

Coarctation and Its Surgical Repair: Mid to Long Term Follow Up in Children and Adults

Sudhir Adalti*, K.N. Bhosale**, Vijay Gupta***, Himani Pandya****

Abstract

Introduction: Coarctation of the aorta is a common defect that accounts for 5-8% of all congenital heart defects. Various surgical techniques are described in the literature. We hereby present our experience about surgical management of CoA and their long term outcome. *Materials and Methods:* This is a retrospective analysis of 70 patients operated for the aortic coarctation. Their age ranged from 6-41 years with mean age of 19.4 years. Demographic and clinical data was recorded for all the cases. All cases underwent surgical repair by various techniques like, Patch aortoplasty, Resection with end to end anastomosis and resection with interposition grafting. All patients were followed up for a period spanning from 4 to 12 years. *Results:* There were two early mortalities. Most common complication was recurrent laryngeal nerve injury and post-operative bleeding. Mean follow up was 7.8 years. 84.3% came for follow up. There was no late mortality. Thirty (43%) patients continued to be hypertensive. There was no recurrence in Coarctation. One patient who was operated with Dacron patch aortoplasty developed an aneurysm at the repair site. *Conclusion:* Asymptomatic patients with uncontrolled or refractory hypertension is the presenting symptoms of Coarctation. End to end anastomosis after complete excision of the coarcted segment in younger patients is the best approach and for older patients resection with interposition grafting is the preferred choice. Persistent postoperative hypertension is a concern and needs further analysis and evaluation. Postoperative CT /MRI is helpful in diagnosing complications of repair.

Keywords: Coarctation of the Aorta; Ventricular Septal Defect; Congenital Anomalies.

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Introduction

Coarctation of the aorta is one of the commonest congenital defect of thoracic aorta. The Coarctation of aorta is commonly associated with the bicuspid aortic valve, other associated cardiac malformations include mitral valve abnormality both stenosis and regurgitation or parachute mitral valve, patent ductus arteriosus and ventricular septal defect, etc. [1]. The surgical management is the treatment of choice, and various surgical techniques

have been reported in the literatures [2-4]. However, it has been fraught with controversy and thus there is no single method of choice for the surgical treatment of Coarctation of aorta. There have not been many changes in the surgical techniques already designed for coarctation of aorta three decades ago. The choice of surgical technique for CoA is determined not only by the type of lesion but also by factors like associated anomalies, the age at presentation and the surgeon's preference [5]. It is very difficult to label any surgical technique as ideal. Further, there are also controversies about timing of

repairing coarctation whether in the early and late age, usage of different suture materials, early and late postoperative outcome of surgery, etc. [6,7].

We hereby present our experience about surgical management of CoA and their long term outcome.

Materials and Methods

This is a retrospective study conducted at the Department of Cardiovascular and Thoracic Surgery of Grant Medical College and Sir J.J. Group of Hospitals, Mumbai. Seventy patients were operated from January 2000 to June, 2012 for the aortic Coarctation. All the patients in this study were above six years in age and their age ranged from 6 to 41 years with mean age of 19.4 years. There were 44 Male subjects and 26 female subjects.

Their clinical presentation varied from asymptomatic patients (28.6%) to Headache, breathlessness, easy fatigability, chest pain and palpitations. One patient presented with subarachnoid hemorrhage and one other presented with hypertensive heart failure. Overall, 88% of the patients were hypertensive at the time of presentation. Sixteen (22.9%) patients had associated cardiac anomalies. About 5.7% patients had hypoplasia of both the lower limbs.

All the patients were evaluated by checking their blood pressure in all the four limbs by an appropriate sized cuff and entered in their case record (Table-1) with a detailed clinical examination findings. The laboratory investigations included analysis of blood for hemoglobin content, cell counts, creatinine, electrolytes, coagulation profile, liver function tests and cardiac enzymes in some older patients. Any abnormal test result was taken into consideration before planning surgery. Chest X ray was done in all patients (Table-2). Typical rib notching depicting of collaterals was present in 40 (57.1%) patients. Every patient was subjected to electrocardiography. Further, both M-mode and two dimensional echocardiography was performed in all the patients followed by a Doppler study. Major emphasis during Echocardiography was on the coarcted aortic segment so as to look for any associated post stenotic dilatation, any valvular cardiac lesions or some other associated intracardiac / extracardiac lesions and LV dimensions. Prior to 2008, angiography and catheterization of aorta was performed preoperatively to make an assessment of the lesion and have a preliminary preoperative surgical plan. However, after 2008, CT aortography was preferentially used, it delineated the associated

cardiac lesion(s), if any and the pressure gradient across the Coarctation was also calculated. The patients were planned and prepared for surgery once the diagnosis of Coarctation was confirmed. Associated cardiac anomalies were Aortic valve disease in 8 (11.4%), Patent ductus arteriosus 6 (8.5%) and ventricular septal defect in 2 (2.8%).

Operative Procedure

Prior to surgery, nature of the disease and the surgical procedure was explained to the patient and their respective attendants and a valid consent was obtained. Antihypertensive therapy, especially ACE-inhibitors was continued till one day before the surgery. Preoperatively, mild sedation in the form of diazepam, 1 mg/kg body weight was given to all the patients including the prophylactic antibiotic in the form of injection Ceftriaxone 35 mg/kg body weight two hours before surgery.

Under general anesthesia, using a double lumen endotracheal tube, one peripheral venous line, one central venous line and one peripheral arterial line usually on the right side were secured. After usual sterile draping a left posterolateral thoracotomy was performed through 4th intercostal space. The high pressure collateral vessels in the chest wall were ligated with fine silk sutures. Pleural cavity was entered and the anatomy of aortic arch, descending thoracic aorta and their branches (if seen) was assessed. Mediastinal pleura were dissected off from the first three centimeters of left subclavian artery down to the upper descending thoracic aorta including coarcted segment and the part distal to it. Due care was taken to prevent injury to dilated and tortuous aneurysmal intercostal arteries. First 3 centimeters of left subclavian artery was always mobilized and looped on a thick cotton tape. The aorta was mobilized and looped on the two sides of coarctation. Any bleeding or lymphatic injury was stopped using fine absorbable sutures. The site and type of coarctation was assessed and the type of correction planned. Whole of the above procedure was performed at a systolic blood pressure of 70-90 mmHg.

Two 4.0 prolene purse-string sutures, one each were taken proximal and distal to the coarctation and were used for measuring the preoperative pressure gradient across the coarcted portion under general anesthesia using the invasive Spacelab and Datex monitors. The two pressures and the gradient across them were noted and recorded (Table 2). The aorta was cross clamped according to the type of operation to be performed saving as many intercostal

arteries as possible. Various surgical techniques which were used for Coarctation were: Patch aortoplasty, Resection with end to end anastomosis, Jump graft (Left subclavian artery to Descending thoracic aorta), Ascending aorta to descending aorta

bypass (exposing the descending thoracic aorta through the posterior pericardium and placing the graft around the left margin of the heart), Resection with interposition grafting. Operative findings and procedures performed are showed in Table 3 & 4.

Table 1: X-ray chest PA view findings

Findings	No. of Patients	Percentage (N=70)
Cardiomegaly	44	62.8
Rib notching	40	57.1
Prominent left Subclavian artery	20	28.6
Pulmonary congestion	20	28.6

Table 2: Pressure Gradient across two ends of coarctation (in mmHg)

Pressure Range	No. of Patients	Percentage (N=70)
20 - 40	0	0
40 - 60	12	17
60 - 80	38	54.3
80 - 100	12	17
> 100	8	11.4

Table 3: Operative findings

Operative Findings	No. of Patients	Percentage
Collateral Arterial Circulation In Chest Wall	68	97
Tortuous And Dilated Intercostal Arteries	68	97
Dilated Distal Aorta	68	97
Short And Curve Coarcted Segment	56	80
Dilated Left Subclavian Origin	38	54
Patent Ductus Arteriosus	6	9
Long Hypoplasticcoarcted Segment	4	5.7
Left Subclavian Artery Narrow At The Base	4	5.7
Normal Distal Aorta	2	2.8
Left Subclavian Artery Arising Distal To Coarctation	2	2.8

Table 4: Age at operation, method of correction

Age in years	Patch aortoplasty	End to end anastomosis	Jump graft	Ascending aorta to descending thoracic aorta	Interposition graft	Total
6 - 10	0	6	0	0	0	6
11 - 15	4	4	0	0	0	8
16 - 20	10	0	2	0	8	20
21 - 30	13	0	2	2	9	26
31 - 50	8	0	0	2	0	10
Total	35	10	4	4	17	70

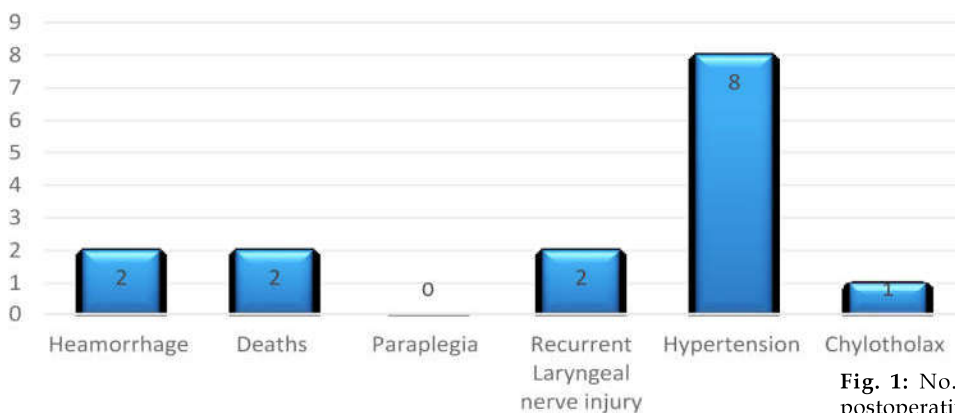


Fig. 1: No. of patients who suffered postoperative complications

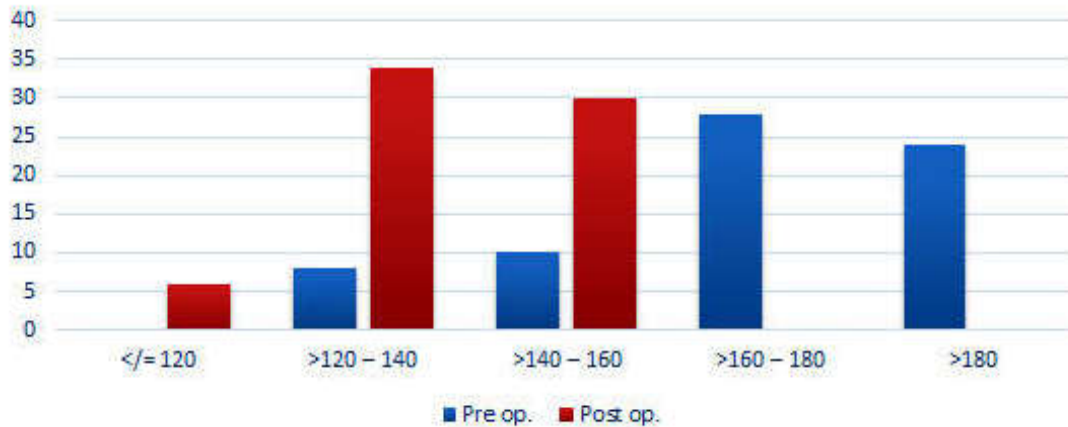


Fig. 2: Blood pressure (in Hg) comparison pre-op. with post-op status

Patch aortoplasty was by far the most common method used in 35 (50%) patients followed by resection with interposition grafting 16 (23%). Resection with end to end anastomosis was done in 10 (14%) patients and all of them were below fifteen years of age. Jump graft (Left subclavian artery to Descending thoracic aorta) was done in five (8.5%) patients who had long coarct segment covered with thin walled collaterals. Ascending aorta to descending aorta bypass was done in 3 (4.2%) patients who had co-existent severe aortic valve disease which necessitated midline approach.

Results

Most common complication (Figure 1) was recurrent laryngeal nerve injury and post-operative bleeding. No patient developed paraplegia indicating that there was no spinal cord ischemic insult. One patient developed chylothorax. Two postoperative deaths occurred. One young female had presented with subarachnoid hemorrhage and later succumbed to complications secondary to long term ventilation. Another boy of six years of age suffering from coexistent VSD with PDA could not be weaned off from the ventilator and died of ventilator associated pneumonia on 8th postoperative day.

Post-operatively a fall in blood pressure (BP) was noticeable in all patients. After the surgery, six (8.5%) patients became normotensive. The maximum BP recorded in this group was below 160 mmHg, with 34 (48.5%) patients had BP between 120-140 mmHg and 30 (42.8%) patients had BP between 140-160 mmHg. Thus, none had BP above 160 mmHg, whereas, prior to Surgery 52 (74.2%) had BP above 160 mmHg. (Figure 2).

The follow up period ranged from 4 to 12 years and 59 patients were followed. 8 patients were lost to the follow up. Mean follow up was 7.8 years. There was no late mortality. Thirty (43%) patients continued to be hypertensive. Although none of them were uncontrolled and all of them had SBP less than 160 mmHg. It has been reported that in post-operative period even after the correction of CoA there can be persistent hypertension. In this study 64 (91.4%) patients remained hypertensive postoperatively. However, after long-term follow up only 30 (43%) remained hypertensive. There was no recurrence in Coarctation. One patient who was operated with Dacron patch aortoplasty developed an aneurysm at the repair site.

Discussion

This study explores outcome of surgical repair of coarctation of aorta, about the relief, recurrence and complication rate. Lifelong follow up of CoA has been indicated for the disease being a type of diffuse aortopathy whereby mere correction of CoA should not be considered as complete cure [8]. In the present study, male to female ratio was approximately 3:2 with patient predominance in 2nd and 3rd decades of life. The age ranged from 6 - 41 years with a mean age of 19.4 years. The preponderance of CoA in males has been previously reported 1,8, and 21. The presentation of patients of CoA occurs in bimodal distribution [22]. There is a group that presents in the first week of life with circulatory symptoms requiring timely diagnosis and management. The other group consists of infants, children or older with CoA but can essentially be asymptomatic. The clinical manifestations, management and outcome are quite different in these two groups. In this study,

a number of patients belonged to the latter group i.e. asymptomatic group [8]. The asymptomatic group was also studied by Fraser, RS et al and W.J. Wells [9, 10]. Many of these patients due to hypertension develop increased left ventricular after-load. This results in certain symptoms and our patients presented to the hospital with these symptoms, viz. breathlessness, easy fatiguability, chestpain, headache and palpitations. They were referred by the physician for evaluation of a precordial murmur or as young hypertensives [11]. E.A. Shine Bourne [12] has observed a similar presentation of patients above 5 years of age. Some of the patients develop extreme form of symptomatology due to hypertension such as hypertensive encephalopathy, even intracranial hemorrhage [8]. In our cohort, presentation as subarachnoid haemorrhage was seen in a young female who was admitted to the Internal Medicine department of our hospital. Her blood pressure was very high (210/140 mmHg) in the upper limbs which made her a coarctation suspect. Another patient presented with severe hypertension and left ventricular failure, though heart failure is relatively rare in cases with late presentation [1].

Hemodynamic features on Physical Examination and Gradient across the Coarctation Segment:

As mentioned previously, all our (100%) patients had upper limb hypertension (differential pressure between brachial and popliteal arteries was high) and all of them also had radio-femoral delay. Both are the pathognomic features of CoA. Further, in our study, per-operatively, we calculated the gradient across the coarctation segment which ranged from 48 to 110 mmHg. A peak-to-peak systolic pressure gradient more than 20 mmHg across aortic coarctation is generally required for intervention [1]. All our patients had a peak-to-peak systolic pressure gradient more than 20 mmHg thus, making mandatory to operate all the cases of this cohort. Whereas, postoperatively, the significant gradient was noted only in 7 (10%) patients. Out of these patients, two had underwent subclavian artery jump graft, two underwent resection with interposition grafting, one underwent end to end anastomosis and two underwent patch aortoplasty.

Imaging Findings

Due to a variety of associated cardiac anomalies the cases of coarctation should be evaluated with vascular imaging modalities [1]. The use of newer imaging techniques like digital subtraction angiography, Intravascular ultrasound and other dynamic scanning have helped to improve the

outcome of complex-Coarctation which are associated with intracardiac defects due to pre-operative detection and appropriate corrective management.

Referring to Table 1, on X-ray chest PA view cardiomegaly was present in 44 (62.8%) patients. Cardiomegaly is known to be present in patients of CoA presenting late, due to hypertension. Rib notching was also noticed in 40 (57.1%) of the patients. Rib notching is not seen in infants because the collateral circulation is not yet well developed but it is developed in later life and further it is rare below the age of five. Rib notching occurs due to erosion of the inferior surfaces of the posterior ribs by dilated and tortuous intercostal arteries [13].

Associated congenital cardiac defect on echocardiography 8 (11.4%) cases of AVD, 6 (8.5%) cases of PDA and 2 cases (2.8%) of PDA were detected. The occurrence of associated cardiac anomalies as listed above are widely reported in literature (1,8,13-18). The cases with isolated CoA are labelled as Simple CoA and those which are with associated cardiac anomalies are called Complex CoA. In our study there were in all 16 (22.8%) cases were of Complex CoA.

Comparison of Surgical Techniques –our Experience

The choice of a particular surgical technique for the treatment of aortic Coarctation is determined not only by the type of Coarctation lesion but also by factors such as the presence of associated anomalies, the age at presentation and not the least, the surgeon's preference. It is therefore very difficult to identify an "ideal" surgical technique. However, objective comparisons can be made based on such determinants as the rate of recoarctation and freedom from re-intervention. Harska et. al. performed [3,19,20] a study of 201 patients undergoing surgery for Coarctation of the aorta over a period of ten years. 139 of these patients had simple or isolated Coarctation, 35 had Coarctation with ventricular septal defects (VSD), while 27 patients had Coarctation with complex intra-cardiac anomalies including hypoplastic left heart syndrome, transposition of the great arteries (TGA) and Shone syndrome. On the whole, 19 cases of recoarctation were recorded, representing 10% of all operated patients. In our series, we did not find recoarctation. Probably, neonates are more prone for recoarctation after end to end anastomosis [3,21]. One of our patient who was operated with Dacron Patch Aortoplasty At 43 years of age had developed an aneurysm at the repair site. He had persistent

hypertension in spite of vigorous antihypertensive therapy [19]. Carl Backer used PTFE for patch aortoplasty and reported no aneurysm over a follow up of 12 years [20]. Marc Cohen has mentioned in his study of 646 patients, that six patients were reoperated for aortic aneurysm formation while as 7% of patients died during long term follow up by rupture of aortic aneurysms [16]. Other investigators have reported that on long term follow-up of surgical outcome of CoA, the patients present with re-Coarctation, pseudo aneurysms. None of the cases in this study developed these complications.

Immediate Post-Op Course

No patient developed paraplegia indicating that there was no spinal cord ischemic insult. It is reported that in older patients due to development of collateral circulation there is very low risk of spinal cord ischemic injury if aortic cross-clamping is employed during the surgery of CoA. All our patients were above 6 years in age [8]. Further, per-operatively also 68 (97%) of our patients had evident collateral arterial circulation (Table 3).

There were two post-operative deaths but both of them were not related to cardiovascular system. A young boy of 6 years suffering from coexistent VSD with PDA could not be weaned off the ventilator as he had severe respiratory acidosis with poor myocardial reserve and died of bilateral lung consolidation on 8th postoperative day. Another death happened in a 32 year old lady who had to be re-explored for bleeding and later on died because of VAP followed by sepsis.

Fate of Blood Pressure in Immediate Post-Op Period

As the population of patients was comprising of older children or adults only, high blood pressure in upper limbs was seen in all our patients. After the surgery, six (8.5%) patients became normotensive. Majority of our patients had significant drop in proximal hypertension and distal perfusion improved quite markedly but their BP still remained in the hypertensive range.

There was an obvious decrease in the gradient between upper and lower limbs. The maximum BP recorded in this group was below 160 mmHg, with 34 (48.5%) patients having BP between 120-140 mmHg and 30 (42.8%) patients had BP between 140-160 mmHg. Thus, none had BP above 160 mmHg, whereas, prior to Surgery 52 (74.2%) had BP above 160 mmHg. [12]. Those patients who remained hypertensive, for them antihypertensive therapy was

continued in the postoperative follow up period. David Lerburg et al [14] reports a study of operating adult Coarctation and has observed that almost 50% of his patients operated at one year of age were discharged with significant hypertension.

A.P. Kappitein et al 1991 [15] have observed hypertension in 49% of patients after surgery. Similar observations have been made by Marc Cohen et al (1989) [16] (44% hypertension in the immediate postoperative period). A Earley and associates (1980) [17] measured blood pressure in a group of patients with Coarctation of aorta before and after surgery. Their 45% patients had hypertension after surgery also. Shine Bourne also reported persistence of hypertension after surgery [12] and also reported by others [14,15,18].

Findings on Long-term Follow up

The study included patient who were operated between years 2000 to 2012. Accordingly, the follow up period ranged from 4 to 12 years and 59 patients were followed. 8 patients were lost to the follow up.

Thirty (43%) patients continued to be hypertensive. Although none of them were uncontrolled and all of them had SBP less than 160 mmHg (Figure 2). It has been reported that in post-operative period even after the correction of CoA there can be persistent hypertension. In this study, 64 (91.4%) patients remained hypertensive postoperatively. However, after long-term follow up only 30 (43%) remained hypertensive. It has been hypothesized that in post-op period the hypertension may persist even after correction of CoA due to increased sympathetic activity [8]. This was also the case in many of our patients. Higher age at the time of surgical correction explains high incidence of postoperative hypertension [23].

However, on follow-up only less than of these remained hypertensive. This finding gives some credence to the above hypothesis.

Thomas Kimball et al (1994) noticed persistent hypertension in patients operated beyond 12 years of age. Thus, like as reported in other studies, on follow up many of the patients who were hypertensive prior to the correction of Coarctation remained hypertensive. All of our patients were above 5 years in age. In the experience of Shine Bourne et. al. If the correction of CoA is performed after 1 year age, there may be persistent systemic hypertension despite relief of obstruction [12] and study from Mayo clinic reports 9 years as cut off age [21].

Limitations

Patients were enrolled retrospectively, thus this study has an inherited selection bias.

We have lost 8 patients to follow-up as we were not able to contact them.

Single surgeon single center experience, hence results cannot be generalized.

Conclusion

Asymptomatic patients with uncontrolled or refractory hypertension coarctation of aorta should be suspected and evaluated. Echocardiography is the preferred modality for the diagnosis. Excision and end to end anastomosis in younger patients is the best approach. For older patients, resection with interposition grafting is the preferred choice now a days, but we have shown equally good results with patch aortoplasty. During long term follow up, examination of pulse and recording of blood pressure in both upper and lower limb should be done routinely. X-ray PA view, chest should also be done annually. Echocardiography and CT Scan/MRI should be used to delineate a complication postoperatively, if suspected. Although LV hypertrophy is present, persistent postoperative hypertension is a concern and needs further analysis and evaluation. Awaiting final answer for the cause of postoperative hypertension, antihypertensive therapy is to be continued whenever indicated.

Conflict of Interest Statement: None declared.

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