital heart disease who underwent corrective surgery at the Brompton Hospital, London.

PATIENTS AND METHODS

The records of 97 patients over the age of 16 years with cyanotic congenital heart defects, who underwent corrective surgery between June 1971 and June 1983, were retrospectively reviewed. There were 51 women and 46 men, aged 16 to 58 years with a mean Of 29.4yrs + 11.5SD. Their diagnoses together with their respective age ranges are listed in table I.

TABLE I - Distribution of Cardiac Anomalies and Age at Surgical Repair.

Anomaly	Age (years)	Mean + SD (years)	No. of Cases
Tetralogy of Fallot	16 - 55	28.4 + 11.8	40
Pulmonary stenosis	16 - 58	30.4 + 12.2	30
Double-outlet right ventricle	18 - 44	30.6 + 7.4	12
Transposition of the great arteries	16 - 30	22.6 + 13.6	9
Tricuspid atresia	16 - 24	20.0 + 3.2	3
Pulmonary atresia	21 - 41	36.7 + 8.2	3

SD = Standart deviation

The following factors were analyzed with respect to 30 day hospital mortality: 1) General variables - age, haemoglobin and body surface area; 2) Morphologic variables - diagnosis, associated anomalies and prior surgery; 3) Preoperative haemodynamics - venous saturation, arterial saturation, right ventricular end diastolic pressure and left ventricular end diastolic pressure; 4) Operative technique - aortic crossclamp time, lowest myocardial temperature reached,

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method of myocardial protection and type of right ventricular outflow tract reconstruction.

The minimum haemoglobin was 17.5g/dl and the maximum 26.6g/dl with mean of 16.6g/dl + 3.8SD. The body surface area ranged from 1.16 to 1.75 square meter with mean of 1.30 + 0.19SD.

Thirty one of the patients had undergone previous cardiac surgery. Closed infundibulectomy (Brock's-operation) and pulmonary valvotomy had been performed in 12 patients with tetralogy of Fallot and 2 with double-outlet right ventricle. Twelve Blalock Taussig shunts had been performed in eleven patients. Two patients had had Waterston's anastomoses constructed and 2 Brock's infundibulectomy in association with a systemic artery to pulmonary artery shunt. One patient with transposition of the great arteries with ventricular sepal defect had had a Blalock Hanlon operation. A Gleen operation had been performed in one patient with tricuspid atresia. None of the patients with isolated pulmonary valve stenosis had undergone previous surgery.

The venous saturation ranged from 29.0% to 83.0% with a mean of 62.3% + 9.6SD and the arterial saturation ranged from 44.0% to 99.0% with mean of 87.3% + 12.1SD. The right ventricular end diastolic pressure (RVEDF) ranged from 3.00-24.00 mmHg with mean of 9.8 mmHg + 4.4SD and the left from 3.0-28.0 mmHg with mean of 10.6 mmHg + 6.2SD.

Means and standard deviations were calculated for each group. Differences between groups were analyzed by Gaussian or distribution-free tests as seemed appropriate. Differences in proportions were investigated by Chisquare and Fisher's Exact tests. The relationships between variables were considered using multiple regression analysis.

The actuarial survival curve was obtained with an end point determined solely by the patients death or the last follow-up visit. This analysis took no account of the number of serious incidents or the clinical condition of the patients.

Pre operative symptomatology and complications - The spectrum of symptom and signs are listed in table II. All the patients had detectable cyanosis at rest, thereafter the most common symptom was effort dyspnoea in 79 patients (81.4%). Squatting was a feature in 15 patients (15.5%).

Technique of intracardiac repair - The techniques and types of repair used varied, as the study covers a 12 year period. Thus a variety of oxygenators and methods of myocardial preservation were used. Most recently myocardial preservation with cold cardioplegia and external cardiac cooling has been pre ferred.

Tetralogy of Fallot - The ventricular septal defect and infundibulum were aproached through a vertical incision made in the infundibulum of the right ventricle. The usual hypertrophy of the septal and parental insertions of the infundibular septum were excised and the ventricular septal defect delineated. The ventricular septal defect was closed using a woven dacron patch sewn in place with buttressed interrupted sutures. Right ventricular outflow tract obstruction was relieved in different ways. Pulmonary valvotomy with or without infundibulectomy was performed in 5. Infundibulectomy plus transannular enlargement of the outflow tract using prosthetic or biological materials was required in 32 patients. An external valved conduit was necessary in three patients. An atrial septal defect was closed in four patients and one patient who had aortic regurgitation needed aortic valve replacement with a Starr Edwards prosthesis. Six patients (15%) died due to low cardiac output, one of these had equal pressure in both ventricle after the repair and another a had poor right ventricular function. Two patients died as a result of bleeding, one from damage to a calcified Blalock-Taussig shunt. One patient needed reoperation for closure of a residual ventricular septal defect and died subsequently due to bacterial endocarditis.

TABLE II - Preoperative Symptomatology, Complications and Clinical Findings.

	Tetralogy of Fallot	Pulmonary stenosis	DORV	TGA	TA	PA
Cyanosis	40	30	12	9	3	3
Effort dyspnoea	28	24	12	9	3	3
Squatting	10	1	1	-	1	2
Angina pectoris	2	5	1	-	1	-
Effort syncope	6	1	1	1	-	1
Paroxysmal nocturnal dyspnoea	5	-	-	1	-	-
Vertigo	5	-	-	-	-	-
Frequent repiratory infection	5	-	-	-	-	-
Headache	4	-	1	1	-	-
Congestive heart failure	3	1	1	1	-	-
Endocarditis	2	-	2	-	-	-
Orthopnoea	2	1	-	-	-	-
Sub acute rheumatism	1	-	-	-	-	-
Paresthesia, (cerebral thrombosis)	1	-	-	-	-	-
Calcification of the aortic valve	1	-	-	-	-	-

DORV = double-outlet right ventricle; TGA transposition of the great arteries; TA = tricuspid. atresia; PA pulmonary atresia.

Pulmonary stenosis - Twenty-three patients open pulmonary valvotomy, or a combination of valvotomy with infundibulectomy. This was performed through a vertical incision made in the pulmonary artery and continued into the infundibulum of the right ventricle, if necessary, to allow excision of hypertrophied septal and parietal insertions of the infundibular septum. Seven patients had a infundibulotomy plus a transannular patch. Atrial defects were closed in six patients and ventricular septal defects in five.

Double-outlet right ventricle - Ten patients a vertical incision made in the right ventricle which demonstrated a large doubly-committed ventricular septal defect. The aorta arose from the right ventricular. The left ventricle was directed to the aorta using a large patch of "dacron" cloth sewn in with interrupted buttressed sutures.

The reconstruction of the right ventricular out-flow was performed using a gusset in 6 patients, including a valved homograft in one and an external valved conduit from right ventricle to pulmonary trunk in four.

A palliative Mustard operation was carried out in one patient who had pulmonary hypertension. An exploratory choracotomy was performed in another patient with double-outlet right ventricle, pulmonary stenosis, complete atrioventricular canal and common atrium. After careful inspection through the right ventricle it was mot found to be possible to route the blood from the left ventricle to the aorta. A palliative infundibulectomy and open pulmonary valvotomy was therefore carried out.

Transposition of the great arteries - The Mustard procedure was performed in two patients. One bad transposition of the great arteries, ventricular septal defect and pulmonary hypertension. The other had transposition of the great arteries, ventricular septal defect and pulmonary stenosis. Both patients also had large atrial septal defects. Systemic venous inflow was switched in each case using a previously fashioned bovine pericardial baffle according to the design of Brom. In one patient the ventricular septal defect was closed through the right ventricle and an open pulmonary performed. In the case with pulmonary hypertension the ventricular septal defect was left open.

Two patients with transposition of the great arteries, ventricular septal defect and pulmonary stenosis underwent a Rastelli operation. A vertical incision was made in the infundibulum of the right ventricle. Large sub aortic ventricular septal defects were closed, directing the left ventricle to the aorta. A transverse incision was then made in line with the confluence of the left, right and main pulmonary arteries. The pulmonary valve was closed with a continuous suture. A previously prepared, dacron and homograft composite conduit was anastomosed end-to-side with the pulmonary artery using a continuous suture and end-to-side to the ventriculotomy using interrupted and continuous sutures.

Two patients had corrected transposition of the great arteries. In one, this was associated with an atrial septal defect, a ventricular septal defect and pulmonary stenosis. In the other it was associated with pulmonary stenosis and an aneurysm of the membranous septum. The atrial septal defects were closed with a continuous suture. In the first patient, the ventricular septal defect was also approached through the right atrium and atrioventricular valve. The interrupted sutures for the patch closure were placed through the ventricular septal defect, picking up the left side of the septum, so as to avoid the conduction tissue. Pulmonary was performed through a vertical incision in the pulmonary trunk. In the second patient, the large aneurysm of the septum obstructed the pulmonary outflow tract. This patient had a homograft external valved conduit placed between the morphologically left ventricle and pulmonary trunk.

Three patients had transposition of the great arteries with single ventricle (double-inlet atrioventricular connexion with ventrículo-arterial discordance). One patient had a modified Fontan's procedure. The tricuspid valve was closed with two layers of continuous sutures. A porcine heterograft conduit was interposed between the right atrium and the pulmonary trunk. The large atrial septal defect was closed directly. Following satisfactory postoperative progress initially, this patient developed persistent bilateral effusions and died after a massive gastro-intestinal haemorrhage. One patient had septation of a main chamber and Mustard's operation and died on the third postoperative day due to hypoxia secondary to lung disease. The third patient was explored with a view to septation, but this proved impossible.

Tricuspid atresia - Two patients with tricuspid atresia underwent Fontan's procedure. The right atrial appendage was opened at its apex and the patient foramen ovale closed with a continuous suture. The main pulmonary artery was cut at the level of the valve and the malformed valve closed off. An aortic homograft was interposed between the right atrium and the pulmonary trunk. One patient died from low output failure. One patient had a Fontan's procedure following a Glenn operation. Cardiopulmonary bypass was established with femoral artery and venous cannulation. No attempt was made to take down the Glenn anastomosis.

Pulmonary atresia - in the three patients with pulmonary atresia, and ventricular septal defect a vertical incision was made into the infundibulum of the right ventricle. This entered a densely hypertrophied infundibular chamber. Ventricular septal defects were closed in three patients and a patent foramen ovale closed through the right atrium in one. A previously prepared homograft external valved conduit was then placed between the infundibulum of the right ventricle and the pulmonary trunk, thereby re-establishing continuity between the main pulmonary artery and right ventricle.

RESULTS

Overall hospital mortality - There were 12 early deaths (12.4%) ie. within 30 days of surgery (table III). Most deaths occured between 1971 and 1978; 61 patients were operated on with 10 deaths (16.4%). Since 1978, 36 patients have been operated on with only 2 deaths (5.5%).

The main postoperative complication was congestive heart failure which was noted in 11 patients, bleeding in 10 patients of whom 8 required reexploration and 2 died; dysrrhythmias were observed in 4 patients and respiratory failure in 2 (table IV).

No estatistically significant differences were found in the general variables when related to outcome.

TABLE III - Operative mortality in adult patients with cyanotic congenital heart disease.

Anomaly	No. Patients	Hospital Death	%
Tetralogy of Fallot	40	9	22
Pulmonary stenosis	30	-	
Double out Met right ventricle	12	-	
Transposition of the great arteries	9	2	22
Tricuspid atresia	3	1	33
Pulmonary artery	3	-	

TABLE IV - Postoperative complications.

	T.F.	P.S.	D.O.R.V.	T.G.A.	T.A.	P.A.
Congestive heart failure	6	2	2	-	-	1
Bleeding	6	-	2	1	-	1
Dysrythmias	3	1	-	-	-	-
Respiratory failure	-	1	-	-	1	-

TF = tetralogy of Fallot; ps = pulmonary stenosis; DORV = double-outlet right ventricle; TGA = transposition of the great arteries; TA = tricuspid atresia; PA = pulmonary atresia.

Sixty patients had an associated lesion, and five (8.3%) died. Of all the patients without associated lesions, seven (18.9%) died. This difference was not significant.

Five patients (16.1%) who had previous palliative procedure died. The mortality in this group was higher than in the group of patients who had mot had previous surgery (10.6%), but not significantly so.

Preoperative variables - None of the preoperative variable - venous saturation, arterial saturation, and right and right and left ventricular diastolic pressures - had a statistically significant influence on postoperative mortality.

Operative variables -The group of patients who had an aortic cross clamping time in excess of one hour had a significant higher mortality (45%) than the patients who had less than one hour (6.5%) ($p < 0.001$).

Lowest temperature during the cardiopulmonary bypass ranged from 20 C to 36 C with mean of 30 C + 4SD. No death was noticed in a group of patients whom had normothermia (36C) and the higher mortality rate was found in a group of patient with moderate hypothermia (18.2%) . There was no significant statistical difference in hospital mortality according to the degree of hypothermia. However a combination of the lowest myocardial temperature and aortic cross clamp time relating to death is shown to be a very good predictor when analysed by discriminant analysis (fig. 1). A straight line gives the two group (dead/alive) and shows, for any given temperature, the maximum cross clamping time which is safe or vice-versa.

There was no significant difference in hospital mortality which related to the type of myocardial protection used.

Twenty-eight patients required a pulmonary valvotomy with or without infundibulectomy (management of right ventricular outflow tract obstruction). In this group two

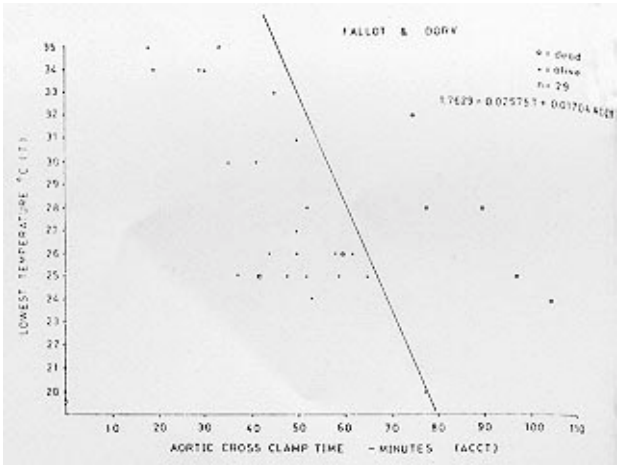

Fig. 1 - Discriminant analysis of myocardial temperature and aortic cross clamp time relating to death for patients with tetralogy of Fallot and double-outlet right ventricle.

patients died (7.1%). Seven hospital death (15.5%) occured in 45 patients who required infundibulectomy with a transannular patch. No death occured among the 7 patients who required, an external conduit from right ventricle to pulmonary artery. The risk of operation was not significantly affected by any particular management of the right ventricular outflow tract obstruction/among he 81 patients who had a diagnosis of tetralogy of Fallot, pulmonary stenosis or double outlet right ventricle.

Follow-up - The 85 survivors were followed up for period ranging from 1 to 11 years (mean 3.5 years). The actuarial survival rate is shown in figure 2. The overall survival for the group with a clinical diagnosis of tetralogy of Fallot and pulmonary stenosis were 73% and 93%, respectively, at 5 years; and 54% and 82%, respectively, at 10 years.

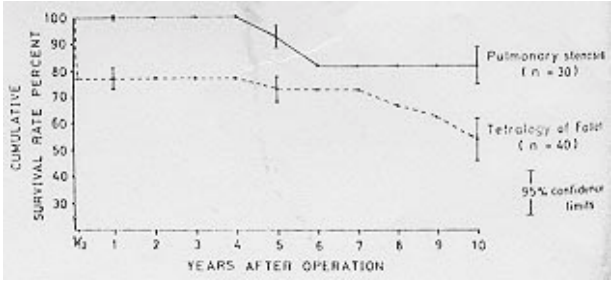

Fig. 2 - Actuarial survival curves for adults patients with tetralogy of pulmonary stenosis.

DISCUSSION

This study is a retrospective review of patients with cyanotic congenital heart disease who reached without previous corrective surgery. have attempted to define the factors that may influence the results of surgical correction in this group.

A review of the literature, shows a wide variation in the frequency with which adult patients with cyanotic congenital heart disease are seen in different centres [3,6-11]. In our series there were 97 patients (1.1

percent) out of a total number of 8.967 adults who underwent cardiac surgery between 1971 and 1983. Among these patients those with tetralogy of Fallot survive longest without surgery, although less than 35 percent survive beyond 10 years of age [1,2,6-8]. In our series, tetralogy of Fallot was the most common lesion found adult cyanotics. Pulmonary stenosis is the second most frequently seen congenital malformation of the heart in adult life [7]. In our study there were thirty such patients and this group was the second largest, followed by double outlet right ventricle, transposition of the great arteries, tricuspid atresia and pulmonary atresia in decreasing order of frequency.

In general adults with cyanotic heart disease present with similar clinical features to children [11]. A great number of patients presenting *de novo* in adult life do so because of a deterioration in their symptoms or become symptomatic again, having been previously well palliated as a child [4,5]. In our series all patients were cyanosed and 66 patients, who had not been palliated previously, had had a change in their symptomatic status. This probably reflects the development of excessive collateral blood flow to the lungs during childhood to the point where the hemodynamic consequences of pulmonary hyperaemia become greatly exacerbated.

Previously published series have indicated a varied operative mortality for patients with adult cyanotic heart disease. Cooley and associates [10] in 1966 reported a mortality rate of 29% for patients with tetralogy of Fallot and no death in the group with pulmonary stenosis. Beach and associates [13] reported in 1971 a series of 64 who had undergone correction of tetralogy of Fallot between the ages of 15 and 48 years with four operative deaths (6.2%). Lavin and colleagues [6] reported a series of 18 patients in 1975 with tetralogy of Fallot and seven with pulmonary stenosis. The mortality rate was 16% and 14 Percent respectively. Kay and colleagues [2] reported in 1976 a 20 percent mortality for patients with tetralogy of Fallot. and patients with pulmonary stenosis. There were no hospital deaths in a series of 11 patients with tetralogy of Fallot and seven with pulmonary stenosis reported by Leidenfrost and his colleagues [5] in 1978. In the same study Leidenfrost reported a high hospital mortality rate (60%) in five adults with complex forms of congenital heart disease. The results obtained in our study are comparable to those of other series. Although in the tetralogy of Fallot group the overall mortality was 22 percent, there has been no death in the past three years. In the smaller group of patients with complex defects, the results were variable, bu the overall mortality acceptably low.

The presence of a collateral circulation, arterial desaturation, increased blood viscosity, and ventricular contraction against an obstructive lesion over many years tend to compromise the myocardium, eventually causing myocardial fibrosis. A low cardiac output state was responsible for 58.3% of the operative mortality emphasizing the importance of good myocardial preservation. The collateral circulation may cause troublesome bleeding. Beach and colleagues [13] also noted increased postoperative bleeding in adult patients due to the abundant collateral circulation. Indeed, postoperative hemorrhage was the second most frequent complication and resulted in the death of 2 patients (2.1%), and angina pectoris was a remarkable clinical feature in our patients (9.3%). It can be postulated that relative coronary insufficiency may result from the increased myocardial oxygen demands of ventricular hypertrophy, superimposed upon the decreased capacity of the coronary circulation to deliver oxygen because of increasing arterial hypoxemia and blood viscosity. The electrocardiograms of patients experiencing chest pain do not differ characteristically from those without pain [1]. The resting electrocardiograms in our patients were unremarkable.

The development of congestive heart failure in tetralogy of Fallot is said to be very rare. However, six (6.2%) of our patient had the symptoms of congestive heart failure. The reasons for the onset of congestive heart failure could be due to the volume loading of the left ventricle secondary to the spontaneous development of a large compensatory systemic to pulmonary arterial collaterals. A similar experience in adults has been reported by others [1].

Abnormalities of the aortic valve in patients with teratology of Fallot are unusual, but aortic insufficiency is occasionally seem in older patients [12-14]. The aortic root, subjected to high flow over a period of years, dilates which renders the valve incompetent. Turbulence at the valve leaflets may result in marked calcification [31]. One patient in our series had aortic insufficiency.

Neurologic disturbances have been the most frequent complications and causes of death in older patients with cyanotic heart disease. The gradual progression of right ventricular outflow tract obstruction over the years results in increasing polycythemia and hypoxemia [1]. In our series five patients had dizziness but only one had transient paresthesia, along with the low cardiac output state was responsible for 9 futher deaths (9.3%). Also the systemic-pulmonary collaterals may cause a persistently elevated vascular resistance contributing to an intractable low cardiac output state and respiratory failure. One patient (1%) in our series died due to respiratory failure.

It has been suggested that a previous systemic to pulmonary artery anastomosis is associated with a higher surgical mortality at the time of complete repair [4-8-15]. There has been one published report which suggested there is a lower mortality and incidence of complication in the previously shunted group [16]. The general consensus is that a previous shunt does not mitigate against a good result. Our data showed a higher mortality in palliated group although the numbers are too small for conclusion to be drawn.

Richardson and colleagues [17] showed a significant increase in mortality and morbidity in patients with tetralogy of Fallot in which the right ventricular pressure remained high at the end of operation. Yankah and colleagues [4] reported a significantly increased right ventricular end diastolic pressure as well as left ventricular end diastolic pressure when compared to the younger population in other series who underwent repair of tetralogy of. Fallot in childhood. It was found that the left ventricular function was slightly decreased in the postoperative hemodynamics. studies. In our series no correlation between these two factors and outcome was observed.

Improvements in the techinique of cardiopulmonary bypass has greatly reduced the incidence of postoperative coagulopathy and myocardial damage during open heart surgery during recent years, as has use of cold cardioplegia, although in our series differences in the technique of myocardial protection could not be correlated with outcome. These improvements in the technical aspects of circulation support, surgical techniques and the myocardial protection have led to a reduced morbidity and mortality in cardiac surgery. From the analysis thus far it seemed that a prolonged aortic cross clamp time is associated with an increased risk with decreased morbidity at lower temperature. By combining these two factors (figure 1) it was possible to define a safe aortic cross clamp time according to a specific myocardial temperature.

The actuarial analysis observed in this report showed a survival probability of 73% and 93%, at five years, for the patients with tetralogy of Fallot and pulmonary stenosis, respectively. These figures show a significant difference between groups, but it should be emphasized that at 10 years the numbers of patients were low and the survival curves roughly parallel following significantly different early mortality.

RESUMO

De junho de 1971 a junho de 1983, 97 pacientes (idade acima de 16 anos) portadores de cardiopatias congênitas cianóticas foram operados no Brompton Hospital (Londres). A tetralogia de Fallot esteve presente em 40 pacientes, a estenose pulmonar em 30, a dupla via de saída do ventrículo direito em 12, a transposição dos grandes vasos em nove, a atresia tricúspide em três e a atresia, pulmonar em três. A comunicação interatrial, a comunicação interventricular e a estenose pulmonar foram as anomalias associadas mais freqüentemente encontradas. Trinta e um pacientes foram submetidos à cirurgia paliativa prévia. A mortalidade global foi de 12,4%, variando de acordo com a má formação. A síndrome de baixo débito cardíaco contribuiu com 58,3% dos óbitos, particularmente no grupo de pacientes que tiveram tempo de pinçamento aórtico acima de 60 minutos (p < 0,00l) 0 pinçamento aórtico e a temperatura de e , esfriamento do miocárdio foram fatores importantes na determinação da mortalidade. Com um período médio de "seguimento" de cinco anos, o estudo da curva atuarial demonstrou proporção de sobreviventes de 93% e 73% para estenose pulmonar e tetralogia de Fallot, respectivamente. Este estudo demonstrou que a correção cirúrgica da cardiopatia congênita cianótica em pacientes adultos pode ser acompanhada de baixa mortalidade.

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