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Early Surgery Outcomes in Congenital Aortic Valve Stenosis in Children: A Cross-Sectional Study

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Abstract

Background: Aortic valve stenosis is a relatively common disorder, and many patients undergo surgical treatment annually. Thus, this study was designed to assess the results of aortic valve replacement in Tabriz's referral hospital from 2006 to 2018.

Methods: Children aged 1 month to 15 years who were diagnosed with congenital aortic valve stenosis and undergone valvular surgery, were included. Based on the findings of echocardiography, the extent of remaining aortic stenosis and aortic insufficiency were evaluated and compared according to different types of surgery, immediately after the surgery and during the 6 months of follow-up. Finally, the results were analyzed comparing the morbidity and mortality of surgical methods.

Results: Among the included patients, 73.8% had left ventricular outflow stenosis at one level, and the remaining had more than one level of stenosis. Prior to surgery, 82.2% of patients had severe ventricular outflow tract stenosis. Immediately after surgery, 91.25% of the patients had no stenosis, or showed mild stenosis. The overall mortality of the operations was 5%. Six months after surgery, only 20% of the patients showed moderate-to-severe stenosis. Web resection and myomectomy showed higher insufficiency rates, and commissurotomy showed increased insufficiency and stenosis. Benthal surgery was shown to reduce remaining rates of insufficiency. Also, a correlation was observed between the mortality rate and commissurotomy surgery. There was no significant relationship between mortality and different levels of aortic stenosis.

Conclusion: The overall success rate of surgery in aortic valve stenosis was acceptable. As different surgical methods implemented in aortic valve stenosis have their own specific pros and cons, regular pediatrician visits are necessary to map-out any possible future complications.

Key Words: Aortic valve insufficiency, Aortic valve stenosis, Congenital heart disease, Pediatric cardiology, Surgical outcome, Valvuloplasty.

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1- INTRODUCTION

Aortic Valve Stenosis (AVS) is a relatively common disorder that is seen in about 5% of children with congenital heart diseases and is caused by the abnormal development of the aortic valve, and the influence of several types of acquired diseases (1-3). The causes of aortic valve diseases differ depending on the ethnicities and the geographical location (4); for instance, the most common cause in Europe and North America is congenital heart disease and in developing countries, it is rheumatic fever (5).

Infants and young children with AVS are often presented with critical clinical symptoms and severe AVS conditions, which account for less than 10% of patients; while the rest of the patients are identified at older ages. The prognosis of infants and young children without intervention is often very poor; on the other hand surgery has been proven to be effective especially in long-term follow ups (5, 6).

Recent advances in cardiothoracic interventions allow earlier treatments of congenital AVS; and current documentation supports aortic valve repair as the primary curative treatment and the last intervention in children with aortic valve disease (6). However, in children with obvious aortic valve destruction or in cases where repair and surgery have failed, the aortic valve replacement is the only curative option (7, 8).

Aortic valve replacement (AVR) surgery in children is associated with distinct clinical and technical problems caused by many anatomical, social and mechanical valve factors (7, 8).

In children with aortic valve stenosis requiring surgery, aortic valve repair should be chosen as the last treatment and aortic valve replacement is best to be used in patients whose aortic valves cannot be repaired or the repairment has failed. Although AVR with mechanical prostheses (MP) can be used as a new option for patients with obvious aortic valve insufficiency and aortic valve annulus dilatation, in infants and young children which the implementation of pulmonary valve autograft is not possible or in patients with invasive endocarditis, the use of homograft valves is considered (7, 9).

Setting the pros and cons of AVR aside, the success of surgery, its adverse events, and its outcomes vary from one center to the other. This study was, thus, conducted to determine the results of AVR in Tabriz's referral hospital.

2- MATERIALS AND METHODS

In this single-center descriptive cross-sectional study, from 2006 to 2018 infants and children aged one month to 15 years with a diagnosis of congenital aortic stenosis, who had undergone surgery at Shahid Madani Hospital in Tabriz, were included; and children who had aortic valve stenosis due to acquired causes, such as rheumatic heart diseases, were excluded from the study. Using the records of these patients, all the necessary information, including demographics, type of aortic valve stenosis, Duration of Cardiopulmonary Bypass (CPB), type of the performed surgery, rate of regurgitation and stenosis immediately after surgery and during 6 months of follow-up, and mortality rate were extracted and analyzed.

2-1. Data Analysis

Data was analyzed using SPSS statistical software (version 26). The normality of the data was checked using the Kolmogrov-Smirnov test. Frequency (with percentage) was used to describe qualitative data. Mean (with standard deviation) was used for quantitative data if it had a normal

distribution; otherwise median (25th and 75th percentile) was used.

Spearman's correlation test was used to investigate the relationship between the type of heart surgery and the degree of remaining aortic valve stenosis and regurgitation after surgery; and also the relationship between the type of aortic valve stenosis and the type of surgery with mortality. The level of statistical significance was considered lower than 0.05.

3- RESULTS

In this cross-sectional study, 84 children with congenital aortic valve stenosis who had undergone open heart surgery were included. The mean ± standard deviation of the current age of the studied patients was 9.154 years. The minimum age of the patients was 1 month and the maximum was 15 years. Forty-seven of the patients (56%) were male and 37 patients (44%) were female. The average weight of the patients was 29.26 ± 20.38 kg. Four of the included patients died during surgery, but no deaths were observed during 6 months of follow-up.

As shown in **Table 1**, the majority of patients had isolated valvular stenosis (45.23%), followed by isolated subvalvular stenosis. Combined conditions were fewer compared to the isolated conditions.

Table-1: Types of surgeries performed in all types of stenosis

type of stenosis	Type of surgery	Number	Total number (Percentage)
Valvular	Commissurotomy	25	38 (45.23%)
	Benthal	3	
	AVR	10	
Subvalvular	Web resection + myomectomy	13	13 (15.47%)
Supravalvular	Aortoplasty	11	11 (13.09%)
Valvular + Subvalvular	AVR + web resection + myomectomy	5	13 (15.74%)
	Commissurotomy + web resection + myomectomy	8	
Valvular + Supravalvular	Aortoplasty + commissurotomy	3	6 (7.14%)
	Aortoplasty	1	
	Aortoplasty + Benthal	1	
	Aortoplasty + AVR	1	
Valvular + Subvalvular + Supravalvular	Commissurotomy + web resection + myomectomy + aortoplasty	1	3 (3.57%)
	Web resection + myomectomy + aortoplasty + AVR	2	

In patients who had valvular aortic stenosis, commissurotomy was performed in 25 patients, Benthal surgery in three patients, and Aortic Valve Replacement (AVR) in 10 cases. Web resection surgery + myomectomy was performed in patients who had only subvalvular stenosis. Only those who had supravalvular stenosis underwent aortoplasty. In five of the patients who had valvular stenosis and infra valvular stenosis, AVR, web resection and myomectomy were performed; while commissurotomy, web resection, and myomectomy were performed in 8 patients. In patients with valvular stenosis and supravalvular stenosis, aortoplasty and commissurotomy were performed in 3 patients; while the

other three cases underwent aortoplasty, aortoplasty and Bentall, and aortoplasty with AVR. In patients who had all three types of stenosis, web resection, myomectomy, aortoplasty and commissurotomy were performed in one patient while AVR, web resection, myomectomy and aortoplasty were performed in two patients (**Table 1**).

In patients who had ventricular outflow tract stenosis only at the valvular level, the majority of patients (86.85%) had severe aortic valve stenosis before surgery. Immediately after surgery 57.9% of the patients showed no valvular stenosis; unfortunately three patients died in the operating room. In the follow-up examinations conducted 6 months after surgery, the results were similar to the immediate echocardiography after surgery, and only 2.9% showed severe stenosis (**Table 2**).

In patients who had isolated subvalvular stenosis, before surgery, the majority of the patients had severe ventricular outflow stenosis (76.93%), and immediately after surgery, 53.84% of the patients showed no stenosis. In the evaluations performed 6 months after surgery, the ratio of the patients with no stenosis increased to 60.54%; while only one patient had severe stenosis. In patients who had isolated supravalvular ventricular outflow tract stenosis, before surgery, approximately 91% of the patients had moderate-to-severe stenosis; immediately after surgery, all of the patients fell into the mild or without stenosis category. In the investigations conducted 6 months after surgery, only 1 patient had developed moderate stenosis; while there were no severe cases observed. In patients who had ventricular outflow stenosis at the valvular and subvalvular level, before surgery, 92.3% of the patients had severe stenosis; immediately after surgery the severe cases fell into the mild, or without stenosis category. Six months after surgery, 69.2% of the patients showed mild-to-moderate stenosis. Unfortunately, the echocardiography result of one patient remained unclear.

All of the patients who had ventricular outflow stenosis at the valvular and supravalvular level before surgery, had severe valvular stenosis. Immediately after surgery, two-thirds of the patients showed mild-to-moderate stenosis; and one patient unfortunately died in the operating room. In the examinations performed 6 months after surgery, no severe cases were detected, and one-third of the patients were stenosis-free.

All patients with ventricular outflow stenosis in all three valvular levels, subvalvular and supravalvular, before surgery, had severe ventricular outflow stenosis, and immediately after surgery, the patients showed mild ventricular outflow stenosis. The investigations were performed 6 months after surgery. One patient had no stenosis and two patients had mild ventricular outflow tract stenosis. The amount of remaining aortic valve stenosis based on echocardiography in all patients is presented in **Table 2**.

As shown in **Table 3**, among the patients who had aortic valvular stenosis before surgery, 68.42% of the patients were without aortic regurgitation and almost 30% had moderate to severe regurgitation based on echocardiography. After surgery, 50% of the patients were regurgitation-free, and moderate to severe rates were reduced to almost 10%.

Table-2: The ratio and number of remaining aortic valve stenosis (AVS) before and after surgery based on echocardiography in patients with prior AVS

Condition	Before surgery	Immediately after surgery	6 month after surgery

Aortic valve stenosis	Without stenosis	-	22 (57.9 %)	20 (57.1 %)
	Mild stenosis (<25 mmHg)	-	11 (28.94 %)	9 (25.7 %)
	Moderate stenosis (25-50 mmHg)	5 (13.15 %)	2 (5.26 %)	5 (14.3 %)
	Severe stenosis (>50 mmHg)	33 (86.85 %)	-	1 (2.9 %)
	Unclear	-	-	-
	Death	-	3 (7.89 %)	-
Aortic subvalvular stenosis	Without stenosis	-	7 (53.84 %)	8 (60.54 %)
	Mild stenosis (<25 mmHg)	-	5 (38.46 %)	2 (15.35 %)
	Moderate stenosis (25-50 mmHg)	3 (23.07 %)	1 (7.7 %)	2 (15.35 %)
	Severe stenosis (>50 mmHg)	10 (76.93 %)	-	1 (7.7 %)
	Unclear	-	-	-
	Death	-	-	-
Aortic supravalvular stenosis	Without stenosis	-	5 (45.45 %)	6 (54.55 %)
	Mild stenosis (<25 mmHg)	1 (9.1 %)	6 (54.55 %)	4 (36.35 %)
	Moderate stenosis (25-50 mmHg)	5 (45.45 %)	-	1 (9.1 %)
	Severe stenosis (>50 mmHg)	5 (45.45 %)	-	-
	Unclear	-	-	-
	Death	-	-	-
Aortic valvular + subvalvular stenosis	Without stenosis	-	6 (46.2 %)	3 (23.1 %)
	Mild stenosis (<25 mmHg)	-	5(38.5 %)	5 (38.5 %)
	Moderate stenosis (25-50 mmHg)	1 (7.7 %)	2 (15.3 %)	4 (30.7 %)
	Severe stenosis (>50 mmHg)	12 (92.3 %)	-	-
	Unclear	-	-	1 (7.7 %)
	Death	-	-	-
Aortic valvular + supravalvular stenosis	Without stenosis	-	1 (16.66 %)	2 (33.33 %)
	Mild stenosis (<25 mmHg)	-	2(33.33 %)	1(16.66 %)
	Moderate stenosis(25-50 mmHg)	-	2 (33.33 %)	2 (33.33 %)
	Severe stenosis (>50 mmHg)	6(100 %)	-	-
	Unclear	-	-	-
	Death	-	1 (16.66 %)	-
Aortic valvular + subvalvular + supravalvular stenosis	Without stenosis	-	-	1 (33.33 %)
	Mild stenosis (<25 mmHg)	-	3 (100 %)	2 (66.66 %)
	Moderate stenosis (25-50 mmHg)	-	-	-
	Severe stenosis (>50 mmHg)	3 (100 %)	-	-
	Unclear	-	-	-
	Death	-	-	-
Aortic valve stenosis in all patients combined	Without stenosis	-	41 (51.25 %)	40 (50 %)
	Mild stenosis (<25 mmHg)	1 (1.2 %)	32 (40 %)	23 (28.75 %)
	Moderate stenosis(25-50 mmHg)	14 (16.6 %)	7 (8.75 %)	14(17.5 %)
	Severe stenosis (>50 mmHg)	69 (82.2 %)	-	2 (2.5 %)
	Unclear	-	-	1 (1.25 %)
	Death	4 (5 %)	-	-

Table-3: The ratio and number of aortic valve regurgitation before and after surgery based on echocardiography in patients with prior aortic valve stenosis (AVS)

Condition	Before surgery	After surgery

Aortic valve stenosis	Without regurgitation	26 (68.42%)	19 (50%)
	Mild regurgitation	1 (2.63%)	12 (31.5%)
	Moderate regurgitation	5 (13.15%)	3 (7.9%)
	Severe regurgitation	6 (15.8%)	1 (2.7%)
	Death	-	3 (7.9%)
Aortic subvalvular stenosis	Without regurgitation	6 (46.15%)	4 (30.76%)
	Mild regurgitation	3 (23.07%)	8 (61.53%)
	Moderate regurgitation	2 (15.38%)	-
	Severe regurgitation	2(15.38%)	1 (7.69%)
	Death	-	-
Aortic supravalvular stenosis	Without regurgitation	7 (63.63%)	8 (72.72%)
	Mild regurgitation	2(18.18%)	2 (18.18%)
	Moderate regurgitation	-	1 (9.09%)
	Severe regurgitation	2 (18.18%)	-
	Death	-	-
Aortic valvular + subvalvular stenosis	Without regurgitation	4 (30.76%)	2 (15.38%)
	Mild regurgitation	2 (15.38%)	8 (61.52%)
	Moderate regurgitation	2 (15.38%)	-
	Severe regurgitation	5 (38.46%)	3 (23.07%)
	Death	-	-
Aortic valvular + supravalvular stenosis	Without regurgitation	4 (66.66%)	2 (33.33%)
	Mild regurgitation	1 (16.66%)	3(50%)
	Moderate regurgitation	-	-
	Severe regurgitation	1 (16.66%)	-
	Death	-	1 (16.66%)
Aortic valvular + subvalvular + supravalvular stenosis	Without regurgitation	-	-
	Mild regurgitation	1(33.33%)	3 (100%)
	Moderate regurgitation	2 (66.66%)	-
	Severe regurgitation	-	-
	Death	-	-
Aortic valve regurgitation in all patients with aortic valve stenosis combined	Without regurgitation	47 (55.95%)	35 (41.66%)
	Mild regurgitation	10 (11.9%)	36 (42.85%)
	Moderate regurgitation	11 (13.09%)	4 (4.76%)
	Severe regurgitation	16 (19.04%)	5 (5.95%)
	Death	-	4 (4.76%)

Almost half of the patients with aortic subvalvular stenosis were without aortic regurgitation; while post-surgical evaluations showed that 61.53% of the cases had developed mild regurgitation. The majority of the patients with aortic supravalvular stenosis before surgery had no aortic regurgitation, and almost 18% had severe regurgitation. On the post-surgical evaluation, no severe regurgitation was detected. In patients who had left ventricular outflow stenosis at the valvular and subvalvular level, before surgery, approximately 54% had moderate-to-severe regurgitation; while after surgery, almost 61% showed mild regurgitation. One-third of the patients with left ventricular outflow stenosis at the valvular and supravalvular level, before surgery, had no regurgitation; while after surgery, half of the patients showed mild regurgitation. Two-thirds of the patients

with left ventricular outflow stenosis in all 3 levels, before surgery, had moderate regurgitation; while after surgery, all of the patients showed mild regurgitation. The degrees of aortic regurgitation based on Echocardiography in all patients are shown in **Table 3**.

The results of evaluating the relationship between the type of heart surgery and the degree of remaining aortic valve stenosis and regurgitation after surgery showed that "web resection + myomectomy" surgery did not increase or decrease the remaining degree of aortic valve stenosis; while the amount of remaining AI was higher in patients who underwent this type of surgery. The analysis showed that the remaining AS and AI levels were higher in patients who had undergone commissurotomy surgery.

The analyses revealed that "Bental" surgery does not increase or decrease the rate of remaining aortic valve stenosis, but it decreases the rate of AI. Furthermore, the analysis showed that there was no significant relationship between aortoplasty and AVR with the remaining rate of aortic valve stenosis and regurgitation.

The results of correlation test between the type of aortic valve stenosis and the type of surgery with mortality showed no significant relationship between mortality and aortic subvalvular, valvular, and supravalvular stenosis; and "Web resection and myomectomy" surgery, Benthal, AVR and aortoplasty. The statistical analysis showed that the mortality rate was higher in patients who had undergone commissurotomy.

Four of the studied cases died immediately after surgery in the operating room (4.76%), among whom one patient (1.19%) had valvular stenosis, and three patients (3.57%) had isolated valvular stenosis. During the 6-month follow-up, there were no deaths in the patients. It should be mentioned that three of these patients were less than 6 months old and one of them was 1 year old; all four patients had severe ventricular outflow tract stenosis and had unstable hemodynamics.

4- DISCUSSION

In a previous and similar study conducted by Brown et al., between 1960 and 2002, 508 patients aged from 1 day to 19 years were evaluated, among whom 81% had ventricular outflow tract stenosis at one level and the remaining had ventricular outflow stenosis at more than one level (10). In another study by Liu et al., left ventricular outflow stenosis at the valvular level was reported to be the most common type (9). Similarly, in the present study, 62 cases (73.8%) of 84 patients had left ventricular outflow tract stenosis at a single level, and only 26.2% had aortic stenosis at more than one level.

The mortality rate in our study was 4.76% (four patients), from which one patient had valvular + supravalvular stenosis (25% mortality) and the other three had valvular stenosis (75% mortality). It should be mentioned that three of these patients were less than 6 months old and one of them was 1 year old; all four patients had severe ventricular outflow tract stenosis and displayed unstable hemodynamics. Similar to our results, in the study by Brown et al., 8% mortality was observed, the largest part of which comprised infants with critical ventricular outflow tract stenosis (10). These results show that prior critical hemodynamics and ventricular outflow dysfunction majorly determine the prognosis of surgery.

In another study by Brown et al., between 1962 and 2000 on 101 patients between the ages of 3 months and 17 years with aortic supravalvular stenosis, one case of premature death was reported (first year overall survival 98%) (10, 11). In an older study by the same author, between the

years of 1957 and 1986, the mortality rate following surgery in children older than 6 months was 4%, while this rate was 60% in infants with severe aortic valve stenosis (1, 11). In a study conducted by Alexiou et al., between 1979 and 2000, two infants (2.1%) died immediately after surgery (12). In other similar studies, either premature death rate was not reported, or the investigated population was scarce (9, 10).

Brown et al., in a study investigating the effects of aortoplasty and endarterectomy on patients, reported that the patients with supravalvular stenosis of the aorta had improved NYHA classifications after undergoing surgery, as most of the NYHA classes III and IV had improved to lower classes (10). In the present study, 82.2% of patients had severe left ventricular outflow tract stenosis before surgery, and 6 months after surgery, 52% of patients had no stenosis, 28.75% had mild stenosis, 17.5% had moderate stenosis, and only 2.5% had severe left ventricular outflow tract stenosis. In a similar study conducted by Alexiou et al., 17 out of 97 patients experienced AS recurrence (1, 12). Furthermore, in a study by Liu et al., the patients who underwent open heart surgery showed a decrease of about 40 mm Hg in the ventricular-aortic gradient; while the patients who did not undergo open heart surgery showed an increase in gradient of about 15-5 mmHg (13). Similar results were achieved by Brown et al., as the patients undergoing surgery showed a mean of 57 mmHg in their ventricular aortic gradient (1).

Witsenburg et al. reported that during a 27-month period, 21 children aged 0.1 to 7.15 years with isolated aortic valve stenosis underwent balloon valvuloplasty, 10 of whom underwent early valvotomy surgery, with a maximum left ventricular systolic pressure of about 35 mm Hg; and the average gradient showed a decrease of about 50 mm Hg (14).

In recent systematic reviews and meta-analyses comparing balloon valvuloplasty and surgical valvotomy, the mortality rate, complications, and short-term results were shown to be similar; although balloon valvuloplasty may require more re-interventions compared to the surgical method (15, 16)

In our study, it is shown that Benthal surgery reduces AI, commissurotomy surgery increases the rate of AI and AS, and webectomy increases the rate of AI; while in the study by Brown et al., valvotomy increased the rate of aortic regurgitation (11). Also, according to the statistical analysis, the mortality rate was correlated with commissurotomy surgery, which can be justifiable by the unstable hemodynamic conditions of the patients.

5- CONCLUSION

According to the findings, AVS presents majorly at a single level; although combined conditions are not uncommon. This condition has been shown to be highly responsive to surgical or interventional methods. Aortic valve regurgitation, as a complication of surgery, was seen more in patients who had undergone web resection and myomectomy. To prevent this complication, these patients should regularly visit a pediatric cardiologist for a visit and echocardiography, and if necessary, aortic valve repair is advised. Also, in patients who had undergone commissurotomy surgery, the remaining amounts of post-surgical AS and AI were high. In these cases, regular care of a pediatric cardiologist is recommended. Furthermore, 6 months after surgery, the prevalence of resolved left ventricular outflow tract stenosis was 50% in all patients, which indicates the high success rate of the operations. The remaining stenosis in the patients can be justified with the level of stenosis being on more than one site, making the operation more challenging and complex.

Finally, it is suggested that the long-term results of aortic valve stenosis surgery should also be investigated, particularly as a cohort study. Also, similar studies should be done in other centers and with a wider scope and dimension.

6- ETHICAL CONSIDERATIONS

The present study has been approved by the Ethics Committee of Tabriz University of Medical Sciences (ethical code: IR.TBZMED.REC.1399.366). The entire patient's information in the study was completely kept confidential.

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8- CONFLICT OF INTEREST

None.

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