Total cavopulmonary connection in children with body weight less than 10 kg

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Received 7 September 1999; received in revised form 30 December 1999; accepted 22 February 2000

Abstract

Objective: To evaluate the results after total cavopulmonary connection (TCPC) in small children, our clinical experience was retrospectively reviewed.

Methods: Of 164 patients undergoing TCPC, the body weight at operation was less than 10 kg (8.8 ± 1.1 kg) in 54, including 21 with visceral heterotaxy. The superior caval vein (SVC) was anastomosed to the pulmonary arteries in a bidirectional fashion. To construct a channel draining the inferior caval vein (IVC), an extended polytetrafluoroethylene (ePTFE) tube was placed intraatrially (in 15 patients) or outside the heart (in 13), its diameter being 14 mm in two patients, 16 mm in 12, and 18 mm or greater in 14. A heterologous pericardial baffle was used for intraatrial rerouting in 12 patients. A pedicled autologous pericardial roll was tailored as an extracardiac conduit in 11 patients, and the pulmonary trunk was directly anastomosed to IVC orifice in three.

Results: Seven patients, including five with right isomerism, died in the intermediate term because of infection of the ePTFE tube in two, respiratory problems in three, atrioventricular valvar regurgitation in one, and pulmonary venous obstruction in one. Postoperative catheterization showed; SVC pressure, 11 ± 2 mmHg without a pressure gradient between SVC and IVC; systemic ventricular end diastolic pressure, 5 ± 2 mmHg; end diastolic volume, 122 ± 54% of the anticipated normal value; ejection fraction, 0.56 ± 0.11; and cardiac index, 2.9 ± 0.7 l/min per m². With the follow-up of 1–116 (35 ± 31) months, the IVC channel has not become obstructive in all, except for one, in whom a pedicled pericardial roll was severely obstructed because of its tortuous extracardiac course crossing in front of the vertebrae. Postoperative growth was generally stable, although body weights and heights were smaller in the majority of patients when compared with the anticipated standards for Japanese children.

Conclusion: TCPC can be justifiably established in small children. The use of autologous tissues seemed preferable for constructing the IVC channel unless anatomic orientation was unsuitable. © 2000 Elsevier Science B.V. All rights reserved.

Keywords: Fontan procedure; Total cavopulmonary connection; Extracardiac Fontan; Pedicled autologous pericardial roll tube; Extended polytetrafluoroethylene tube graft; Growth

1. Introduction

The age at operation has been considered to be one of the limiting factors when successfully establishing the Fontan circulation [1]. The initially proposed limitation of older than 4 years of age at operation [2], nonetheless, has been extended towards younger ages [3,4], partly because earlier establishment of the Fontan circulation may avoid developments of deleterious conditions, in terms of either the ventricular function or the pulmonary circulation [5,6], and might provide better cardiac performance in the long term [7]. Earlier employment of the Fontan procedure, in contrast, obviously has a potential problem in that the sizes of the patients’ bodies are commonly small. The purpose of our present study is to evaluate the results after total cavopulmonary connection (TCPC) in our series of small children.

2. Materials and methods

Since 1988, 164 patients have undergone the Fontan type procedure by TCPC at our institution. Of these, the body weight was less than 10 kg (a minimum of 6.1 kg, a mean of 8.8 ± 1.1 kg) at operation in 54 patients, including 21 with visceral heterotaxy (left isomerism in five and right isomerism in 16). The morphology of the hearts in these patients is listed in Table 1.

The age at TCPC was 10–51 months, at a mean of 22 ± 9 months. Thirty-eight patients were younger than 2 years old, including two undergoing the definitive procedure within the first year of life. The construction of a systemic-to-
Table 1
Morphologic diagnoses in the present series of patients

<table>
<thead>
<tr>
<th>Diagnosis</th>
<th>Patients</th>
</tr>
</thead>
<tbody>
<tr>
<td>A dominant LV with a hypoplastic RV</td>
<td>22</td>
</tr>
<tr>
<td>Tricuspid atresia</td>
<td>11</td>
</tr>
<tr>
<td>Severe tricuspid stenosis</td>
<td>1</td>
</tr>
<tr>
<td>Pulmonary atresia with intact ventricular septum</td>
<td>3</td>
</tr>
<tr>
<td>Double inlet LV (2AVV)</td>
<td>5</td>
</tr>
<tr>
<td>Double inlet LV (CAVV)</td>
<td>2</td>
</tr>
<tr>
<td>A dominant RV with a hypoplastic LV</td>
<td>32</td>
</tr>
<tr>
<td>Mitral atresia and discordant VA connections</td>
<td>1</td>
</tr>
<tr>
<td>Severe mitral stenosis and DORV</td>
<td>3</td>
</tr>
<tr>
<td>Hypoplastic left heart syndrome</td>
<td>3</td>
</tr>
<tr>
<td>Double inlet RV (2AVV)</td>
<td>2</td>
</tr>
<tr>
<td>Double inlet RV (CAVV)</td>
<td>8</td>
</tr>
<tr>
<td>AVSD (CAVV) and DORV</td>
<td>8</td>
</tr>
<tr>
<td>DORV with pulmonary stenosis or atresia</td>
<td>5</td>
</tr>
<tr>
<td>AVSD (CAVV)</td>
<td>1</td>
</tr>
<tr>
<td>Straddling mitral valve and discordant VA connections</td>
<td>1</td>
</tr>
</tbody>
</table>

* LV, morphologically left ventricle; RV, morphologically right ventricle; 2AVV, two atrioventricular valves; CAVV, a common atrioventricular valve; VA, ventriculo-arterial; DORV, double outlet right ventricle; AVSD, atroioventricular septal defect.

pulmonary shunt had been previously carried out in 26 patients, and the Norwood procedure in three. The pulmonary trunk had been banded in 12 patients, including three concomitantly undergoing reconstruction of the obstructed aortic arch. The bidirectional Glenn procedure had been previously employed in 15 patients, at the age of 6–32 (14 ± 7) months, the interval between the partial right heart bypass and TCPC being 3–19 (10 ± 5) months.

The superior caval vein (SVC) was anastomosed to the pulmonary arteries in a bidirectional fashion. For the channel to drain the inferior caval vein (IVC), an extended polytetrafluoroethylene (ePTFE) tube was placed intraatrially in 15 patients [7], or outside the heart in 13 [8]. Its diameter was 14 mm in two patients, 16 mm in ten, and 18 mm in three, in the group of patients undergoing intraaerial grafting, while it was 16 mm in two, 18 mm in nine, and 20 mm in two undergoing extracardiac grafting. In another 12 patients, a heterologous pericardial baffle was used for intraaerial rerouting [7]. Our preference, when employing this option, was the use of the atrial appendage, although pectinated, as an anterior wall of the channel rather than a lateral tunnel technique [9], in order to place the site of the sinus node to a low-pressure atrial chamber connected to the systemic ventricle. The width of the native tissue included within the pathway was designed to be no more than one third of the circumference at any cross-section of the created route [7]. A pediced autologous pericardial roll tube was tailored as an extracardiac conduit [10,11] in 11 patients. The roll was designed to possess 17–20 (18.4 ± 0.9) mm of internal diameter. In the remaining three patients, the pulmonary trunk was directly anastomosed to the orifice of the IVC outside the heart [12], since the pulmonary trunk had a sufficient size for the channel, and direct anastomosis was morphologically feasible by means of extensive mobilization of the pulmonary arteries. Of the 27 patients undergoing extracardiac construction of the inferior caval venous channel, cardiopulmonary bypass was not used [11] in 12.

Fenestration was placed in seven patients. The indication for fenestration was hypoplastic left heart syndrome in three patients, and the preoperatively estimated value of pulmonary resistance was greater than 3 units/m² in four. No interventional management has been subsequently carried out on the fenestration. Other additional surgical procedures were concomitantly necessary with the establishment of the Fontan circulation in 25 patients. In order to avoid the progression of subaortic stenosis, additional aortopulmonary anastomosis [13] was carried out in three patients, and subaortic myectomy in two. Plasty to the atroioventricular valve was considered necessary in nine patients, as was the repair of totally anomalous pulmonary venous connection in five, resection of the atrial septum in five, plasty to the pulmonary arteries in nine, and the Sealy’s procedure for WPW (Wolff–Parkinson–White) syndrome in one.

Anticoagulation was postoperatively continued for 1 year, heparin sodium being infused immediately after the procedure for 1 week, followed by the administration of warfarin potassium and dipyridamole for 1 year. This protocol was chosen even in patients undergoing construction of the venous channel with their autologous tissues, because disorders of liver function and coagulation are commonly seen in patients with the Fontan circulation.

3. Results

No patients died within the first postoperative month. Seven patients, however, died in the longer terms, five of these having right isomerism. The actuarial survival rate was 97 ± 3% at 1 year, and 92 ± 6% at 5 years in the group of patients with usual atrial arrangement, while in those with visceral heterotaxy, this was 79 ± 10% and 72 ± 11% at 1 and 5 years, respectively (Fig. 1). The cause of death was bacterial infection of the ePTFE tube in two patients, upper respiratory tract infection in two,
bronchial bleeding in one, recurrent atrioventricular valvar regurgitation in one, and progressive pulmonary venous obstruction in one. In this particular patient with pulmonary venous obstruction, the venoatrial connection, although not the extracardiac type, was obviously abnormal [14] and obstructive. The pulmonary venous orifices had been surgically cut back twice at the time of the bidirectional Glenn and the Fontan procedures. The obstruction was not related to the surgical procedure for establishment of the Fontan circulation by extracardiac grafting at all.

Postoperative catheterization was carried out 1 year after TCPC in 36 patients thus far. The superior caval venous pressure was $11 \pm 2$ mmHg, and no pressure gradients were present between the SVC, the IVC and the pulmonary arteries in these patients. The end diastolic pressure, end diastolic volume and ejection fraction of the systemic ventricle were $5 \pm 2$ mmHg, $122 \pm 54\%$ of the anticipated normal value, and $0.56 \pm 0.11$, respectively. The cardiac index was calculated as $2.9 \pm 0.7$ l/min per m$^2$.

With the follow-up of $1$–$116$ ($35 \pm 31$) months, no obvious stenosis was found at the bidirectional cavopulmonary anastomosis between the SVC and the pulmonary arteries. The channel for the IVC did not become obstructive in all, except for one, in whom a pedicled pericardial roll became severely obstructive a few months after the Fontan procedure because of its tortuous extracardiac course crossing in front of the vertebrae. Even in two patients undergoing intraatrial grafting using a 14 mm ePTFE tube, the channel remained unobstructive 38 and 116 months after TCPC, respectively. In one of these, angiography was carried out 61 months after TCPC, and the internal diameter of the ePTFE tube proved to be 11.5 mm at its narrowest, probably with a slight peel formation inside the graft (Fig. 2). No obvious dilatation of the inferior caval venous channel was noted in those undergoing construction of the channel using a pedicled autologous pericardial roll (Fig. 3a). Of 12 patients undergoing intraatrial baffling, a minor baffle leakage was noted at the suture line between the heterologous pericardial baffle and the pectinated atrial wall in ten, even 5 years after the procedure (Fig. 3b).

No episodes of atrial flutter or fibrillation were noted in this series of patients. In one patient with right isomerism, paroxysmal supraventricular tachycardia was repeatedly noted, and administration of an antiarrhythmic agent has been effective. In another patient with left isomerism, ventricular tachycardia occurred. In this particular patient, familial QT long syndrome was eventually disclosed. The recurrence of ventricular tachycardia has not been noted since a pacemaker was implanted.

Body weights and heights were basically smaller when compared with the anticipated standards for Japanese children, the calculated $Z$-values being less than zero in the majority of patients (Fig. 4). The $Z$-value for body weight significantly increased at 1 year after TCPC in 37 patients who survived 1 year or more after the procedure ($P < 0.0001$ by the paired $t$-test). No further improvement, however, was noted in the longer terms. No obvious tendency was noted in changes of the $Z$-value for body height. Body height reached an average level in some patients, and its $Z$-value remained less than $-1$ in others.

4. Discussion

When employing TCPC in small children, it is essential
for the surgeon to construct the channels for the SVC and the IVC in a well-designed orientation, since these structures are smaller compared with those seen in older patients. Bidirectional cavopulmonary anastomosis should be achieved without major difficulty. In recent years, the bidirectional Glenn procedure was successfully employed, even during early infancy [15,16]. It remains contentious, in contrast, whether the channel for inferior caval venous drainage has been efficiently constructed or not. Our results demonstrated that the channel was functioning extremely well, even in the intermediate term.

When a prosthetic tube graft is used, the material of the graft and its size must be chosen carefully [17]. Some surgeons place Dacron tube grafts for the venous channel [8,18,19], and others prefer the use of ePTFE tube grafts [7,11,20,21]. As for the size of the tube graft, some surgeons place relatively small conduits, such as a 14 mm-diameter tubes, even in patients with body weights of over 10 kg [20]. Before June 1996, intraatrial baffling using an ePTFE tube graft was our first choice in patients with abnormal venoatrial connections [22]. To avoid the obstruction of blood flow within the atrial cavity caused by the intraatrially-present tube graft, the size of the graft (16.1 ± 1.2 mm) was smaller than that used in patients undergoing extracardiac grafting (18.0 ± 1.2 mm, P = 0.0003 by the t-test). The channel outside the heart has much less influence on either pulmonary venous drainage or inflow to the atrioventricular
valve [22]. Of course, the larger size of the tube graft does not necessarily guarantee that the channel will remain unobstructive throughout the patient’s life. The prosthesis does not grow at all, and peel formation inside the graft is more or less inevitable [23] sooner or later. Although one of our 14 mm tube grafts used did not become obstructive for the first decade, we predict that reoperation will be needed in the future for up-sizing of the conduit. It is unclear at this stage whether an 18 mm tube graft will require replacement. We hope that a 20 mm tube graft could remain sufficiently large until the patient reaches adulthood.

Another obvious disadvantageous aspect of the use of prosthetic materials is infection. As occurred in two of our patients, bacterial infection of the tube graft can be intractable. These patients died before reoperation for the graft replacement. The ePTFE tube had been placed intrathoracically in one and outside the heart in the other. The use of a homograft tube, if available, could be the alternative choice for constructing the inferior caval venous channel. Such an autollogous, however, cannot be ideal, particularly in terms of calcification in the long term when used in small children. In this respect, the use of autologous tissues as external conduits is attractive. No episode of bacterial infection occurred in patients undergoing the extracardiac Fontan procedure either using a pedicled autologous pericardial roll, or by direct anastomosis between the pulmonary arterial trunk and the inferior caval venous orifice. Presumably, such autologous tissues are also more durable in terms of thrombosis than prosthetic materials. Furthermore, hopefully, the potential of growth might persist after construction of the channel, without considerable calcification. The surgeon, however, should pay attention to the precise indication of these options.

Direct anastomosis between the pulmonary arterial trunk and the inferior caval venous orifice requires particular morphological features, such as sufficient diameter of the pulmonary trunk and suitable spatial orientation of the cardiac structures [12]. The orientation between the pulmonary arterial trunk and the IVC is also of surgical importance when a pedicled autologous pericardial roll tube is to be used. As seen in one of our patients, obstruction of the pathway can occur when the channel is constructed to course crossing in front of the vertebrae. Since the autologous roll tube is very flexible, the pathway can be readily compressed by the adjacent rigid structure. In such a circumstance, we should probably have used an ePTFE tube graft with external support. Another indication to be considered for the use of a pedicled pericardial roll is the absence of a history of previous surgical procedures via a median sternotomy and pericardotomy. Adhesion of the autologous pericardium will produce problems when preparing a pericardial roll in a pedicled fashion. In our patients previously undergoing the bidirectional Glenn procedure, a pedicled autologous pericardial roll was unavailable. If earlier establishment of the Fontan circulation by the extracardiac TCPC using a pedicled autologous pericardium is considered justifiable, the primary Fontan procedure, rather than the staged Fontan approach, would be the surgical strategy of choice. Of course, earlier TCPC should not be promoted in those patients with considerable risk factors. In a selected group of patients who are good candidates for the Fontan circulation, the age at TCPC can be younger than 2 years, or even within the first year of life.

It is surely necessary for us to comment briefly on our previous option of intraatrial rerouting. The modification, if employed, should be skillfully carried out with no residual leakage across the suture line between the baffle and the pectinated atrial wall. The presence of the pectinate muscle within the channel draining the IVC, as seen in Fig. 3b, could be another potential problem in the long term. Such an uneven internal surface may produce turbulence within the channel, and could be intimately related to thrombosis.

In this respect, the conventional lateral tunnel maneuver seems superior, because the orientation of the channel to be constructed is straighter and the internal surface of the atrium is smoother at its venous component. An alternative resolution is the construction of an extracardiac channel using autologous tissues, which is our current preference. With this option, the sinus node can be readily placed to the low-pressured atrial chamber, and no residual intracardiac right-to-left shunt can unintentionally persist, since the extracardiac channel is completely divided from the atrial cavity.

Apart from the surgical point of view, the functional result in the long term after TCPC in small children is undoubtedly another matter of concern. Mortality and morbidity were likely to have been related to features of the complicated malformations, rather than low body weight at operation, since six out of seven patients dying in the longer term had right isomerism (in five) or hypoplastic left heart syndrome (in one). The activities and cheerfulness of the survivors are considerably good, with no limitations seen in their daily and school lives. Subjectively, the postoperative exercise capacity, such as running, seems no worse, or even better, in these patients than that seen in our patients undergoing the Fontan procedure at older ages. Objective examinations for determining exercise tolerance, however, have yet to be carried out in the majority of the present series of patients because of their young ages. We will be able to obtain further information concerning this matter several years later. Currently, body weight and height were the only data objectively available. The postoperative patient growth did not prove to be pessimistic in the intermediate term, although Z-values below —1 were not uncommon for their body weights and heights. We must still carefully follow up all the survivors for longer in this respect.

In conclusion, TCPC can be established in small children with body weights of less than 10 kg. The use of autologous tissues would be preferable for constructing the inferior caval venous channel, particularly in patients undergoing no previous surgical procedures via a median sternotomy,
unless anatomic orientation is unsuitable. Otherwise, the extracardiac placement of an ePTFE tube graft is justifiably employed, providing no obvious obstruction across the channel in the intermediate term.

References


Appendix A. Conference discussion

Dr J.A.M. van Son (Leipzig, Germany): By staging the Fontan operation in a bidirectional Glenn operation at an early age and subsequent completion of the Fontan circulation at a body weight of about 18–20 kg, the patient is protected from volume overload of the single ventricle and its related detrimental sequelae, and secondly, a (nearly) adult sized conduit, preferably in an extracardiac fashion, can almost always be incorporated. I am concerned about the heterogeneity of your Fontan procedures, such as tube grafts and pericardial baffles, constructed both intraatrially and extracardially, and about the concept of performing the Fontan operation in one stage. Seven of your 48 patients (15%) died at early or intermediate follow-up. One of the most important advantages of performing a two-stage Fontan operation is that the bidirectional Glenn allows the single ventricle to adapt and remodel in accordance with the reduced volume loading with a consequently much reduced risk of mortality; in fact, this process, together with early volume reduction, is the most important cause for the much improved Fontan results during the last decade in some centers. Why don’t you follow a two-stage Fontan strategy, because I am concerned about using a 12–16 mm conduit, as you did, as there may be quite an early need for reoperation because of obstruction. Secondly, you indicated that you observed calcification of the intraatrial baffle or tube graft in some of your patients. Are you not concerned about systemic embolization?

Dr Uemura: The first question, our preference is the primary establishment of the Fontan circulation at a body weight of about 18–20 kg, and the patient is protected from volume overload of the single ventricle and its related detrimental sequelae, and secondly, a (nearly) adult sized conduit, preferably in an extracardiac fashion, can almost always be incorporated. I am concerned about the heterogeneity of your Fontan procedures, such as tube grafts and pericardial baffles, constructed both intraatrially and extracardially, and about the concept of performing the Fontan operation in one stage. Seven of your 48 patients (15%) died at early or intermediate follow-up. One of the most important advantages of performing a two-stage Fontan operation is that the bidirectional Glenn allows the single ventricle to adapt and remodel in accordance with the reduced volume loading with a consequently much reduced risk of mortality; in fact, this process, together with early volume reduction, is the most important cause for the much improved Fontan results during the last decade in some centers. Why don’t you follow a two-stage Fontan strategy, because I am concerned about using a 12–16 mm conduit, as you did, as there may be quite an early need for reoperation because of obstruction. Secondly, you indicated that you observed calcification of the intraatrial baffle or tube graft in some of your patients. Are you not concerned about systemic embolization?

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Dr A. Uhran (St. Augustin, Germany): I would like to support the concept of early TCPCs, as a Glenn may be avoided. Doing a Glenn means doing another operation, which probably is not necessary. Early TCPCs are Glenn-sparing operations.

Secondly, if you choose your tube, whether it is intracardiac or extracardiac, too big, then you lose kinetic energy. I think that the TCPCs with too big a tube are probably not as well off as the ones who have normally-sized or even smallish tubes, like you showed in the two postoperative
patients who had restudy, and the tube was 14 and 11 mm. I think if the patients are well, smallish tubes are okay.

**Dr M.A. Nawabi (Shiraz, Iran):** In your pedicled pericardial tube, you will have a circumferential suture line anyway, which will compromise the vascularity of the pedicled pericardial tube. That may create a growth problem, which was present in one of your cases.

**Dr Uemura:** Yes, I agree with that. We cannot say that the channel is alive or not, we cannot determine it, but fortunately we have not experienced severe obstruction if the anatomic orientation is not unsuitable for the use of the autologous pedicled roll. Probably, in terms of anticoagulation or thrombosis and infection, the autologous tissue should be better; I believe it. In the long term, we can determine whether the autologous pericardial roll can grow or not.

**Dr B. Maruszewski (Warsaw, Poland):** I think the major task in this group of patients is to volume unload the single ventricle as early as possible, and secondly, to avoid the second operation. Which technique, because there are various techniques, do you think is the best to fulfill these two expectations?

**Dr Uemura:** In our series, the incidence of patients with isomerism or visceral heterotaxy is relatively high. Therefore, probably, postoperative problems can occur even after this kind of definitive repair. Anyway, our policy is the construction of an extracardiac tube, because the option has some advantageous points, such as the fact that we can put the sinus node or all venous drainage of the ventricles to the systemic atrial chamber, and we can also avoid obstruction of the pulmonary vein within the atrium, and probably earlier establishment of the Fontan procedure will preserve the ventricular function much better.

Our current preference is the early establishment of Fontan circulation, at around 1 year of age, or even before 1 year of age, if the patient has very good pulmonary circulation and ventricular function. Otherwise, we will proceed to a staged Fontan.

**Dr R. Guerrero (Newcastle-upon-Tyne, England):** The issue in pediatric patients about quality of life is an important one at the moment. In your experience, have you fenestrated all your cases, or which ones have you fenestrated?

Secondly, how many of these patients have problems secondary to a Fontan circulation, such as recurrent pleural effusions etc. These will have an impact on the quality of life, and will make us decide if we should do babies in the early stage or later on in life?

**Dr Uemura:** That is a very important point. We have placed seven fenestrations in this group, and the majority of such patients have undergone previous bidirectional Glenn procedures. That means that such a patient was in a relatively high risk group within this group. If a previous bidirectional Glenn procedure is not needed, usually we need not put in the fenestration. Even after the construction of a fenestration, we can close outside the heart if the circulation is very good.

And the second point, chylothorax or effusion of the thoracic cavity. It depends on the condition of the patient. If the pulmonary artery is sufficiently large, then the postoperative course would be smooth, or lower pulmonary resistance will lead to a shorter time of placement of the drainage tubes. Younger age does not necessarily mean a prolonged postoperative course, I believe.