Outcomes of Operated Partial-Intermediate Atrioventricular Septal Defect Patients

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ABSTRACT

Introduction: Follow-up results of patients with partial-intermediate atrioventricular septal defect (AVSD) operated in 1996-2016 at Baskent University are presented.

Patients and Methods: Data obtained from hospital records consists of echocardiographic and angiographic details before surgery, age and weight at surgery, operative details, presence of Down's syndrome, details of postoperative care, early postoperative and latest echocardiographic findings and hospitalization for reintervention.

Results: One hundred seventy eight patient-files were reviewed including 41.6% (n= 74) male, 58.4% (n= 104) female patients. The mean age of patients were 47.09 ± 44.25 (median, 30; 25 and 75 percentiles, 17 and 66.5, respectively) months. The mean body weight during the operation was 15.00 ± 11.22 (median, 11; 25 and 75 percentiles, 8.27 and 17, respectively) kg. Study group included 152 patients (85.3%) with partial AVSD and 26 of patients (14.7%) with partial AVSD. A total of 39 patients (19.1%) were diagnosed with down syndrome. Associated cardiac anomalies were present in 21.3% of patients. Operative technique was modified single-patch technique (Wilcox) in 14.6% (26 patients), pericardial patch in 25.8% (128 patients) and pericardial patch and annuloplasty in 13.5% (24 patients). The cleft in the left atrioventricular (AV) valve was closed in 92.1% all of patients.The early mortality and morbidity in the postoperative first month were calculated as 5.6 and 21.2% and the late mortality (> 1 month) and morbidity rates were calculated as 1.2% and 17%, respectively. The most common cause of late morbidity was left AV valve insufficiency, left ventricular outflow tract obstruction and therefore reoperations (15.2%).

Conclusion: Although the mortality and morbidity rates are low in partial AVSD operations, the rate of reoperations for left AV valve insufficiency and left ventricular outflow tract obstruction are still high. Patients should be done corrective surgery around age two and follow up should be performed in terms of reoperating requirement.

Key Words: Partial atrioventricular septal defect; left atrioventricular valve insufficiency; left outflow tract obstruction; reoperation.

Ameliyat Edilmiş Parsiyel-İntermediate Tip Atriyoventriküler Septal Defektli Hastalarımızın Sonuçları

ÖZ

Giriş: Bu çalışmada, Başkent Üniversitesi Tıp Fakültesinde 1996-2016 yılları arasında ameliyat edilen parsiyel ve intermediate tip atriyoventriküler septal defektli (AVSD) hastaların sonuçları değerlendirilmiştir.

Hastalar ve Yöntem: Hastane dosyaları incelenerek hastaların ameliyat öncesi ekokardiyografi ve anjiyografi bulguları, ameliyat yaşları ve ağırlıkları, ameliyat teknikleri ve süreleri, down sendromu varlığı, postoperatif dönemdeki takip ve ekokardiyografi bilgileri kaydedilmiştir.

Bulgular: Ameliyat edilen toplam 178 hastanın %41.6 (n= 74)'sı erkek, %58.4 (n= 104)'ü kızlardan oluşmuştur. Ameliyat sırasındaki yaş ortalamaları 47.09 ± 44.25 ay (median yaş 30 ay, 25. persentil 17 ay, 75. persentil 66.5 ay), ortalama vücut ağırlıkları 15.00 ± 11.22 kg (median 11 kg, 25. persentil 8.27 kg, 75. persentil 17 kg) olarak hesaplanmıştır. Hastalardan 152 (%85.3)'sinin parsiyel, 26 (%14.7)'sının intermediate tip AVSD hastaları olduğu belirlenmiştir. Hastaların %19.1 (34 hasta)'ini down sendromlu hastalar oluşturmuştur. %21.3'ünde ek kardiyak anomali tespit edilmiştir. Hastaların %14.6 (intermediate tip olan 26 hasta)'sı modifiye tek yama tekniği (wilcox) ile, %71.9 (128 hasta)'u perikard yama, %13.5 (24 hasta)'i perikard yama + annuloplasti yapılarak ameliyat edilmiştir. Hastaların %92.1'inin mevcut klefti kapatılmıştır.

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Hastaların ilk bir ay içindeki erken morbidite oranı %21.2 (35 hasta), erken mortalite oranı ise %5.6 (10 hasta) olarak hesaplanmıştır. Geç morbidite (> 1 ay) %17 (27 hasta), geç mortalite ise %1.2 (2 hasta) olarak hesaplanmıştır. En sık geç morbidite nedeni sol atriyoventriküler kapak yetersizliği, sol ventrikül çıkış yolu darlığı ve bu nedenle yapılan reoperasyonlardır (%15.2).

Sonuç: Parsiyel AVSD'lerin ameliyat sonrası erken mortalite ve morbiditesi düşük olmasına rağmen sol atriyoventriküler kapak yetersizliği ve sol ventrikül çıkış yolu darlığı nedeniyle reoperasyon oranı hala yüksektir. Hastalara çok geciktirmeden iki yaş civarında tüm düzeltme ameliyatı yapılmalı ve reoperasyon gereksinimi açısından yakın izlem yapılmalıdır.

Anahtar Kelimeler: Parsiyel atriyoventriküler septal defekt; sol atriyoventriküler kapak yetersizliği; sol ventrikül çıkış yolu darlığı; reoperasyon.

INTRODUCTION

Atrioventricular septal defect (AVSD) is a congenital heart disease involving the atrioventricular valves, the inlet part of the interventricular septum and the primum part of the atrial septum to varying degrees. Among congenital heart diseases, AVSD is seen with a frequency of 2.9% and approximately 25% of them are partial type AVSD[1]. If there is no symptom in partial AVSD, corrective surgery at an early age is recommended at preschool age but if there are signs of congestive heart failure due to severe left atrioventricular valve insufficiency and atrial shunt[1-3]. Atrioventricular block requiring pacemaker, significant mitral valve insufficiency (MR), left ventricular outflow tract obstruction (LVOTO) may occur after surgery, and patients may need to be operated again[3].

In our study, we aimed to present long-term results by evaluating the data of patients with partial and intermediate type AVSD who were operated at Baskent University Medical Faculty Hospital between 1996 and 2016.

PATIENTS and METHODS

The data of 178 patients with partial and intermediate type AVSD who were operated at Baskent University Medical Faculty Hospital between 1996 and 2016 were retrospectively reviewed. This study was certified by Baskent University Medical and Health Sciences Research Board (Project number: KA19/163). Patients' surgical age, intraoperative weight, preoperative echocardiography findings (especially AV valve insufficiency), presence of down syndrome, pulmonary artery pressures and pulmonary vascular resistance (PVR) determined in preoperative catheter angiography, technique used in surgery, cardiopulmonary bypass and aortic clamp times were examined. Postoperative mechanical ventilation times, inotropic agent administration times, inotrope scores, length of stay in intensive care, presence of infection, postoperative AV block and arrhythmia, early mortality and morbidity, discharge time, postoperative early echocardiography findings, follow-up times, and the last echocardiographic findings, late mortality and morbidity during follow-up were recorded. The inotrop score was calculated as follows[4,5]; dopamine μg/kg/min x 1 + dobutamine μg/kg/min x 1 + adrenaline μg/kg/min x 100 + milrinone μg/kg/min x 10.

Statistical Analysis

Statistical analysis were performed using the "Statistical Package for the Social Sciences software (version 20 for Windows; SPSS, Chicago, IL, USA)" program. T test and analysis of variance (ANOVA) were used for comparisons of mean between groups, and Chi-square test was used to compare categorical variables. For the reoperation risk factor significance, cox regression analysis was used. Statistically, a p value of < 0.05 was considered significant.

RESULTS

Of the 178 operated patients, 41.6% (n= 74) were male and 58.4% (n= 104) were female. Mean age at surgery 47.09 ± 44.25 months (median age 30 months, 25th percentile 17 months, 75th percentile 66.5 months), mean body weight 15.00 ± 11.22 kilograms (kg) (median 11 kg, 25th percentile 8.27 kg, 75th percentile 17 kg). One hundred fifty two of the patients were partial and 26 were intermediate type AVSD. 19.1% of the patients (34 patients) were diagnosed with down syndrome. 21.3% had additional cardiac anomaly. The most common additional cardiac anomalies were secundum atrial septal defect (ASD) and patent ductus arteriosus (PDA). General characteristics of the patients are shown in Table 1. 14.6% of the patients were operated with modified single patch technique (wilcox), 71.9% (128 patients) pericardial patch, 13.5% (24 patients) pericardial patch + annuloplasty. The cleft was closed in 92.1% of the patients.

The early morbidity of the patients in the first month was 21.2% (35 patients), the most common cause of morbidity was pericardial effusion and infection. Early mortality was calculated as 5.6% (10 patients). When early mortality is calculated separately; It was found that 15.3% (four patients) for the intermediate type and 3.6% (six patients) for the partial type. Five of the patients died in the first 48 hours postoperatively due to hemodynamic instability (four patients partial, one patient intermediate type), four patients sepsis (one partial, three intermediate type), one patient died on the 13th postoperative day due to sudden arrhythmia and cardiac arrest (partial type). In 36% of the patients, in the preoperative period, in 27.6% in the postoperative period, and in 48% of

Table 1. General characteristics of patients

	Mean ± SD (min-max)
Age (month)	47.09 ± 44.25 (3-240)
Weight (kg)	15.00 ± 11.22 (3-64)
Pulmonary artery pressure (mmHg)	22.83 ± 8.54 (9-62)
Pulmonary vascular resistance	1.51 ± 1.19 (0.1-7.9)
Cardiopulmonary bypass time (min)	71.93 ± 24.59 (36-185)
Aortic clamp time (min)	48.48 ± 19.32 (22-141)
Inotrop time (day)	2.65 ± 5.10 (0.2-53)
Inotrop score	7.61 ± 12.38 (2-122)
Mechanical ventilator duration (hour)	20.40 ± 19.32 (4-456)
Intensive care unit stay (day)	3.97 ± 9.22 (1-110)
Time to discharge (day)	6.58 ± 4.59 (3-24)
AV valve reoperation time (year)	4.51 ± 2.73 (1-10)
LVOTO reoperation time (year)	2.62 ± 1.88 (0.5-5)

AV: Atrioventricular, LVOTO: Left ventricular outflow tract obstruction, SD: Standard deviation.

the patients, 3rd and 4th degree mitral insufficiency (MR) were detected in the last echocardiography. No significant difference was found between the surgical techniques in terms of the degree of MR in the postoperative period (p= 0.135). There was no significant difference between preoperative MR (p= 0.640) and postoperative MR (p= 0.639) in patients with partial and intermediate defects.

Postoperative infections, confirmed by blood, urine, or tracheal aspirate cultures, were seen in 7.8% of the patients. Postoperative complete AV block developed in 21 patients (11.8%), of which only five patients had permanent AV block and pacemaker was implanted. Late morbidity (> 1 month) was 17% (27 patients), and late mortality was 1.2% (two patients). The most common late morbidity reasons were MR, left ventricular outflow tract obstruction (LVOTO) and reoperations performed for this reason (15.2%).

Reoperations

Valve surgery was performed in 21 patients (11.8%) due to significant MR. Mitral valve repair was performed in 7 patients (3.9%) and mitral valve replacement in 14 patients (7.9%). Nineteen of these patients had partial defects and two of them were intermediate type. Postoperative LVOTO developed in nine patients and 4 (2.2%) of these were operated. All of these patients were those who were repaired with partial type and pericardial patch. It was observed that two patients had undergone reoperation for both valve and LVOTO. The mean time for reoperation for MR was 4.48 ± 2.65 (1-10) years, and for LVOTO was 2.62 ± 1.88 (0.5-5) years.

In terms of reoperation for left AV valve and LVOTO, no significant risk factor was found for reoperation in the analysis performed by determining the absence of down syndrome, surgery age and weight, mitral insufficiency before and after surgery, type of AVSD, surgical technique, not closing the cleft, as risk factors.

Comparison of Patients with and Without Down Syndrome

Patients with and without Down's syndrome, gender, age of surgery, body weight during surgery, AVSD type, preoperative pulmonary artery pressure (PAP), preoperative and postoperative MR degrees, cardiopulmonary bypass (CPB), aortic clamp times, intensive care inotrope score, the durations of mechanical ventilation were compared in duration of stay in the intensive care unit, duration of discharge, postoperative AV block and infection, early mortality and morbidity, late mortality and morbidity, reoperations due to valve or LVOTO (Table 2). It was observed that patients with down syndrome had more postoperative infections (p= 0.038). While reoperation was required in patients who did not have down syndrome due to MR and LVOTO, it was determined that patients with down syndrome did not undergo reoperation (p= 0.013). There was no significant difference between the groups in terms of other parameters.

Comparison of Surgical Techniques

Patients were grouped according to the surgical techniques and compared according to the same parameters. Preoperative MR of the patients who underwent annuloplasty was higher than the others (p= 0.008) and the discharge time of these patients was shorter (p= 0.008). Intermediate type AVSD was operated with the Wilcox technique, these patients were found to have higher preoperative PAP (p= 0.001) and PVR (p= 0.001), early morbidity higher in this group (p= 0.003), and more hospital stay (p= 0.008). There was no significant difference between groups in terms of other parameters (Table 3).

Early Mortality

In terms of early mortality, the presence of down syndrome, AVSD type, surgery age and weight, surgery technique, CPB, aortic clamp time, postoperative AV block development and infection were compared. It was found that early mortality was higher in the intermediate type (p= 0.019), in patients with lower age (p= 0.011) and lower weight during surgery (p= 0.013), higher preoperative PAP (p= 0.019), longer CPB duration (p= 0.016), and long aortic clamp time (p= 0.001).

Table 2. Comparison of down and not affected by down syndromes patients

	Down syndrome (n= 34) (mean ± SD)	Not affected by down syndrome (n= 144) (mean ± SD)	p value
Age (month)	47.20 ± 42.47	47.06 ± 44.81	0.987
Weight (kg)	14.81 ± 13.09	15.05 ± 10.78	0.923
Pulmonary artery pressure (mmHg)	24.75 ± 9.85	22.41 ± 8.18	0.702
Pulmonary vascular resistance	1.76 ± 1.44	1.44 ± 1.12	0.231
Cardiopulmonary bypass time (min)	73.41 ± 24.98	71.58 ± 24.57	0.702
Aortic clamp time (min)	48.12 ± 16.87	48.57 ± 19.91	0.893
Inotrop time (day)	1.71 ± 1.05	2.89 ± 5.69	0.269
Inotrop score	6.04 ± 4.42	8.02 ± 13.70	0.206
Mechanical ventilator duration (hour)	15.21 ± 12.59	21.52 ± 49.22	0.195
Intensive care unit stay (day)	3.28 ± 2.05	4.12 ± 10.11	0.375
Time to discharge (day)	7.75 ± 5.73	6.19 ± 4.17	0.400
AV valve reoperation time (year)	-	4.15 ± 2.66	
LVOTO reoperation time (year)	-	2.62 ± 1.88	

AV: Atrioventricular, LVOTO: Left ventricular outflow tract obstruction, SD: Standard deviation.

Table 3. Comparison of surgical techniques

	Modified single patch (Wilcox) (mean ± SD)	Pericardium patch (mean ± SD)	Pericardium patch + annuloplasty (mean ± SD)	p value
Age (month)	29.51 ± 33.73	50.99 ± 44.90	46.20 ± 47.90	0.978
Weight (kg)	10.35 ± 6.18	15.80 ± 11.57	16.03 ± 12.76	0.655
Pulmonary artery pressure (mmHg)	28.85 ± 10.68	21.92 ± 7.71	20.14 ± 6.56	**0.001**
Pulmonary vascular resistance	2.31 ± 1.98	1.44 ± 0.95	0.94 ± 0.62	**0.001**
Cardiopulmonary bypass time (min)	74.81 ± 18.99	69.41 ± 25.23	82.04 ± 24.61	0.384
Aortic clamp time (min)	53.70 ± 13.49	46.43 ± 20.98	53.46 ± 13.20	0.278
Inotrop time (day)	4.34 ± 5.67	2.56 ± 5.52	1.45 ± 0.94	0.154
Inotrop score	6.76 ± 12.82	7.67 ± 13.47	8.13 ± 6.23	0.847
Mechanical ventilator duration (hour)	29.20 ± 52.19	20.07 ± 47.78	12.79 ± 6.23	0.072
Intensive care unit stay (day)	4.81 ± 5.25	4.12 ± 10.73	2.33 ± 1.09	0.096
Time to discharge (day)	11.60 ± 6.02	5.08 ± 2.01	7.28 ± 5.73	**0.008**
AV valve reoperation time (year)	4.95 ± 2.89	4.23 ± 2.77	3.16 ± 2.56	0.757
LVOTO reoperation time (year)	-	2.62 ± 1.88	-	

AV: Atrioventricular, LVOTO: Left ventricular outflow tract obstruction, SD: Standard deviation.

DISCUSSION

In the previous years in partial type AVSD, if there are no signs of heart failure, it was recommended to perform all correction surgery at preschool ages, but nowadays, it has been shown that the results of surgery are better at earlier ages[2,6-9]. Many centers prefer the age of three to four years as the age of surgery due to lower surgical complications. However, Devlin et al. did not find an increase in early mortality, left AV insufficiency or stenosis, pacemaker need, need for reoperation for AV valve or LVOTO in patients who underwent early repair around the median 1.5 years in their series, and they recommended surgery around the age of two years[8].

The early mortality of partial AVSDs in the first 1 month postoperatively has decreased from 10% to 1-5% in the last 20 years[2,3,7,8]. In our study, early mortality was 5.6% in all patients, 3.6% in the partial type, and the median age at surgery was 30 months.

Although early mortality is low in atrioventricular septal defects, the main problem is the high rate of reoperation due to left AV valve insufficiency and/or LVOTO. The reoperation rate for AVSD has been reported as 6-28%[2,7,8,10]. The most common cause of reoperation is AV valve insufficiency and its frequency is 5-19%[2,11]. Moderate or severe left AV valve insufficiency in the early postoperative period causes reoperation[2,7,11-14]. Bove et al., stated that left AV valve insufficiency was caused by technical deficiencies such as separation of the cleft suture or incomplete cleft closure, or insufficient repair caused by not clear recognition of the morphological anomalies of the valve[11]. Anatomic residual lesions of the left AV valve are common causes of reoperation in children with partial AVSD. The most common anomalies; anomaly adhering chordae and/or additional papillary muscle anomalies are asymmetric development of one of the superior leaflets. Subvalvular apparatus anomalies of the valve are also common in partial AVSD[11,13]. Therefore, surgery in partial AVSD should be performed before these secondary fibrotic changes begin, and it is preferably recommended before two years of age[8,11]. Failure to close the cleft in the left AV valve causes significant valve insufficiency in the postoperative period[2,14,15]. Kleft closure and postoperative annular dilatation are the most important factors affecting the results of valve repair[14,15]. Palada et al. recommended ring-shaped reduction with cleft repair to reduce reoperation rates and prevent long-term valve insufficiency[15]. The reoperation rate was 11.8% in our patients due to left AV valve insufficiency. It was observed that 92.1% of the patients had cleft closed during surgery. However, no difference was found between surgical techniques in terms of postoperative mitral insufficiency and reoperation. In our patient group, preoperative and postoperative MR, keeping the cleft open were not found as risk factors for reoperation for the left AV valve. Left ventricular outflow tract obstruction is the second most common cause of operation in AVSD and its frequency is reported as 1-10%[3,8,16-19]. Since the aortic valve is located in front and to the right in AVSDs, it cannot be placed between the AV valves normally. The LVOT is longer and its distal part is narrower. Elonge and abnormal angled LVOT and various intrinsic anomalies facilitate the formation of stenosis. The abnormal AV valve and subvalvar apparatus anatomy also greatly affects the LVOT geometry. Abnormal adhering AV valve chordates and accessory fibrous bands also cause stenosis. Due to these anatomical features in AVSD, LVOT stenosis may develop at many different levels after surgery[3,8,16-19].

Although the modified single patch technique was held responsible for LVOTO in the past, many publications have shown that it is not related[16-20]. In this study, we compared surgical techniques and types of AVSD in terms of LVOTO development, and we found no statistically significant difference. Again, there was no statistically significant difference between surgical techniques and types of AVSD between reoperation for AV valve and LVOTO. In previous studies, it was reported that there was no difference between AVSD types in terms of reoperation frequency due to AV valve and LVOTO[13,15]. However, in our study in which we compared partial and complete AVSD, we reported that postoperative valve insufficiency and therefore the rate of reoperation were high in partial AVSD[21]. Since patients with down syndrome have a higher tendency to pulmonary vascular reactivity and respiratory complications, the postoperative period of mechanical ventilation is longer, the risk of infection is higher, and the duration of intensive care stay is longer[22,23]. However, in many studies, down syndrome was not found as a risk factor for operative mortality. Mortality rates are the same as patients without down syndrome, reoperation rates were lower[23-25]. We also found that patients with down syndrome had significantly more infections in the postoperative period. However, no difference was found between inotrope score, mechanical ventilator, intensive care and discharge times. There was no reoperation in down syndrome patients due to AV valve insufficiency or LVOTO.

CONCLUSION

Postoperative early mortality of partial AVSDs is low, but the reoperation rate is high in the late period due to left AV valve insufficiency and LVOTO. All correction surgery should be performed at the age of two years without delay and close monitoring should be performed in terms of the need for reoperation.

Ethics Committee Approval: This study was approved by Baskent University Medical and Health Sciences Research Board (Date: 07.05.2019 - Project Number: KA19/163).

Informed Consent: Informed consent was obtained.

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