Determination of Stenosis and Arrhythmia After Surgical Operation of Partial Anomalous Pulmonary Venous Drainage in Mid & Longterm (Comparison of Double/Single Patch and Warden Operation in Partial Anomalous Pulmonary Venous Drainage): A Comparative Study of Two Different Time Intervals

Parsiyel Pulmoner Venöz Dönüş Anomalisinin Cerrahi Onarımı Sonrası Orta ve Uzun Dönemde Aritmi ve Stenozun Değerlendirilmesi (Parsiyel Pulmoner Venöz Dönüş Anomalisinde Tek/Çift Yama ve Warden Operasyonlarının Karşılaştırılması): 2 Farklı Dönemin Karşılaştırmalı Çalışması

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Öz

Çalışmamızda, Hacettepe Üniversitesi Tıp Fakültesi Kalp-Damar Cerrahisi Bölümü'nde, parsiyel pulmoner venöz dönüş anomalisi (PAPVD) tanısıyla tek yama- çift yama ve Warden prosedürü ile opere edilen 20 hasta (grup A) ve Muğla Sıtkı Koçman Üniversitesi Tıp Fakültesi'nde opere edilen 10 hasta ile Eskişehir Osmangazi Üniversitesi Tıp Fakültesi'nde opere edilen 62 hasta (grup B 72 hasta) retrospektif olarak incelendi. Farklı cerrahi tekniklerinin postoperative v. cava superior darlığı, pulmoner ven darlığı ve aritmi insidansı ile ilişkisi karşılaştırıldı. 2 farklı grup oluşturuldu: 2005-2011 yılları arasında, Hacettepe Üniversitesi Tıp Fakültesi'nde 20 hasta (A grubu) ve 2015-2022 yılları arasında Eskişehir Osmangazi Üniversitesi ile Muğla Sıtkı Koçman Üniversitesi Tıp Fakültesi'nde opere edilen toplam 72 hasta (B grubu). A grubunda 12 kız, 8 erkek hasta ortalama yaş sırasıyla 5.7±3 yıl (0.42-11yıl), ortalama ağırlık 18.67±9.01 kg (5.5-41 kg) idi. B grubunda 39 kız, 33 erkek hasta ise ortalama yaş 6.68±3.70 yıl (1-17 yıl), ortalama ağırlık 23.58±14.75 kg (8-80 kg) idi. Klinik, elektrokardiyogram ve ekokardiyografi incelemeleri derlendi. Erken ya da geç mortalite yoktu. Ortalama izlem süresi sırasıyla 39.73 ay ve 49.82 aydı. İzlemde pulmoner ven,v. cava superior stenozu gelişmedi. A grubunda, Warden prosedürü ve çift yama tekniği ile opere edilmiş birer hastaya rezidüel atrial septal defekt nedeniyle reoperasyon yapıldı. B grubunda ise reoperasyon yoktu. A grubundan 6 hastada, B grubunda da 4 hastada postoperative erken dönemde aritmi gelişti. Takipte tüm hastalar sinus ritmine geri döndü. Pacemaker hiçbir hastada gerekmedi. PAPVD farklı cerrahi tekniklerle güvenli olarak opere edilebilir. Tüm yöntemlerde sinus nodu (SN) hasarından kaçınmak için, çevresinde dikkatli doku diseksiyonu yapılmalıdır.

Anahtar Kelimeler: Aritmi, Obstrüksiyon, Parsiyel Anormal Pulmoner Venöz Dönüş, Stenoz

Abstract

We retrospectively investigated 20 patients who underwent partial anomalous pulmonary venous drainage (PAPVD) surgery with single-double patch, and Warden operation after surgical treatment in Hacettepe Medical School, department of cardiovascular surgery (group A) and 62 patients who underwent surgery with double patch in Eskişehir Osmangazi Medical School and 10 patients in Muğla Sıtkı Koçman Medical School (group B including 72 patients) at different time intervals. We compared postoperative v. cava superior stenosis, pulmonary vein stenosis and arrhythmia in regards of different techniques. We constituted 2 groups: 20 patients of Hacettepe Medical School between 2005-2011 (group A) and 72 patients of Muğla Sıtkı Koçman and Eskişehir Osmangazi Medical School between 2015-2022 (group B). Group A included 12 females, 8 males, the mean age was 5.7±3 years, ranging between 0.42 and 11 years, the mean weight was 18.67±9.01 kg, ranging between 5.50 and 41 kg. Group B included 39 females, 33 males, the mean age was 6.68±3.70 years, ranging between 1 and 17 years, mean weight was 23.58±14.75 kg, ranging between 8 and 80 kg. Clinical findings, electrocardiographic, echocardiographic evaluations were obtained. We found neither early nor late mortality in both groups. The mean follow-up duration were 39.73 months and 49.82 months respectively. No pulmonary venous or v. cava superior stenosis occurred in both groups. Reoperation for residual atrial septal defects required in 1 patient who underwent Warden procedure,1 patient who underwent double patch technique in group A. It didn't require reoperation in group B. In group A, 6 patients presented rhythm disturbance in early postoperative period, as well as 4 patients in group B. In follow-up all patient recovered to sinus rhythm in both groups. Pacemaker wasn't required. PAPVD can be safely operated using different procedures. Meticulous dissection nearby sinus node (SN) should be emphasized to avoid injury of SN for all techniques.

Keywords: Arrhythmia, Obstruction, Partial Anomalous Pulmonary Venous Return, Stenosis

Introduction

Partial anomalous pulmonary venous drainage (PAPVD) is a congenital malformation that represents a physiologic left-to-right shunt with a risk for pulmonary vascular disease, Eisenmenger syndrome, and congestive heart failure (1). PAPVD may be present isolated or concomitant with atrial septal defect (ASD). The most frequent is sinus venosum ASD. Other types include right pulmonary vein (PV) draining into right atrium, connection of right PV to IVC (Scimitar Syndrome), and less frequent right PV draining to the azygos vein or coronary sinus (2,3). The left PV can connect to the left v. brachiocephalica through a vertical venous structure, and bilateral PAPVD is rare (3).

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The most common repair technique is using a patch to forward anomalous pulmonary venous return through the sinus venosus ASD with or without performing patch plasty on SVC. Other techniques are the Warden procedure and longitudinal or transverse transcaval incisions with or without superior vena cava (SVC) patch plasty, i.e. the Takahashi atrial flap (4-8).

In this retrospective study, we aimed to compare two different surgical techniques in PAPVD in regard to arrhythmia or obstruction of the PV's and the SVC. Additionally, we compared 2 different time interval and different centers in regards of surgical results and arrhythmia or obstruction of the PV's and the SVC.

Material and Method

Inclusion Criteria

All of 20 patients under 18 years of age who were diagnosed with PAPVD with or without sinus venosus type ASD's between June 2005-December 2011 in Hacettepe University Medical School, were included for group A in this retrospective study. Ethical approval was obtained from the institutional review board (LUT 12/22 05.06.2012), and written informed consent was obtained from legally authorized representatives priorly the study. Similarly, for group B, all of 62 patients under 18 years of age who were diagnosed with PAPVD with or without sinus venosus type ASD's between January 2015-December 2017 in Eskişehir Osmangazi University Medical School and 10 patients under 18 years of age who were diagnosed with PAPVD with or without sinus venosus type ASD's between January 2018-January 2022 in Muğla Sıtkı Koçman University Medical School were taken. Ethical approval for group B, was obtained from the institutional review board of Muğla Sıtkı Koçman University Medical School (19/01/2023: 2/XI) with a special permission of Eskişehir Osmangazi University Medical School. Our study was planned under the rules of declaration of Helsinki.

Controls of patients were made at the 1st, 3rd, and 6th month postoperative visits, and at the time of study. Electrocardiograms were recorded in 12 leads at 10 mm/mV amplitudes. For patients who had arrhythmia, we obtained Holter records for 24 hours. Echocardiographic evaluations were made by pediatric cardiologists.

Postoperative pulmonary venous and SVC stenosis were determined with transthoracic echocardiogram. Postoperative SN dysfunction was defined as follows: persistent sinus bradyarrhythmia (less than 50 per minute), nodal rhythm, or pause more than 3 seconds seen in Holter monitoring or electrocardiogram.

Demographic Features

The study included 2 groups:

Group A included 12 females 8 males, the mean age was 5.7±3 years, ranging between 0.42 and 11 years, the mean weight was 18.67±9.01 kg, ranging between 5.50 and 41 kg. Group B included 39 females, 33 males, the mean age was 6.68±3.70 years, ranging between 1 and 17 years, mean weight was 23.58±14.75 kg, ranging between 8 and 80 kg.

In group A 18 patients had associated ASD: 10 patients (50%) had high venosum ASD, 4 patients (20%) had secundum ASD, 3 (15%) had patent foramen ovale (PFO), 1 (5%) had low venosum ASD, and 2 patients had none (10%) (Table1). Four patients (20%) had left superior vena cava, 3 (15%) had pulmonary stenosis, 2 (10%) had v.azygos continuity, 2 (10%) had Scimitar Syndrome, and 2 (10%) had heterotaxia syndrome (Table 2). The mean value of the main pulmonary artery (MPA) pressure was 18.10 (15-24.25) mmHg for group A (Table 3).

Table 1. Type of ASD and additional PFO in the groups.

ASD types	Group A (n%)	Group B (n%)
High venosum	10 (50%)	35 (48.6%)
Secundum	4 (20%)	28 (38.9%)
PFO	3 (15%)	0 (0%)
Low venosum	1 (5%)	9 (12.5%)
None	2 (10%)	0 (0%)

Table 2. Associated cardiac anomalies in our patients. (n: number of patients).

	Group A (n%)	Group B (n%)
Persistent left SVC	4 (20%)	16 (22.2%)
Pulmonary stenosis	3 (15%)	10 (13,9%)
Azygos vein continuity	2 (10%)	4 (5.6%)
Scimitar Syndrome	2 (10%)	4 (5.6%)
Heterotaxia syndrome	2 (10%)	4 (5.6%)

In group B 72 patients had associated ASD: 35 patients (48.6%) had high venosum ASD, 28 patients (38.9%) had secundum ASD, and 9 (12.5%) had low venosum ASD (Table 1). Sixteen patients (22.2%) had left superior vena cava, 10 patients (13.9%) had pulmonary stenosis, 4 patients (5.6%) had v.azygos continuity, 4 patients (5.46%) had Scimitar Syndrome, and 4 patients (5.6%) had heterotaxia syndrome (Table 2). The median value of the main pulmonary artery (MPA) pressure was 25 (20.50-28) mmHg for group B (Table 3).

Operative Techniques

In group A, the surgical procedures varied according to the surgeons' preference.

All operations were initiated through a median sternotomy under hypothermic conditions (mean 26.9°C, range 22-29°C). The mean CPB time was 75.6 minutes (range 30-124), and the mean cross-clamp time was 49.1 minutes (range 20-93 minutes)

for all patients. For every patient, cold blood cardioplegia was given to obtain cardiac arrest.

In group A, for the subgroup in which the Warden procedure was preferred, after the sternotomy approach, the azygos vein was doubly ligated and transected to allow mobilization of the SVC. Cannulations of the ascending aorta, inferior vena cava and SVC above the highest anomalous PV were made. Then, the SVC was transected. The tip of the right atrial appendage was amputated and enlarged, similar to the diameter of the SVC. Excision of the trabeculae of the appendage was done. The caudal stump of the SVC was sutured with 5/0-6/0 propylene running sutures. After cutting and enlarging anteriorly, the cephalad SVC was sutured to the right atrial appendage with 5/0-6/0 propylene running sutures.

In group A for the subgroup in which single/double patch technique were preferred, standard cava-atrial cannulation was performed. It was preferred patch closure to divide the caval and pulmonary channels to redirect the SVC to the right atrium and the same aim for anomalous PV's to the left atrium. Right atriotomy was performed through the cava-atrial junction on the lateral side of the SVC. In the case of SVC obstruction, a second patch made of autologous pericardium treated with glutaraldehyde was sewn using a running polypropylene 5/0-6/0 suture.

Table 3. Demographic features of patients according to groups.

	Group A (n=20)	Group B (n=72)	p
Age (year)	5.7 (0.42-11)	6.68 (1-17)	0.992
Weight (kg)	18.67 (5.5-41)	23.58 (8-80)	0.310
Gender			
Female	12 (60)	39 (54.2)	0.834
Male	8 (40)	33 (45.8)	
MPA pressure (mmHg)	18.10 (15-24.25)	25 (20.50-28)	**<0.001**
Duration of CPB	76 (56.75-87)	65 (55.50-70)	**0.035**
Duration of clamping time	49.50 (36.75-59.75)	48.50 (41-57)	0.805
Lowest temperature in CPB (°C)	28 (26-28)	32 (31-32)	**<0.001**
Postoperative drainage (cc)	225 (192.50-300)	65 (50-365)	**<0.001**
Extubation time (hour)	7 (6-8.75)	5 (4.5-5)	**<0.001**
Duration of intensive care unit (ICU) (hour)	25 (24-32)	48 (48-48)	**<0.001**
Duration of hospitalization (day)	6.5 (5.25-7)	5 (5-6)	**0.002**
Arrhythmia in hospital stay	6 (30%)	4 (5.6%)	**0.006**
Reoperation	2 (10%)	0 (0%)	**0.045**
Residual ASD	2 (10%)	0 (0%)	**0.045**
Obstruction of systemic venous return	none	none	
Obstruction of pulmonary venous return	none	none	
Mortality	none	none	

In group B, only double patch technique was preferred and operations were done by single surgeon. All operations were initiated through a median sternotomy under moderate hypothermic condition (mean 31.5°C, range 30-33°C). The mean CPB time was 65.3 minutes (range 35-98), and the mean cross-clamp time was 50.5 minutes (range 30-81 minutes) for all patients. For every patient, cold blood cardioplegia was given to obtain cardiac arrest. While performing double patch technique, standard cava-atrial cannulation was performed. It was preferred patch closure to divide the caval and pulmonary channels to redirect the SVC to the right atrium and the same aim for anomalous PV's to the left atrium. Right atriotomy was performed through the cava-atrial junction on the lateral side of the SVC. A second patch made of autologous pericardium treated with glutaraldehyde was sewn using a running polypropylene 5/0-6/0 suture for all patients.

Statistical Analysis

The statistical analysis was executed using Statistical Packages for the Social Science (SPSS, IBM SPSS Statistics 11.5 (Chicago, Illinois, USA)). Quantitative variables were compared with Kolmogorov-Smirnov in order to control if they were appropriate to normal distribution. Independent samples were compared with Mann Whitney U test. Qualitative variables were analyzed with chi-square test. The descriptive statistics of the quantitative variables which were in normal distribution, were indicated as mean ± standard deviation. The descriptive statistics of the quantitative variables which were not in normal distribution, were indicated as median (25-75 percentiles). The descriptive statistics of the qualitative variables were indicated as frequency (%). $p<0.05$ value was considered significant.

Results

Group A and B were statistically homogenous in regards of age, weight and gender ($p>0.05$ Table 3). The mean follow-up duration were 39.73 months (3-82 months) in group A, and 49.82 (4.5-78 months) in group B, respectively. No mortality occurred in both groups. In group A the mean age of all patients at the time of operation was 5.70±3 years, and

6.68±3.70 years in group B. Other descriptive characteristics are shown in table 3.

Comparison was performed by 2 different series, moreover at different time interval. Group A included patient from 2005 to 2011 and group B from 2015 to 2022. Additionally, operations were performed by different surgeons in group A and by single surgeon in group B.

MPA pressure was significantly higher in group B (p=0.035). Duration of CPB was longer in group A (p=0.035). Lowest temperature was also significantly lower in group A (p<0.01). However, there was no significance in regards of the duration of ACT between groups.

Postoperative drainage (p<0.01), extubation time (p<0.001), duration of hospitalization (p=0.02) were significantly higher in group A related to group B. However, ICU stay was significantly longer in group B (p<0.01) due to the surgeon's preference.

There was no dysrhythmia in any patient prior to surgery in both groups. When we compare group A and B in regards of arrhythmia, the arrhythmia was observed significantly higher in group A (p=0.006).

In group A, 6 patients (30%) had arrhythmia (1 atrial fibrillation, 2 sinus bradyarrhythmia, 3 atrio-ventricular 1st degree (AV) block) following CPB, in ICU stay or in hospital stay and all of patients were discharged with sinus rhythm. We did not find any significant difference between the arrhythmia and non-arrhythmia groups according to the duration of CPB (p=0.603), the lowest degree during CPB (p=0.115), the duration of ACT (p=0.570), the duration of ICU stay (p=0.215), or the mean follow-up (p=0.137) among group A.

In group B, 4 patients (5%) had arrhythmia (sinus bradyarrhythmia) following CPB, in ICU stay or in hospital stay and all of the patients were discharged with sinus rhythm. We did not find any significant difference between the arrhythmia and non-arrhythmia groups according to the duration of CPB (p=0.502), the lowest degree during CPB (p=0.189), the duration of ACT (p=0.450), the duration of ICU stay (p=0.384), or the mean follow-up (p=0.196) among group B.

The duration of postoperative hospital stay was significantly higher in the arrhythmia group among group A (p=0.004). In group B, probably due to the sinus bradyarrhythmia, postoperative hospital stay was not significantly higher (p=0.332).

The arrhythmia subgroup of the group A had significantly lower mean age (p=0.032) and a mean weight (p=0.026) during the operation. Similarly, the arrhythmia subgroup of the group B had significantly lower mean age (p=0.003) and mean weight (p=0.015) were significantly lower during the operation.

Postoperative pulmonary venous obstruction, SVC or inferior vena cava (IVC) obstruction were not detected in both groups. Consequently, no significant difference was obtained in postoperative development of PV/SVC stenosis between two groups. Moreover reoperation (p=0.045) and residual ASD (p=0.045) were significantly higher in group A.

According to post-hoc power analysis results performed by arrhythmia in hospital stay statistics, the power of the study is calculated and 87.43% power was obtained with a value of 0.324 effect size.

Discussion

Even though the results in different time interval and those of different centers were compared, we concluded that in group A, duration of CPB, amount of drainage, duration of hospitalization, postoperative arrhythmia, reoperation, residual ASD were higher. Lowest degree during CPB was lower in group A. Previously, surgeons preferred lower temperature and this might extend the duration of CPB due to the weaning process and as a consequence, these reasons might cause more bleeding and longer hospitalization time. Residual ASD and reoperation in group A were observed in younger and lower-weight patients. The arrhythmia might occur due to the fact that the operations were performed by different surgeons with different techniques and in lower-weight patients which is the reason of difficult exploration.

In the literature, many studies have investigated arrhythmia in the pediatric population following PAPVD surgery (9). In childhood, the incidence of postoperative arrhythmia following congenital heart surgery is 48% (10). Arrhythmia has a close correlation with atrial incision length, myocardial damage, injury of the sinus node or sinus node artery due to cannulation of the SVC, sutures near the conduction system, and acute alterations in physiological parameters postoperatively (11-13). The duration of CPB and deep hypothermia have also important relationships with arrhythmia (14). Dramatic changes of the pressure in cardiac chambers caused by volume and pressure overload may be other reasons for postoperative early arrhythmia (10).

Injury to the SN or SN artery may lead to serious arrhythmias. Yet there is no consensus on the best technique to avoid this risk. The presence of long-term SN dysfunction following single or double-patch technique is contradictory; Alsoufi and Iyer reported no SN dysfunction with the single/double-patch technique (10,15-17). In contrast, Agarwal and Baron showed that the risk is diminished using the single patch technique (18-20). Nevertheless, Buz et al. presented a counterview of the continuity of the postoperative arrhythmia ratio, and they stated that the ratio was 7.3%. Buz et al. and Jaschinski et al. reported that extension of the incision through the cavoatrial union at the time of double-patch repair causes postoperative arrhythmias (44%) (21,22). For the Warden procedure, the ratio of arrhythmia is

10%, and the long-term incidence of arrhythmia is 2.5% (23). Nakahira et al. determined the incidence of arrhythmia following the Warden procedure to be 4% (24).

Some studies have concluded that redo surgery is a reason for postoperative arrhythmia (25). Postoperative AF developed in one of our patients who also underwent redo surgery.

Some studies have concluded that postoperative arrythmia after pediatric heart surgery has an association with different factors such as younger age, lower weight, electrolyte imbalance, prolonged CPB time, deep hypothermic circulatory arrest, surgery type, inotropic use, and cyanotic congenital heart disease (10,11,13,26,27).

Said didn't find any PV or SVC obstruction because of the contraction related with patch in spite of their material in his study (8). Hu and colleagues reported, the dimension of the patch in single-patch technique is the most important point for long-term systemic or pulmonary venous stenosis. A smaller patch increases the risk of PV obstruction; in contrast, a larger patch causes SVC obstruction (8). Alsoufi has been found to avoid cavoatrial stenosis, and anastomosis should be accomplished without tension by providing adequate mobilization of the brachiocephalic vein and SVC (15). In our study, SVC obstruction wasn't detected in both groups. Additionally, if superior enlargement of ASD enlargement is required in PAPVD surgery, it may lead to postoperative PV stenosis, as Hu has reported (8). In contrast, Hu and colleagues did not identify any PV stenosis, SVC stenosis or SN dysfunction (8).

Conclusion

We retrospectively investigated patients who underwent PAPVD surgery with single-patch, double-patch, and Warden procedures but at different time intervals and from different centers.

In the pediatric population, age and weight have a close relation. In some instances, failure of thrive is encountered in addition to congenital heart anomalies, and low weight is not uncommon in this population. At the time of surgery, lower weight may add difficulties for exposure during the operation and undesirable injuries to cardiac structures.

We believe that careful dissection and incision near the sinus node must be emphasized to prevent injury to the sinus node for all types of surgery and by means of the development in surgical era. Moreover, difficulties in exposure in low-weight pediatric patients lead tendency toward postoperative arrhythmia in our study.

We did not observe any postoperative PV/SVC stenosis between the single/double patch and Warden procedure among group A and between group A and group B.

The limitations of our study

Our study does not include a high volume of patients. The comparison was made between 2 different time interval and different centers: Surgical material options and opportunities obtained in time by congenital heart surgery teams might be better in time. In addition, while patients in group B were operated by single surgeon, those of group A were operated by different surgeons.

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