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SURGICAL TREATEMENT AND OPERATIVE RESULTS FOR PATIENTS WITH TRANSPOSITION OF THE GREAT ARTERIES*

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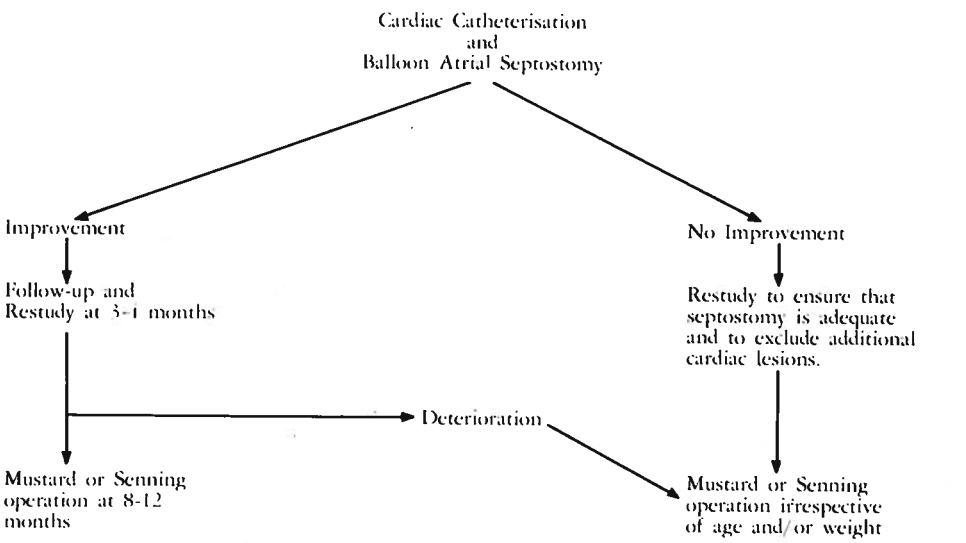
The incidence of congenital heart disease was underestimated by earlier studies, as it is now apparent that it occurs in about 1% of live-born infants. Transposition of the great arteries (TGA) is one of the most frequently occurring forms of cyanotic congenital heart disease, and is the most common form of fatal cyanotic heart disease in the first year of life. According to the New England Regional Infant Cardiac Programme (Nadas et al 1973), 10.8% of 1,564 infants studied with congenital heart disease had TGA. In this paper we report our experience with TGA and outline our present management policy for treatment of various forms of TGA.

TGA+Intact Ventricular Septum

About 50% of the patients with TGA have an intact ventricular septum (IVS) and owe their survival either to an atrial septal defect or a patent foramen ovale. Our policy is to perform Rashkind's balloon atrial septostomy (Rashkind et al 1966) at the initial cardiac catheterisation in every patient with TGA and IVS (Table 1).

Table 1

Transposition of the great arteries with intact ventricular septum
Management Policy



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If the child improves, he is carefully followed up. Recatheterisation and angiocardiogram are performed at the age of 3-4 months. An intraatrial redirection of venous inflow (by Mustard or Senning operation) (Mustard 1964; Senning 1959) is then scheduled for the age of 8-12 months. If the child does not improve following atrial septostomy, recatheterisation is performed to ensure that the septostomy is adequate and also to exclude additional cardiac lesions. If arterial oxigen saturation remains low (less than 20-25 mm Hg) and metabolic acidosis is present, a Mustard or Senning operation is performed without delay, irrespective of age or weight of the patient. The same policy of early intraatrial redirection of venous return is applied for patients who deteriorate after an initial improvement following septostomy.

The alternative form of early management would be surgical septectomy (Blalock and Hanlon 1950). The disadvantage of surgical septectomy is the mortality associated with the procedure and the fact that in some infants even a resection of a large part of the atrial septum does not improve mixing. This is presumably the result of a similar diastolic compliance of the two ventricles, the pressure difference between the two atria being too small to allow bidirectional flow. Surgical septectomy will also cause adhesions which may increase morbidity at subsequent *corrective* operations.

The hospital mortality rate following the Mustard operation for TGA+IVS has been reported from various centres and these results are shown in Table 2. Currently, a 90-95% survival rate can be expected. The risk of the Mustard operation in the first year of life is no higher than the risk in older children, and in our experience this has been even lower (Table 3). In the small group of sick infants who require operation on an emergency basis, the operation carries a high risk (Alfieri et al 1977; Subramanian et al 1973; Stark 1973).

Table 2

Mustard operation for transposition of the great arteries and intact ventricular septum

Author	Year	No.	Died	(%)
Trusler et al	1974	97	11	11%
Oelert et al	1977	21	0	—
Turley et al	1978	54	4	7%
Zavanella et al	1978	44	4	9%
Great Ormond Street	1978	307	27	9%

Table 3

Mustard operation for transposition of the great arteries and intact ventricular septum (First year of life)

Author & Year	No. of Patients	Hospital (No.)	Deaths (%)
Alfieri et al 1977	28	5	18%
Bailey et al 1976	27	0	—
Barrat-Boyes 1973	17	2	12%
Oelert et al 1977	26	0	—
Stark 1977	80	5	6%
Zavanella et al 1978	44	4	8.5%

During recent years, there has been a revival of the Senning operation. Quaegebaur and colleagues (1977) presented an impressive series demonstrating that the high risk of this operation in earlier years was probably due to the selection of patients rather than the operation itself. Their results, as well as those of others (Table 4) compare favourably with the best series of Mustard operations. One advantage of the Senning operation is that only living cardiac tissue is used for repair, increasing the effectiveness of the atrial contraction, compared with other techniques, Another reported feature of the Senning operation has been the low incidence of dysrhythmias. Our own experience with the Senning operation is limited to 19 children. Currently, we are using both techniques and plan to restudy and compare both groups of patients.

Table 4

Senning operation for T.G.A.

Author	Number	Died
Quaegebeur et al 1977	20	2
Parenzan et al 1978	24	0
Pacifico 1978	8	1
Stark 1979	19	2
Total	71	5

TGA+Ventricular Septal Defect

A ventricular septal defect (VSD) occurs in about 25% of patients with TGA. Pulmonary vascular disease develops early in patients with TGA+VSD (Newfeld et al 1974; Stark et al 1970; Wagenwoort 1968) and, therefore, operation should be performed at an earlier age than in patients with simple TGA.

Our current management policy for patients with TGA+VSD is illustrated in Table 5. In all infants with TGA+VSD, balloon septostomy is performed at the time of initial catheterisation. If, after septostomy, congestive heart failure is controlled, the child is restudied at three months of age. Patients with a pulmonary artery pressure less than 1/3 systemic are treated in the same way as patients with TGA+IVS. If, however, the pulmonary artery pressure is more than 1/3 systemic, an intraatrial redirection of venous return (Mustard or Senning operation) and closure of the VSD is performed before the age of six months.

Patients who remain in severe heart failure following septostomy, constitute a difficult group whose treatment is still a matter for discussion. Pulmonary artery banding or early correction are the possible alternatives. The results of correction during the first months of life are unfavourable, when the mortality rate of pulmonary artery banding is about 12% (Stark et al 1970). We, therefore, consider pulmonary artery banding in the first 2-3 months of life. The subsequent management of patients with a banded pulmonary artery is controversial. Intraatrial redirection of the venous flow (Mustard or Senning operation) plus closure of the VSD and debanding of the pulmonary artery can be done at the age of 12-18 months. Alternatively, a Rastelli operation (Rastelli et al 1969) can be performed later.

The operative mortality for patients with TGA+VSD is higher than for simple TGA (Table 6). The risk depends partly upon the position of the VSD and its relationship to the tricuspid and mitral valve, and also on the degree of pulmonary vascular disease.

Table 5

Management policy for transposition of great arteries+ventricular septal defect

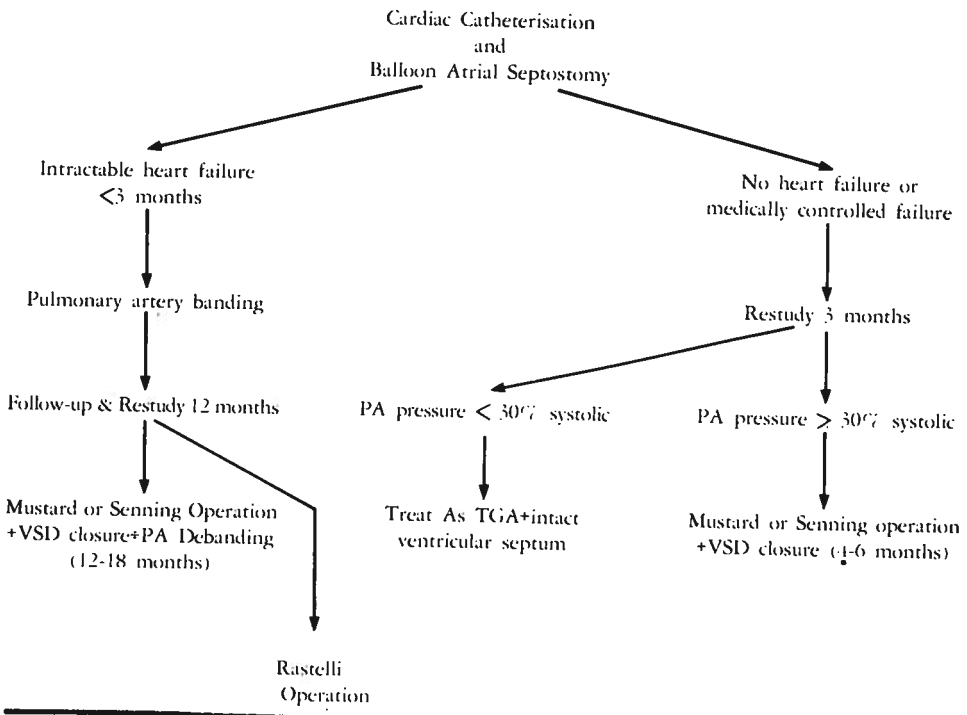


Table 6

Transposition of the great arteries and ventricular septal defect the Mustard operation and ventricular septal defect closure

Author & Year	Number	Hosp. Deaths	Mortality
Castaneda et al 1976	25	2	8%
Champsaur et al 1973	22	10	45%
Danielson et al 1972	29	16	55%
Mori et al 1976	23	9	39%
Oelert et al 1977	13	0	0
Stark et al 1977	95	26	26%

TGA+VSD+Left Ventricular Outflow Tract Obstruction

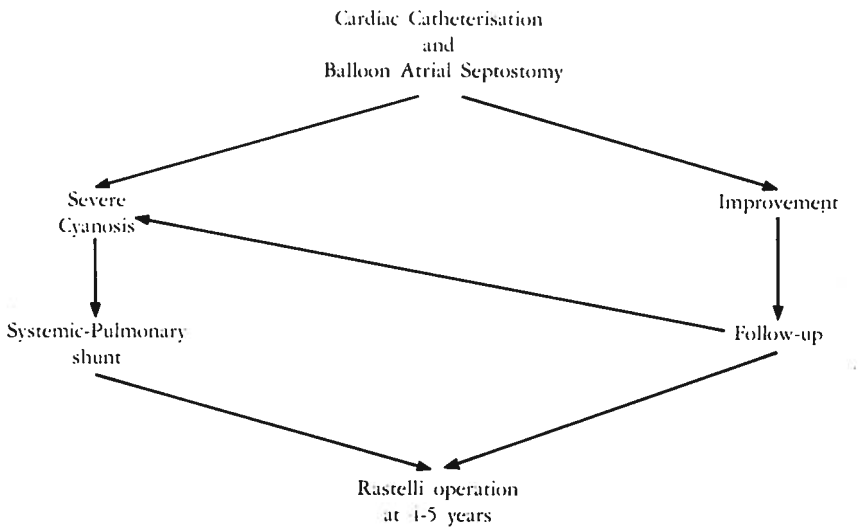
The combination of TGA+VSD and left ventricular outflow tract obstruction (LVOTO) remains rare, occurring in only 0.67% of the patients with congenital heart

disease reviewed by Keith and colleagues in 1978. However, significant obstruction occurred in 31% of their patients with TGA + VSD, an incidence which agrees with figures reported by other centres (Van Praagh et al 1977). The causes of obstructions are multiple and may occur at various levels. Fibrous tunnel between the ventricular septal defect and the pulmonary valve ring, fibrous diaphragm, hypertrophied intraventricular septum, abnormal insertion and/or abnormal movement of the mitral valve, accessory fibrous tissue associated with the mitral valve, aneurysm of the membranous ventricular septum or tricuspid valve tissue and, rarely, pulmonary valve stenosis have all been described as causes of LVOTO (shrivastava et al 1976; Silove et al 1973; Vidne et al 1976). The surgical relief of some of these is difficult (fibromuscular tunnel), or impossible (abnormalities of the mitral valve) and can result in irreversible damage to the adjacent structures such as the bundle of His or the mitral valve. Early attempts to combine the Mustard procedure with closure of the VSD and resection of LVOTO were met with disappointing results (Danielson et al 1971; Breckenridge et al 1972). In 1969, Rastelli and his associates developed an anatomical correction for this combination of lesions (Rastelli et al 1969). The principle of the operation consists of redirection of blood at ventricular level through the VSD in such a way that the left ventricle empties into the aorta. The pulmonary artery is then ligated, or divided and oversewn and the continuity between the right ventricle and pulmonary artery reestablished with an external valved conduit. Both physiological and anatomical correction is achieved since the ventricles and great arteries become concordant.

Our current approach for patients with TGA, VSD and LVOTO is illustrated in Table 7.

Table 7

Management policy for patients with transposition of the great arteries, ventricular septal defect and left ventricular outflow tract obstruction



Balloon atrial septostomy is performed at the time of cardiac catheterisation as in all other infants with TGA. Should cyanosis or polycythaemia increase, it is our policy to perform a systemic to pulmonary shunt. Besides carrying a low risk, the shunt enables us to delay the Rastelli procedure until the patient is 4-5 years old, when an adult size conduit can be used. Our first choice is the Blalock-Taussig anastomosis on the side opposite to the side of the aortic arch. Using magnification, microsurgical techniques and mobilization of the innominate and carotid arteries, the operation can be performed even in the first day or life. In patients in whom the anatomy is not suitable for Blalock-Taussig shunt, one can either perform a Waterston anastomosis (Waterston 1962) or use a Gore-tex prosthetic graft. Our experience with the use of the Gore-tex prosthetic graft has been encouraging. In this operation, the flow is controlled by the diameter of the subclavian artery, thus a 5 or 6mm graft can be inserted irrespective of the age of the patient.

The results of the Rastelli operation as reported from several centres are shown on Table 8. There has been a significant reduction in mortality rates during the last few years. Early deaths were mainly related to unfavourable anatomic features, conduit compression and superimposed sepsis. Considerable late morbidity and mortality has been reported following the Rastelli operation. Marcelletti et al (1976) reported a late mortality of 8.5%. In our series, seven children died in a group of 37 early survivors (late mortality rate of 17.1%). The main factors contributing to late morbidity and mortality were residual ventricular septal defects and infections.

Table 8

The Rastelli operation for transposition of the great arteries ventricular septal defect and left ventricular outflow tract obstruction

Author	Year	No	Hospital (No.)	Deaths (%)
Marcelletti et al	1968-72	34	9	26%
Marcelleti et al	1972-75	25	2	8%
Great Ormond Street	1971-76	28	4	14%
Great Ormond Street	1977-78	13	0	0
Heck et al	1972-77	9	0	0
Norwood et al	1972-76	7	0	0

TGA+IVS+LVOTO

The causes of LVOTO in patients with intact ventricular septum are the same as those described for patients with TGA, VSD and LVOTO. However, a pressure gradient between the left ventricle and pulmonary artery in a patient with TGA and IVS does not necessarily mean that an anatomical obstruction is present, as the gradient may be due to an increased blood flow across the left ventricular outflow tract. Flow gradients up to 100mmHg were abolished, or significantly reduced; following the Mustard or Senning operation.

Our current policy for patients with TGA, IVS and left ventricle to pulmonary artery gradients depends on the underlying cause of the obstruction (Table 9). If no anatomical reason can be identified on angiocardiography, a Mustard or Senning operation is performed using the same criteria as that outlined for patients with simple TGA. If an anatomic obstruction is diagnosed, the approach will depend on its anatomical

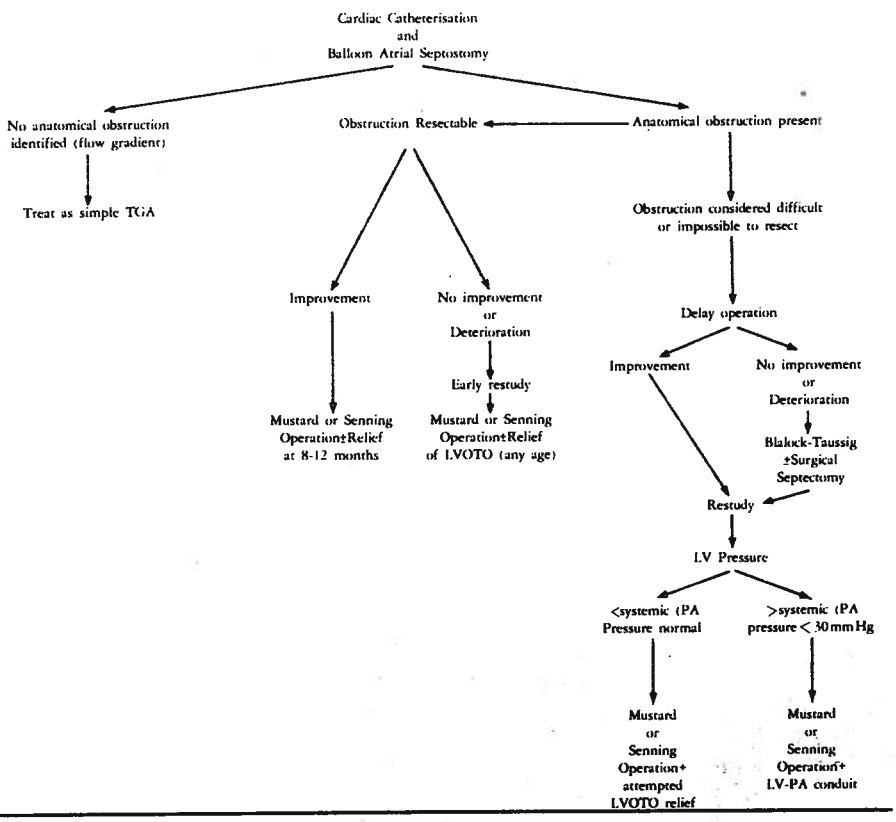
type. If complete or near complete relief of the obstruction is to be expected at operation (fibrous shelf, aneurysm of the membranous intraventricular septum), we follow the same criteria used for patients with simple TGA. At the time of inflow correction, the obstruction is resected through a pulmonary arteriotomy or, occasionally, through a short left ventriculotomy.

If surgical relief of LVOTO is likely to difficult (fibromuscular tunnel) or impossible (abnormalities of the mitral valve) the operation is delayed. Should cyanosis and/or polycythaemia increase, a systemic to pulmonary shunt is performed. If there is any doubt about the efficiency of a balloon atrial septostomy, surgical septectomy may be required at the time of the shunt operation, as it is important that an adequate intraatrial communication is present in such patients.

If relief of the obstruction seems impossible, or if after attempted relief the pressure in the left ventricle remains supra-systemic, with a pulmonary artery pressure less than 30 mm Hg, a left ventricle to pulmonary artery conduit (McGoon 1976; Singh et al 1976) is considered. If the left ventricular and pulmonary pressures do not meet this criteria, an inflow correction alone is performed. In our experience, we have observed that residual pressure gradients were well tolerated clinically up to 12 years after operation, and at repeat cardiac catheterisation the gradients usually did not increase.

Table 9

Management policy for transposition of the great arteries, intact ventricular septum and left ventricular outflow tract obstruction



To operative mortality of patients with TGA, IVS and LVOTO is higher than in simple TGA (Idriss et al 1977). Between 1971 and 1978, we operated upon 26 patients with TGA, IVS and LVOTO with four deaths (15% mortality rate). Between 1974 and 1979, nine children with TGA, IVS and LVOTO had Mustard or Senning operations peformed, and in addition the insertion of a left ventricle to pulmonary artery conduit. There were three early and one late deaths.

TGA + Pulmonary Vascular Obstructive Disease

Patients with TGA are known to develop pulmonary vascular obstructive disease (PVOD) at an early age (Wagenwoort et al 1968). The coexistence of a large ventricular septal defect and/or patent ductus arteriosus accelerates the development of PVOD, and a substantial number of patients with these associated lesions will have a fixed high pulmonary arteriolar resistance by the end of the first year of life (Newfeld et al 1974). Although much less common, PVOD does occur even in patients with TGA and IVS during the first year of life (Lakier et al 1975).

In earlier years, the Mustard operation in patients with severely elevated pulmonary arteriolar resistance (Rp) resulted in a high mortality, or these patients were considered inoperable (Mair et al 1971). Further progress of PVO has been observed after the Mustard operation (Mair et al 1976).

The fate of children with TGA, VSD and PVOD was much improved by the concept of the palliative Mustard operation introduced by Lindesmith in 1972 (Lindesmith et al 1972). The intraatrial redirection of venous inflow is performed, but the VSD is left open. The principle of the palliative operation can be extended to patients with TGA, IVS and PVOD, as reported by us (Stark et al 1976). A Mustard operation is performed and a ventricular septal defect created in the apical portion of the interventricular septum.

Our present policy for patients with TGA and PVOD is as follows: If the Rp is more than 8 units. m^2, a palliative Mustard operation is performed (if there is no VSD, an apical VSD is created). If the Rp is less than 6 units. m^2, a Mustard or Senning operation is performed and the VSD closed. Patients with Rp between 6 and 8 units. m^2 are assessed on an individual basis. The age of the patient, the appearance of the pulmonary vessels on angiocardiography and lung biopsy results are factors that can affect the decision on whether or not to close the VSD.

The results of palliative operations have been good. Not only has the hospital mortality been low (Table 10) but the clinical improvement in most patients has been

Table 10

Palliative Mustard operation for transposition of the great arteries with severe pulmonary vascular obstructive disease

Author	No. of Patients	Mortality (No.)	(%)
Lindesmith 1975	10	0	0
Bernhard 1976	3	0	0
Mair 1976	8	0	0
Oelert 1977	8	1	12%
Great Ormond Street 1978	38*	3	8%

* Includes 6 patients with TGA+IVS.

remarkable. To date, our own experience with the palliative Mustard operation consists of 38 patients, 32 of whom had a VSD and six had IVS. One patient out of 32 with TGA, VSD and PVOD died (3%). There were two deaths among six patients with TGA+IVS who had a VSD created.

Good results achieved with the palliative Mustard procedure should, however, not diminish our efforts to diagnose and treat all children with TGA early, i. e., before severe pulmonary vascular disease develops.

Complications and Late Results of Mustard Operation

Several complications have been reported after Mustard operation. These include systemic venous obstruction, pulmonary venous obstruction, tricuspid valve incompetence, arrhythmias and right ventricular malfunction (Mazzei et al 1971; Stark et al 1972; Tynan et al 1972; El-Said et al 1972; Jarmakani et al 1974).

Vena caval pathway obstruction, usually superior vena cava, has been repeatedly reported. In our own series, obstruction of the vena caval pathway was much more common in patients in whom a Dacron patch was used (30%) compared with those in whom pericardium was used (5%). Obstructions of the inferior vena cava is very unusual, especially if pericardium has been used for the atrial partition.

In the last few years, it was recognised that obstructive complications were mainly due to faulty operative techniques. Some technical modifications were introduced, and since we started using pericardium cut into the trouser-shaped patch, as described by Brom (1975), the incidence of obstructive complications have decreased sharply. During the period 1976-1977, we performed 69 Mustard operations in our unit. Of 52 hospital survivors, only two developed obstruction and were successfully reoperated. We prefer the right thoracotomy approach through the 5th intercostal space when performing an operative revision for systemic or pulmonary venous obstruction (Szarnicki et al 1978). The same approach is also used for reoperations for tricuspid incompetence or residual VSD.

Tricuspid valve regurgitation is another reported complication after the Mustard operation. In 1972, 17 of 173 survivors of the Mustard operation performed in our unit had some degree of tricuspid incompetence, but only in three patients was it judged to be severe. Two of these three patients had abnormal myxomatous valve cusps. In patients with TGA and VSD, retraction or detachment of the septal cusp of the tricuspid valve may be a contributing factor. We are very careful when retracting the valve and never detach it.

Arrhythmias following Mustard operation have long been recognised as an important factor in postoperative morbidity and mortality. (El-Said et al 1972; Isaacson et al 1972; Breckenridge et al 1972). It is now considered that only 24 hour monitoring can evaluate such arrhytmias, and even then it may be difficult as some of the patients may possibly have arrhythmias preoperatively. Modifications in operative techniques were introduced by many surgeons in order to reduce the incidence of postoperative arrhythmias. Direct damage to the atrioventricular node should obviously be avoided. Careful use of intracardiac suction, especially around the coronary sinus is recommended, as well as placement of the superior vena cava cannula well away from the sinus node. Injury to the sinus node artery should also be avoided.

Deterioration in right ventricular function has been reported in some patients after the Mustard operation (Jarmakani et al 1974; Hagler et al 1977). It is still unknown what proportion of patients with TGA are likely to develop this complication and, if so, the degree of severity.

Long-Term Results

Reports on the long-term results of the Mustard operation vary from pessimistic (Hagler et al 1977), to optimistic (Champsaur et al 1973). The late results at The Hospital for Sick Children, reviewed in 1976 (Schmitz et al 1976) showed that there were 25 late deaths (15% in a group of 164 patients surviving the operation peformed during the period 1965-1971). 78% of the patients reviewed were normally developed, attended normal school and enjoyed the same activities as their peers.

To evaluate the exercise tolerance of patients after Mustard operation, a study was conducted in our unit (Weller et al 1978) on 44 asymptomatic patients operated upon between 1965 and 1971. Only patients who had little or no subjective exercise limitation and were living normal lives were studied. The study showed that many of these patients had achieved a normal working capacity, but as a goup there was a statistically significat reduction when compared healthy children ($P<0.001$). There was no significant difference between the group of patients who had Mustard operation in infancy and those operated upon later.

Future Trends

Although intraatrial redirection of venous return (Mustard or Senning operation) has enhanced long-term survival rates and functional capacity, it lacks the achievement of a true anatomical correction. As previously stated, the long-term results are still a matter for discussion and analysis, and serious doubts exist as to the capability of the right ventricle and tricuspid valve to sustain a systemic pressure for a long time. Becu et al (1975) reported the presence of congenitally malformed right ventricular musculature and a high incidence of abnormalities of the tricuspid valve mechanism in specimens with TGA. One possible answer to this problem may be retransposition of the great arteries with their coronary suplly to the appropriate ventricles. In 1975, Jatene and his colleagues achieved the first successful survival by great artery reversal, modifying a technique previously described by Baffes and colleagues in 1961. Since then, the same technique has been occasionally applied to patients with TGA and IVS (Mauck et al 1977). The mortality rate associated with this operation remains high (Jatene et al 1976). An interesting approach has been suggested and practised by Yacoub. Patients with TGA and IVS undergo banding of the pulmonary artery and a Jatene operation is performed a few months later (Yacoub 1977). If this operation can be performed with a lower risk and the long-term results prove acceptable, it may become the operation of choice.

Another possibility could be an anatomically corrective operation described independently by Kaye, Damus and Stansel in 1975. The pulmonary artery is transected and its proximal portion anastomosed end to side to the ascending aorta. The continuity between the right ventricle and the distal pulmonary artery is then reestablished by the insertion of an extracardiac valved conduit. This approach is somewhat less appealing, as it requires the insertion of a conduit in every patient with TGA.

RESUMO

TRATAMENTO CIRÚRGICO E RESULTADOS NA TRANSPOSIÇÃO DOS GRANDES VASOS

Pensava-se que a incidência de cardiopatias congénitas era baixa mas sabe-se actualmente que ocorre em cerca de 1% dos nados-vivos.

A transposição dos grandes vasos (TGV) é uma das mais frequentes cardiopatias congénitas cianosantes e é a mais comum entre as que são mortais no 1.º ano de vida.

Neste artigo descrevemos a nossa experiência com TGV e apresentamos a forma de tratamento que preferimos nas suas diversas variantes anatómicas.

TGV com septo inter ventricular intacto:

Constituem cerca de 50% dos doentes com TGV, e devem a sua sobrevivência a uma comunicação inter auricular ou a um «foramen ovale» patente. Nelas se põe a indicação terapêutica primária de septostomia de Rashkind ao mesmo tempo que se realiza o primeiro cateterismo cardíaco. Se o resultado obtido é bom a criança é seguida até à idade de 3 ou 4 meses, altura em que se repete o cateterismo. Entre os 8 e os 12 meses de idade realiza-se então uma intervenção cirúrgica venosa intra cardíaca.

Descrevem-se as alternativas de terapêutica cirúrgica nestes casos.

Mencionam-se os resultados e a mortalidade da operação de Mustard descritos em vários Centros.

Recentemente a operação de Senning tem de novo passado a usar-se e os resultados obtidos são favoravelmente comparáveis com os da operação de Mustard.

TGV+Comunicação inter-ventricular (CIV):

Em 25% dos casos de TGV existe uma CIV. Nestes doentes aparece precocemente **doença vascular pulmonar e por isso a operação correctiva deve ser realizada mais cedo. A mortalidade operatória em doentes com TGV+CIV é mais elevada do que nas TGV simples.**

TGV+CIV+Obstrução da câmara de saída do ventrículo esquerdo:

Este conjunto de malformações ocorre em 0,67% dos doentes com cardiopatias congénitas. No entanto, em 31% destes doentes a obstrução da câmara de saída do ventrículo esquerdo é significativa. As tentativas de ressecção da estenose ventricular esquerda sempre deram mortalidade alta até que, em 1969, Rastelli et al. descreveram a correcção anatómica deste conjunto de lesões, dirigindo o sangue do ventrículo esquerdo para a aorta através da CIV e ligando o ventrículo direito à artéria pulmonar por intermédio de um conduto valvulado.

TGV+Doença vascular pulmonar:

Depois de se ter considerado esta associação como simónimo de inoperabilidade, passaram a obter-se bons resultados operatórios com a técnica de operação de Mustard paliativa, descrita por Lindesmith, em que não se faz o encerramento da CIV.

Descrevem-se em seguida os resultados a distância das correcções cirúrgicas das TGV e revêem-se as possibilidades futuras de técnicas descritas recentemente como a operação ou de *SWITCH* arterial.

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