e-ISSN: 0976-822X, p-ISSN:2961-6042

Available online on http://www.ijcpr.com/

International Journal of Current Pharmaceutical Review and Research 2023; 15(9); 155-161

Original Research Article

A Study to Ascertain the Early Outcomes Associated with the Surgical Intervention for Congenital Supravalvular Aortic Stenosis (SVAS): An Observational Study

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Received: 04-02-2023 Revised: 11-03-2023 / Accepted: 20-04-2023
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Conflict of interest: Nil

Abstract

Aim: The aim of the present study was to evaluate the early results of the surgical management of congenital supravalvular aortic stenosis (SVAS).

Material & Methods: This single-center descriptive cross-sectional study was conducted at Department of pediatrics for the duration of 2 years infants and children aged one month to 15 years with a diagnosis of congenital aortic stenosis, who had undergone surgery were included. Children who had aortic valve stenosis due to acquired causes, such as rheumatic heart diseases, were excluded from the study. In the present study, 100 children with congenital aortic valve stenosis who had undergone open heart surgery were included.

Results: The mean ± standard deviation of the current age of the studied patients was 9.31±1.46 years. The minimum age of the patients was 1 month and the maximum was 15 years. 55 patients (55%) were male and 45 patients (45%) were female. The average weight of the patients was 28.24 ± 20.36 kg. The majority of patients had isolated valvular stenosis (46%), followed by isolated subvalvular stenosis. Combined conditions were fewer compared to the isolated conditions. In patients who had valvular aortic stenosis, commissurotomy was performed in 28 patients, Benthal surgery in 4 patients, and Aortic Valve Replacement (AVR) in 14 cases. Web resection surgery + myomectomy was performed in patients who had only subvalvular stenosis. Only those who had supravalvular stenosis underwent aortoplasty. In patients who had ventricular outflow tract stenosis only at the valvular level, the majority of patients (83.34%) had severe aortic valve stenosis before surgery. Immediately after surgery 55.55% of the patients showed no valvular stenosis; unfortunately, three patients died in the operating room.

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.

Keywords: Aortic Valve Insufficiency, Aortic Valve Stenosis, Congenital Heart Disease, Pediatric Cardiology, Surgical Outcome, Valvuloplasty.

Introduction

Aortic Valve Stenosis (AVS) is a prevalent condition seen in around 5% of infants diagnosed with congenital heart problems. It arises due to the anomalous development of the aortic valve, as well as the impact of other acquired illnesses. [1-3] Infants and early children diagnosed with AVS often exhibit significant clinical symptoms and have severe AVS problems, constituting a minority of fewer than 10% of all patients. Conversely, the majority of individuals are typically detected at later stages of development. Congenital supravalvular aortic stenosis (SVAS) is an uncommon kind of aortic blockage that may affect both the aortic arch and the brachiocephalic arteries in severe instances. [4,5]

Williams-Beuren syndrome (WBS) is a well-established condition that is characterized by the presence of supravalvular aortic stenosis (SVAS), mental retardation, distinct facial features, and pulmonary artery (PA) stenosis. [6,7] This syndrome is associated with a microdeletion of the elastin gene located on chromosome 7q11.23. This genetic alteration leads to a reduction in elasticity, resulting in increased shear stress, collagen deposition, and thickening of the aortic media. [4,8-10] The prognosis for newborns and young

children who do not get intervention is often unfavorable. Conversely, surgical intervention has shown effectiveness, particularly in long-term follow-up assessments. In recent years, there have been significant developments in cardiothoracic procedures that have enabled the implementation of early therapies for congenital aortic valve stenosis (AVS). Existing literature provides evidence that aortic valve replacement is the preferred and last surgery for children with aortic valve illness, serving as the main curative therapy. [11,12] In situations when there is evident deterioration of the aortic valve in children or when attempts at repair and surgery have been unsuccessful, aortic valve replacement emerges as the only therapeutic alternative. Aortic valve replacement (AVR) surgery in pediatric patients is characterized by unique clinical and technical challenges stemming from various anatomical, social, and mechanical valve variables. [13,14] The utilization of mechanical prostheses (MP) in aortic valve replacement (AVR) has emerged as a viable alternative for patients presenting with evident aortic valve insufficiency and aortic valve annulus dilatation. This option becomes particularly relevant in cases involving infants and young children where the implementation of pulmonary valve autograft is not feasible, or in patients afflicted with invasive endocarditis. Consequently, the application of homograft valves is deemed appropriate in such scenarios. [13,15] When considering the advantages and disadvantages of AVR, it is important to acknowledge that the efficacy of the surgical procedure, as well as the occurrence of adverse events and the resulting results, might differ across different medical centers.

Therefore, the present research was undertaken in order to ascertain the outcomes associated with the surgical intervention for congenital supravalvular aortic stenosis (SVAS).

Material & Methods

This single-center descriptive cross-sectional study was conducted at department of pediatrics, SKMCH, Muzaffarpur, Bihar, India for the duration of 2 years infants and children aged one month to 15 years with a diagnosis of congenital aortic stenosis, who had undergone surgery were included. Children who had aortic valve stenosis due to acquired causes, such as rheumatic heart diseases, were excluded from the study. In the present study, 100 children with congenital aortic valve stenosis who had undergone open heart surgery were included. 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.

Statistical 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.

Results

Table 1: Demographic data

Gender	N%
Male	55 (55)
Female	45 (45)
Average weight	28.24 ± 20.36 kg
Mean age	9.31±1.46

The mean ± standard deviation of the current age of the studied patients was 9.31±1.46 years. The minimum age of the patients was 1 month and the maximum was 15 years. 55 patients (55%) were male and 45 patients (45%) were female. The average weight of the patients was 28.24 ± 20.36 kg.

Table 2: Types of surgeries performed in all types of stenosis

Type of stenosis	Type of surgery	N	Total (%)
Valvular	Commissurotomy	28	46 (46%)
	Benthal	4	
	AVR	14	
Subvalvular	Web resection + myomectomy	16	16 (16%)
Supravalvular	Aortoplasty	13	13 (13%)
Valvular +	AVR + web resection +	7	

Subvalvular	myomectomy		16 (16%)
	Commissurotomy + web resection + myomectomy	9	
Valvular + Supravalvular	Aortoplasty + commissurotomy	4	7 (7%)
	Aortoplasty	1	
	Aortoplasty + Benthal	1	
	Aortoplasty + AVR	1	
Valvular + Subvalvular + Supravalvular	Commissurotomy + web resection + myomectomy + aortoplasty	1	2 (2%)
	Web resection + myomectomy + aortoplasty + AVR	1	

The majority of patients had isolated valvular stenosis (46%), followed by isolated subvalvular stenosis. Combined conditions were fewer compared to the isolated conditions. In patients who had valvular aortic stenosis, commissurotomy was performed in 28 patients, Benthal surgery in 4 patients, and Aortic Valve Replacement (AVR) in 14 cases. Web resection surgery + myomectomy were performed in patients who had only subvalvular stenosis. Only those who had supravalvular stenosis underwent aortoplasty. In 7 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 9 patients. In patients with valvular stenosis and supravalvular stenosis, aortoplasty and commissurotomy were performed in 4 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 1 patient.

Table 3: 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 months after surgery**
Aortic valve stenosis	Without stenosis	-	23 (63.88%)	20 (55.5 %)
	Mild stenosis (<25 mmHg)	-	11 (30.55%)	10 (27.7 %)
	Moderate stenosis (25-50 mmHg)	6 (16.66%)	2 (5.55%)	5 (13.88%)
	Severe stenosis (>50 mmHg)	30 (83.34%)	-	1 (2.7%)
	Unclear	-	-	-
	Death	-	4 (20%)	-
Aortic subvalvular stenosis	Without stenosis	-	8 (40%)	10 (50%)
	Mild stenosis (<25 mmHg)	-	6 (30%)	4 (20%)
	Moderate stenosis (25-50 mmHg)	6 (30%)	2 (10%)	4 (20%)
	Severe stenosis (>50 mmHg)	14 (70%)	-	2 (10%)
	Unclear	-	-	-
	Death	-	-	-
Aortic supravalvularstenosis	Without stenosis	-	5 (50%)	5 (50%)
	Mild stenosis (<25 mmHg)	1 (10%)	5 (50%)	4 (40%)
	Moderate stenosis (25-50 mmHg)	4 (40%)	-	1 (10%)
	Severe stenosis (>50 mmHg)	5 (50%)	-	-
	Unclear	-	-	-
	Death	-	-	-
Aortic valvular + subvalvular stenosis	Without stenosis	-	7 (46.6%)	3 (20%)
	Mild stenosis (<25 mmHg)	-	6(40%)	6 (40%)
	Moderate stenosis (25-50 mmHg)	2 (13.34%)	2 (13.34%)	5 (33.34%)
	Severe stenosis (>50 mmHg)	13 (86.66%)	-	-
	Unclear	-	-	2 (13.34%)
	Death	-	-	-
Aortic valvular + supravalvula	Without stenosis	-	1 (20%)	2 (40%)
	Mild stenosis (<25 mmHg)	-	2(40%)	1(20%)
	Moderate stenosis(25-50 mmHg)	-	1 (20%)	2 (40%)
	Severe stenosis (>50 mmHg)	5 (100 %)	-	-

rstenosis	Unclear	-	-	-
	Death	-	1 (20%)	-
Aortic valvular + subvalvular + supravalvula rstenosis	Without stenosis	-	-	1 (25%)
	Mild stenosis (<25 mmHg)	-	4 (100 %)	3 (75%)
	Moderate stenosis (25-50 mmHg)	-	-	-
	Severe stenosis (>50 mmHg)	4 (100 %)	-	-
	Unclear	-	-	-
	Death	-	-	-
Aortic valve stenosis in all patients combined	Without stenosis	-	50 (55.55%)	45 (50%)
	Mild stenosis (<25 mmHg)	1 (1.11%)	34 (37.77%)	25 (27.77%)
	Moderate stenosis(25-50 mmHg)	14 (15.55%)	6 (6.66%)	16(17.77%)
	Severe stenosis (>50 mmHg)	70 (77.77%)	-	3 (3.33%)
	Unclear	-	-	1 (1.11%)
	Death	5 (5.55%)	-	-

In patients who had ventricular outflow tract stenosis only at the valvular level, the majority of patients (83.34%) had severe aortic valve stenosis before surgery. Immediately after surgery 55.55% 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 2.7% showed severe stenosis. 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.

Table 4: 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	27 (67.50%)	20 (50%)
	Mild regurgitation	1 (2.50%)	12 (30%)
	Moderate regurgitation	5 (12.50%)	4 (8%)
	Severe regurgitation	7 (17.50%)	1 (2.5%)
	Death	-	3 (7.5%)
Aortic subvalvular stenosis	Without regurgitation	7 (46.66%)	5 (33.33%)
	Mild regurgitation	4 (26.66%)	9 (60%)
	Moderate regurgitation	3 (20%)	-
	Severe regurgitation	2 (13.34%)	1 (6.66%)
	Death	-	-
Aortic supravalvular stenosis	Without regurgitation	7 (70%)	7 (70%)
	Mild regurgitation	2(20%)	2 (20%)
	Moderate regurgitation	-	1 (10%)
	Severe regurgitation	1 (10%)	-
	Death	-	-
Aortic valvular + subvalvular stenosis	Without regurgitation	5 (33.33%)	2 (13.33%)
	Mild regurgitation	2 (13.33%)	10 (66.66%)
	Moderate regurgitation	2 (13.33%)	-
	Severe regurgitation	6 (40%)	3 (20%)
	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	50 (55.55%)	40 (44.44%)
	Mild regurgitation	10 (11.11%)	37 (41.11%)
	Moderate regurgitation	12 (13.33%)	4 (4.44%)
	Severe regurgitation	18 (20%)	5 (5.55%)
	Death	-	4 (4.44%)

Among the patients who had aortic valvular stenosis before surgery, 67.50% of the patients were without aortic regurgitation and almost 30% had moderate to severe regurgitation based on echocardiography. Almost half of the patients with aortic subvalvular stenosis were without aortic regurgitation; while post-surgical evaluations showed that 60% of the cases had developed 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.

Discussion

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. [3,16,17] 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. [18,19]

The mean ± standard deviation of the current age of the studied patients was 9.31±1.46 years. The minimum age of the patients was 1 month and the maximum was 15 years. 55 patients (55%) were male and 45 patients (45%) were female. The average weight of the patients was 28.24 ± 20.36 kg. The majority of patients had isolated valvular stenosis (46%), followed by isolated subvalvular stenosis. Combined conditions were fewer compared to the isolated conditions. In patients who had valvular aortic stenosis, commissurotomy was performed in 28 patients, Benthal surgery in 4 patients, and Aortic Valve Replacement (AVR) in 14 cases. Web resection surgery + myomectomy were performed in patients who had only subvalvular stenosis. Only those who had supravalvular stenosis underwent aortoplasty. In 7 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 9 patients. In patients with valvular stenosis and supravalvular stenosis, aortoplasty and commissurotomy were performed in 4 patients; while the other three cases underwent aortoplasty, aortoplasty and Bentall, and aortoplasty with AVR. 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%). [20,21]

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 1 patient. In patients who had ventricular outflow tract stenosis only at the valvular level, the majority of patients (83.34%) had severe aortic valve stenosis before surgery. Immediately after surgery 55.55% 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 2.7% showed severe stenosis. In a study conducted by Alexiou et al., between 1979 and 2000, two infants (2.1%) died immediately after surgery. [22] In other similar studies, either premature death rate was not reported, or the investigated population was scarce. [20,23] 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. [24]

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. Among the patients who had aortic valvular stenosis before surgery, 67.50% of the patients were without aortic regurgitation and almost 30% had moderate to severe regurgitation based on echocardiography. 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. [25]

Almost half of the patients with aortic subvalvular stenosis were without aortic regurgitation; while post-surgical evaluations showed that 60% of the cases had developed 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.

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.

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