Complications after Senning Operation for TGA with and Without VSD

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=국문초록=

대혈관전위증에서 Senning 수술후 합병증에 관한 임상적 고찰

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단순 대혈관 전위증 및 심실중격 결손이 동반된 복잡 대혈관 전위증에 대해 Oklahoma University Health Sciences Center에서 Senning 술식을 적용했던 60명의 환아를 분석하였다. 41명의 단순 대혈관 전위증(1군)과 19명의 복잡 대혈관 전위증(2군)에서 전체 수술 사망율은 20%로 1군에서 4.9%, 2군 52.6% 였으며, 그들을 술후 추적 관찰한 결과 13년 간의 생존율은 1군 95%, 2군 42% 이었다. 술전의 높은 좌심실압 및 폐동맥압이 사망율에 영향을 미치고 있으며($p<0.05$), 술후 다양한 합병증을 관찰할 수 있었던 바, 부정맥은 1군에서 28.2%, 2군에서 55.6%로서 13년 간 부정맥이 발생치 않는 율은 66% 로 1군 71%, 2 군 44% 이었다. 술후 좌심실 유출로 협착은 31.2% 로 15명에서 관찰되었으나 그중 3명에서 재수술이 필요했었다. 폐정맥 경로 협착은 3명에서 관찰되었던 바 모두 1군이었고 그중 1명에서 재수술을 시행했으나(13년 간의 폐정맥 경로 협착이 없는 율 89%), 체정맥 경로의 협착은 관찰되지 않았다. 전체의 재수술율은 14.6% 로서 잔존성 심실중격 결손이 3례, 좌심실 유출로 협착이 3, 폐정맥 경로 협착이 1례, 심방 baffle에서의 누출 3례에서 있었다.

이러한 높은 합병증의 발생 및 2군에서의 높은 수술 사망율로 인해 현재는 Senning 술식을 이용치 않고 수년전 부터는 모든 환자에서 Jatene 술식을 적용하고 있다.

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중심 단어 : Senning 술식, 대혈관 전위증, 술후 합병증

INTRODUCTION

For the treatment of transposition of the great arteries (TGA), we had performed several kinds of corrective surgeries, physiological or anatomical, and encountered their intra-and postoperative problems. Until the Jatene operation became a routine operative modality in neonates, we used to use the atrial switch operations and experienced their usual outcomes and drawbacks. The notorious problems after the Senning operation are postoperative arrhythmia after extensive intraatrial procedure, obstruction of venous pathway, the baffle leakage and late right ventricular failure as a systemic ventricle[1, 2]. Especially when combine with ventricular septal defects (VSD), the early and late mortality and complication rate are much higher than simple TGA, with published mortality ranged from approximately 10 to 60%[3, 4].

We had applied the Senning procedure since 1979, but because of their frequent postoperative complications and high mortality for the coexisting VSD and improved result of the Jatene procedure in neonate, we did not perform

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Table 1. Age distribution of the patients for Senning procedure

age	patients' numbers	accumulated numbers
less than 1 week	24	24
− 6 weeks	16	40
− 1 year	12	52
over 1 year	8	60

that kind of atrial rerouting procedure any more since couple of years ago. Now we review the outcomes and the complications after Senning operation for TGA with or without VSD undergone at our institution and analyse their prevalence and related factors.

PATIENTS AND METHOD

From 1979 through March 1990, total 61 patients had undergone Senning operation for simple and complex TGA at Oklahoma University Health Sciences Center. Except one missing case we could review all of these patients with medical records, echocardiograms, cardiac catheterization data and operation records, and investigated the results of operations and complications.

There were 45 male(75%) and 15 female patients(25%), 41 were simple TGA(group I) and 19 had significant VSD(group II). The age of these 60 patients at operation ranged from 1 day to 8 year old(mean 7.3 ± 15.2 months), 24 were less than 1 week of age, 40 were under 6 weeks of age and only 8 patients were older than 1 year old(Table 1), and body weight from 2.7 to 21.5 kg(mean 5.84 ± 3.35 kg). Associated lesions except VSD were patent ductus arteriosus in 22 patients(36.7%) and mild to moderate left ventricular outflow tract obstruction(LVOTO) in 9 patients(15%). In group I, all patients had undergone previous Balloon Atrial Septostomy(BAS) and 4 had Blalock-Hanlon atrial septectomy before Senning procedure. In group II 14 had BAS, 4 Blalock-Hanlon septectomy, 1 pulmonary arterial banding(PAB) and 1 modified Blalock-Taussig shunt operation, but 5 patients didn't get any procedure before Senning operation.

For analysis of the data of echocardiogram we measured the diastolic dimension of right and left ventricular cavity, left ventricular posterior wall thickness and ejection fraction(EF), and corrected with the diameter of their own descending aorta at diaphragmatic level for standardization[5, 6]. We also studied about the systolic, diastolic pressure of right and left ventricle(RVP and LVP), pulmonary artery(PAP) and aorta(SAP). We investigated the operation record and bypass sheets to know the variable modifications of operation technique, total bypass time, total circulatory arrest period and aortic cross clamp time.

The mean follow up period was 7 years, ranged from 1 year to 13. 5 years. During this postoperative follow-up period,the patients had been checked clinically, with the electrocardiogram(ECG) and echocardiogram regularly and 24 hours Holter monitor recording, cardiac catheterization, if indicated, to detect the complications like arrhythmia, pulmonary and systemic venous pathway obstruction (PVO and SVO), atrioventricular(AV) valve regurgitation, right and left ventricular outflow tract obstruction, and to determine the necessity of reoperation.

Early operative mortality was defined as death within 1 month of the initial operation and late mortality was death at 1 month or more after surgery.

All these values were expressed in terms of mean ± standard deviation and these data were analysed statistically with student T-test and Fisher's exact test and consider the significance if the p-value was less than 0.05. To identifiy the operative risk factors we applied the multivariate analysis.

RESULTS

The patients profile was summarized as Table 2. Those showed several statistically significant difference between two group: In group II, they had less hemoglobin($p<0.05$) and high arterial oxygen saturation, higher LVP and PAP ($p<0.01$), and larger LV diastolic dimension(LVd)($p<0.05$), which mean more mixing and shunting of the systemic and pulmonic circulation at ventricular level.

Mortality

Overall operative mortality of Senning operation for these patients was 20%(12/60), but for Group I, the simple TGA, only 2 patients died among 41(4.9%). One died

Table 2. Patients profile for simple TGA(group I) and TGA with VSD(group II)

	group I	group II	p-value
age (months)	5.44 ± 9.52	11.42 ± 22.92	ns
body weight (kg)	5.66 ± 2.47	6.22 ± 4.81	ns
hemoglobin (gm%)	17.81 ± 2.81	16.07 ± 2.79	$p<0.05$
O_2 saturation (%)	54.18 ± 18.72	63.06 ± 13.33	ns
aorta pressure (mmHg)	84.10 ± 20.01	82.94 ± 16.87	ns
PAP (mmHg)	30.75 ± 11.87	57.33 ± 51.56	$p<0.01$
LV pressure (mmHg)	52.77 ± 20.04	76.25 ± 16.94	$p<0.01$
RV pressure (mmHg)	80.65 ± 13.62	88.92 ± 8.06	ns
LVd dimension (mm)	23.24 ± 6.56	28.93 ± 6.29	$p<0.05$
RVd dimension (mm)	19.71 ± 5.2	19.69 ± 5.43	ns
LVpw thickness (mm)	4.83 ± 0.9	4.84 ± 1.50	ns
EF (%)	63.77 ± 14.3	59.60 ± 9.73	ns
Pump time (min.)	94.50 ± 28.03	115.27 ± 60.70	$p<0.05$
Aortic X time (min.)	49.67 ± 16.52	64.91 ± 19.01	ns
TCA time (min.)	51.78 ± 13.18	57.58 ± 13.93	ns
Postop. LVP (mmHg)	44.00 ± 16.90	37.50 ± 25.98	ns
RVP (mmHg)	103.60 ± 28.36	108.33 ± 12.58	ns
LVd (mm)	25.57 ± 6.04	25.71 ± 9.64	ns
RVd (mm)	22.14 ± 8.70	23.29 ± 8.86	ns
LVpw (mm)	4.35 ± 1.50	5.00 ± 1.55	ns
EF (%)	59.22 ± 8.95	58.33 ± 10.63	ns

TGA : transposition of great artery, VSD : ventricular septal defect, PAP : systolic pulmonary artery pressure, LV : left ventricle, RV : right ventricle, LVd : diastolic LV, RVd : diastolic RV, LVpw : LV posterior wall, EF : ejection fraction, X : cross-clamp, TCA : total circulatory arrest

immediate postoperatively due to the myocardial failure with low ouput and another patient, 1 day old, died at postoperative 11th day due to the hypoxic cerebral damage. In group II, 10 patients died(52.6%) caused by ventricular failure, endocardial fibrosis, small left ventricular cavity, acute renal failure and ventilator related accident. There was one late death in group II who got reoperation for VSD leakage and LVOTO at 2 years after initial operation, who died during the third reoperation for relief of pulmonary stenosis on 2 years and 8 months after first operation. Age related mortality of group I and group II were on Table 3. In group I only 2 patient died in their 1 and 3 days of age. In group II less than 1 week old showed the lowest mortality, and oldest patient(8 years old) died with preexisting severe pulmonary hypertension (155/80). The actuarial survival at 13 years were 78% in total patients, 95% in group I, 42% in group II(Fig. 1). When we reviewed the preoperative catheterization data, among the death patients the preoperative LVP were much higher than survival group(82.5 ± 7.7 mmHg vs. 56.0 ± 3.5 mmHg, $p<0.05$), which might be related with high PAP (77.3 ± 50.01 mmHg vs. 33.57 ± 12.26 mmHg, $p<0.05$) and pulmonary vascular resistance(PVR). Compared the variables between the survival and mortality group, we could not find the definite discrepancy in the preoperative data like age, body weight, hemoglobin, oxygen saturation, echocardiogram and cardiac catheterization data and also in the cardiopulmonary bypass(CPB) data. The longer bypass time in death group(134.8 ± 27.8 min.) than the survival group(94.1 ± 5.1 min) did not have statistical meaning, but longer bypass time means the complexity of sur-

Table 3. Age related mortality in group I and group II

	group I	group II	cumulative total
less than 1 week	2/17 (11.8%)	2/7 (28.6%)	4/24 (16.7%)
− 6 weeks	0/14	2/2 (100%)	6/40 (15%)
− 1 year	0/7	4/5 (80%)	10/52 (19.2%)
over 1 year	0/3	2/5 (40%)	12/60 (20%)
	2/41 (4.9%)	10/19 (52.6%)	

Table 4. Comparison of survival and nonsurvival patients

	survival group	nonsurvival group	p-value
age (months)	6.05 ± 9.25	12.47 ± 13.95	ns
body weight (kg)	5.80 ± 0.38	6.02 ± 1.58	ns
hemoglobin (gm %)	17.6 ± 0.4	16.1 ± 1.1	ns
O_2 saturation (%)	58.4 ± 2.8	52.1 ± 5.6	ns
Aorta pressure (mmHg)	86.9 ± 21.02	78.2 ± 15.76	ns
PAP (mmHg)	33.57 ± 12.76	77.3 ± 50.01	$p<0.05$
LVP (mmHg)	56.0 ± 3.5	82.5 ± 7.7	$p<0.01$
RVP (mmHg)	80.1 ± 2.5	104.0 ± 15.6	ns
LVd (mm)	17.5 ± 1.1	18.1 ± 1.3	ns
RVd (mm)	14.1 ± 0.8	11.7 ± 1.5	ns
LVpw (mm)	3.5 ± 0.2	3.0 ± 0.2	ns
EF (%)	62.5 ± 3.1	59.5 ± 4.9	ns
pump time (min.)	94.1 ± 5.1	134.8 ± 27.8	ns
aortic X time (min.)	52.0 ± 3.0	62.4 ± 7.9	ns
TCA time (min.)	52.3 ± 2.9	58.3 ± 4.6	ns
with VSD	2/41 (4.9 %)	10/19 (52.6 %)	$p<0.01$
atrial patch	6/40 (15 %)	6/20 (30 %)	ns

PAP : pulmonary arterial pressure, LVP : LV pressure, RVP : RV pressure, LVd : LV diastolic dimension, RVd : RV diastolic dimension, LVpw : LV posterior wall thickness, EF : ejection fraction, X : cross-clamp, TCA : total circulatory arrest

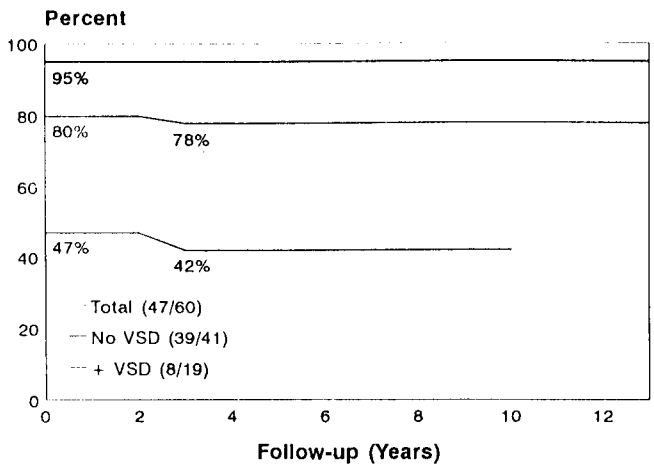

Fig. 1. The actuarial survival curve after Senning operation for simple TGA and TGA with VSD

gery and can affect the mortality (Table 4).

Arrhythmia

They developed junctional rhythm in 7 patients, one of whom might be indicated for permanent pacemaker implantation but still observed, first degree AV block in 3 and one sick sinus syndrome. A complete AV block developed in one patient who died at first postoperative day and another patient who got complete AV block needed permanent pacemaker implantation immediate postoperatively. One patient had got a pacemaker insertion in his postoperative fourth year because of the aggrevating sinus bradycardia. The total incidence of rhythm disturbance in this series was 33.3 % (16/48) : 28.2 % (11/39) in group I, 55.6 % (5/9) in group II, but there was no statistically significant difference. The actuarial rate of freedom from arrhythmia was 66 % in 13 years postoperatively (71 % in group I, 44 % in group II) (Fig. 2). After the BAS or Blalock-Hanlon septectomy, the native atrial septum might be insufficient for rerouting the new right atrial pathway. The usage of additional prosthesis or left atrial appendage for reconstruction of part of the new atrial roofing did not affect the incidence of arryhthmia (29.4 % vs. 42.9 %). Arrhythmia did not show any relationship with the postoperative AV valve regurgitation (42.9 % for AV valve regurgitation vs. 29.4 % for non-regurgitation). Among the variables in CPB data, increased aortic cross clamp time had a tendency to cause more frequent arrhythmia (60.0 ± 4.2 min. in arrhythmic group vs. 47.6 ± 3.8 min. in non-arrhythmic group, $p<0.05$).

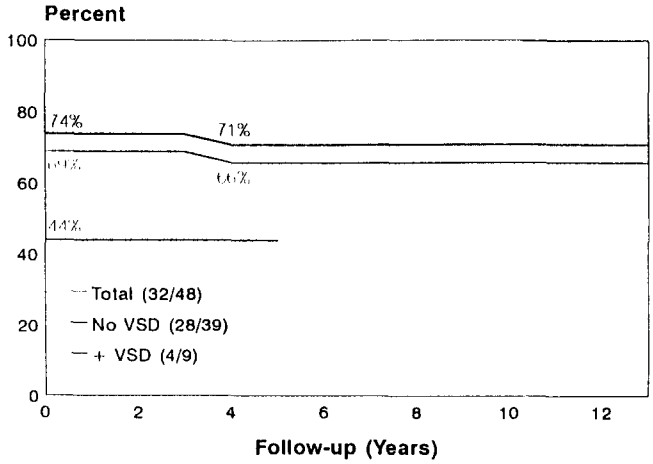

Fig. 2. The estimated Freedom from arrythmia at 13 years after Senning operation for simple TGA and TGA with VSD

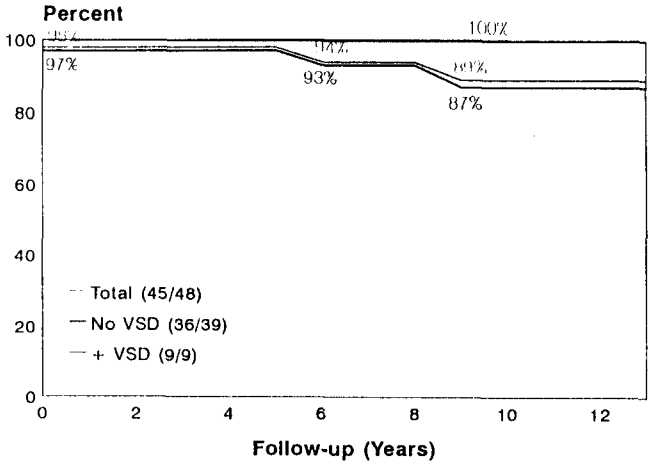

Fig. 3. The estimated Freedom from pulmonary venous pathway obstruction at 13 years after Senning operation for simple TGA and TGA with VSD

LV outflow tract obstruction

The degree of LVOTO could be categorized as mild, moderate and severe according to the gradient between LVP and peak systolic PAP less than 25 mmHg, between 25 and 45 mmHg and more than 45 mmHg. There were 9 mild, 3 moderate and 3 severe LVOTO, and all the severe patients had got reoperation for relief of pulmonary stenosis in their second and eighth postoperative years. The overall incidence of LVOTO was 31.3% (15/48) : 25.6% (10/39) in group I, 55.6% (5/9) in group II, and there was no significant difference between two groups. But we have to cosider the preoperative mild to moderate LVOTO in 9 patients, who were tried to be corrected during operation. We couldn't find any factors influencing LVOTO in our studied variables.

Pulmonary venous pathway obstruction

PVO was defined as a gradient between the simultaneous mean pulmonary capillary wedge pressure and RV end diastolic pressure of 5 mmHg or more, or clinical sign, characteristic angiographic appearance and/or elevated PAP[7]. The overall incidence of PVO was 6.3% (3/48) : in group I 7.7% (3/39), group II 0% (Fig. 3). When using the additional prosthesis for reconstruction of part of new atrial pathway, the obstruction were occurred 3 patients among 34 (8.8%). In these three patients, only one patient required the reoperation.

Systemic venous pathway obstruction

We diagnosed SVO when the gradient between the mean superior vena caval pressure or inferior vena caval pressure, and right atrial pressure is more than 3 mmHg and/or angiographic evidence of SVC or IVC obstruction[7]. We could not find any SVO cases in our series.

Atrioventricular valve regurgitation

The patients were evaluated with two dimensional color Doppler echocardiography and/or cardiac catheterization, if any significant evidence. There occurred AV valve regurgitation in 14 patients among 48 (29.2%) : in group I 28.9% (11/38), group II 33.3% (3/9). For mitral valve there were 5 regurgitation (4 in group I, 1 in group II), 11 for tricuspid valve (9 in group I, 2 group II) and 2 for both valve concomitantly, but these regurgitation did not require any surgical intervention until now. Those mean 14.6% incidence (7/48) of mild mitral regurgitation and 27.1% (13/48) mild tricuspid regurgitation. When using a prosthetic or self tissue patch for defect of ASD, the incidence of regurgitation was 23.5% (8/34), and 42.9% (6/14) for without patch. Other pre-and postoperative hemodynamic data did not affect the result. There was no late right ventricular failure related with tricuspid regurgitation in our series.

Reoperation

Late reoperation rate for residual VSD, PVO, baffle leakage or LVOTO was 14.6% (7/48): group I 12.8% (5/39), group II 22.2% (2/9). We found residual VSD in 4 patients : one in group I was regarded as insignificant at preoperatively and 3 in group II showed the VSD shunt flow postoperatively, one of them, who also manifested LVOTO, required reoperation. During postoperative follow-up period, we detected 3 intraatrial baffle leakages and all of them were corrected with reoperation. As we mentioned before, 3 patients among 15 LVOTO cases had got reoperation for relief of obstruction like subvalvular membrane resection and LVOT mymectomy. One of 3 PVO required reoperation. We couldn't find any difference whether using the additional patch for atrial septal defect (14.7% in using patch vs. 14.3% in non patch).

DISCUSSION

Since 1959, Senning introduced an intraatrial baffle repair for correction of TGA, that procedure had been used at most institution together with the Mustard operation, but long term follow-up studies have documented a remarkable incidence of morbidity and mortality related to venous baffle obstruction as well as arrhythmia[8].

Mortality

According to the recent results of Senning operation for simple TGA, the operative mortality was 5.4%[1], and 17% in the first week of life[9]. Children with simple TGA may develop pulmonary vascular obstructive disease (PVOD) as early as 2 years of age and in rare instance even under 1 year of age, so operation at a younger age may prevent the development of PVOD. When combined with VSD, PVOD developed much earlier than simple TGA, that maybe related to the high operative mortality in TGA with VSD[10]. We found significantly higher LVP in mortality group, which related with the high PAP and PVR. In multi-institutional analysis the overall mortality was 15%: 14% for simple TGA and nearly 50% for complex TGA[11, 12]. Also in our series, mortality for simple TGA is only 4.9% but in combined with VSD the mortality is much higher than other reports. Our recent result of the Jatene procedure for combined VSD, the mortality is 6.3%, which should be compared with these data. Figure 1 shows the actuarial survival curve for simple TGA and TGA with concomitant VSD.

Arrhythmia

Postoperative arrhythmia have been noted since the early days of atrial repair of TGA and modifications of the surgical technique have been developed without successful result to decrease the incidence and severity of these arrhythmia[2]. The cause of these arrhythmia are thought to be due to the extensive dissection around the sinus node with fibrosis in this area or sinoatrial nodal artery thrombosis, interruption of intraatrial communications by baffle suturing and future stretching contraction around the sinus node[13~15]. According to the recent data, 70% of free from serious rhythm disturbance 10 years postoperatively[1] and 53% of patients can be expected to be free from junctional rhythm 20 years after atrial repair[16]. In our results the incidence of various arrhythmia was 33.3% and the freedom from arrhythmia was 66% in 13 years postoperatively (Fig. 2). This might be comparable to other report but this complication rate should be regarded still high, that was one of the reason we change our policy for managing TGA with Jatene operation.

LVOT obstruction

LVOTO has been reported in 13 to 38% of all patients with TGA and in about 8% of those without VSD[17], and in our previous report about less than 1 week of life we showed 11% incidence[9]. Malaligned VSD with posteriorly deviated infundibular septum can cause varying degrees of LVOTO[3]. Subvalvular obstruction is the most common type of LVOTO and occurs in various forms including circumferential fibrous ridge, a fibromuscular tunnel, anomalous attachments of the mitral valve, a pouch of tricuspid leaflet tissue or an aneurysm of the membranous septum protruding into the LVOT and posterior bulging of the ventricular septum in combination with systemic anterior movement of mitral valve. Severe LVOTO is difficult to treat effectively. Successful use of LV to PA conduits may have a role in selected patients[18]. In our series 3 severely

obstructed LVOT required reoperation and one of them died during reoperation. They all showed subvalvular diaphragm and one concomitant fibromuscular tunnel.

Pulmonary venous pathway obstruction

Senning operation utilizes very little foreign material to reroute systemic and pulmonary venous return, thereby optimizing the potential for atrial growth and conduction[1]. The reported complication rate of PVO were 6-16%[2, 3, 8] which were comparable to our series. One of our patients needed reoperation for PVO and relieved successfully followed by uneventful postoperative course. Figure 3 shows the estimated rate of freedom from PVO.

Systemic venous pathway obstruction

SVO was not found in our series of patients. Only the SVC obstruction rarely required reoperation, but when combined with IVC obstruction reoperation was often necessary. It appears that if adequate collateral circulation to a patient IVC is present, the obstructed SVC can decompress[19]. Our series can compare favorably with the reported incidence of up to 5% in infant and up to 10% in older patients[7~9].

Tricuspid regurgitation and Right Ventricular Failure

RV dysfunction after atrial repair of TGA occurs in 10% or more of patients within 10 years of operation[20, 21]. This complication appears to be much more common in those patients with TGA in whom a VSD was closed surgically. The clinical course is often progressive with increasing tricuspid valve regurgitation, aortic regurgitation and further

=Abstract=

Complications after Senning Operation for TGA with and Without VSD

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We analysed 60 consecutive patients who got Senning operation for transposition of the great arteries (TGA) with or without ventricular septal defects (VSD). There were 41 simple TGA (group I) and 19 TGA with VSD (Group II), the operative mortality was 20% (in group I 4.9%, group II 52.6%). Among the survivors (n=48), the mean follow-up period was 7 years (range, 1 year to 13.5 years) and the actuarial survival rate at 13 years were 95% in group I and 42% in group II. Preoperative high left ventricular pressure and high pulmonary arterial pressure affected the surviving ($p<0.01$). There occurred various type of arrhythmia like junctional rhythm, first degree atrioventricular (AV) block, sick sinus syndrome and complete AV block, and we inserted 2 permanent pacemakers for these patients. The incidence of arrhythmia were 28.2% (11/39) in group I and 55. 6% (5/9) in group II, and the actuarial freedom from arrhythmia at 13 years after operation was 66% (71% in group I, 44% in group II). Increased aortic cross clamping time had affected the development of arrhythmia ($p<0.05$) which meant the complexity of the operation. The total incidence of left ventricular outflow tract obstruction (LVOTO) was 31.3% (15/48), but only 3 patients (6.25%) showed the significant gradient requiring reoperation. The pulmonary venous pathway obstruction (PVO) were found in 3 patients, all in group I, and among them only one required the reoperation. The estimated freedom from PVO was 89% at 13 years (87% in group I, 100% in group II), but we couldn't find any significant systemic venous obstruction in our series. There occurred 27.1% (13/48) mild degree tricuspid valve regurgitation without necessary surgical correction. We experienced 14.6% (7/48) reoperation rate: 3 residual VSD, 3 LVOTO, 1 PVO, 3 atrial baffle leakage.

For this high incidence of complication rate after Senning operation and high mortality in TGA with VSD, We do not use this kind of surgical modality any more and do the Jatene operation for all the TGA patients since several years ago.

Key words : Senning operation, Simple TGA, Complex TGA, Complication

RV distention and dysfunction. The cause of tricuspid regurgitation in these patients is multifactorial and can be assumpted 1) primary RV dysfunction leading to increase RV dimensions may result in stretching of tricuspid valve ring and regurgitation, 2) organic damage to the tricuspid valve occurs sometimes as a result of VSD patching or repair 3) organic damage to the tricuspid valve result from the VSD, namely, thickening and shortening of adjacent leaflet tissue perhaps as a result of jet trauma[22]. In addition to these, the previously existed intrinsic structural abnormalities of the tricuspid valve were considered the other cause of postoperative regurgitation in patients with d-TGA with VSD[23]. This tricuspid valve regurgitation were reported 6.5- 33%[3, 4] that could be comparable to our series, but some report only included the patients who required the valve reoperation, so we could not compare directly the result. In our series there was no patient who required the surgical correction of AV valve. Graham reported that RV function could be well preserved if the intraatrial repair were performed at a younger age, better preoperative function and good intraoperative protection. They also considered postoperative abnormalities may be due to preoprative hypoxia, intraoperative myocardial hypoxia and right ventricle subjected to an abnormal afterload[24, 25]. We observed only one patient showed moderately enlarged RV with grade I tricuspid regurgitation, but she is still in NYHA class I during the more than 5 years follow-up.

Since this series, with its significant late morbidity of Senning operation and high mortality for TGA with concomitant VSD, we abandoned the atrial rerouting operation and have performed the arterial switch operation especially for combined VSD with 6. 25% mortality.

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