

Coarctation of aorta in paediatric age group: mode of presentation, early and intermediate outcomes of interventional treatment.

An experience from Ibn Al-Bitar Hospital in Baghdad

Munaf Jarallah Yaseen¹, Sanaa Noori Mohammed Hasan², Amer Jabar Ali³

¹ MBCHB, DCH, FICMS (paediatric), FICMS (cardiology) interventional cardiologist at Iraqi Centre for Heart Diseases, Medical City, Baghdad, Iraq.

² MBCHB, DCH, FICMS (paediatric). Paediatrician, Child Welfare Teaching Hospital, Medical city, Baghdad, Iraq.

³ MBCHB, FICMS (paediatric). Paediatrician, Child Welfare Teaching Hospital, Medical city, Baghdad, Iraq.

Corresponding Author:

Munaf Jarallah Yaseen.
Iraqi Centre for Heart Diseases, Medical City, Baghdad, Iraq.
E mail:
iraqiii1978@gmail.com

ABSTRACT

INTRODUCTION: Although rare, coarctation of aorta is a treatable cause of hypertension, transcatheter dilatation by balloon with or without stent are a well-known treatment strategy.

OBJECTIVE: This study is designed to review the mode of presentation of patients with coarctation of aorta referred to Ibn Al-Bitar Hospital and to compare the immediate and intermediate outcomes of transcatheter balloon angioplasty with and without stenting.

METHODS: A case series study conducted at Ibn Al-Bitar Cardiac Centre from February 2009 to February 2010. Forty-five patients that diagnosed by echocardiography as coarctation of aorta. Their demographic features, way of presentation, family history of congenital heart diseases and other echocardiographic associated lesions were identified. Forty-two patients have undergone intervention. Either balloon dilation with or without stenting. Their intra-operative complications were reported in addition to immediate and intermediate success rates.

RESULTS: Boys are predominant than girls, their age ranged from 1 month to 18 years. Undiagnosed cardiac murmur is the commonest cause of referral. More than 50 % (26 patient) of cases were simple coarctation of aorta. Intimal tear was reported in 4 patients (9.52%) while aortic aneurysm, transient complete heart block, and death were reported in one patient for each. Both interventions have an equivalent good success rate immediately after the intervention; however, stenting has maintained better success rate after 6 months of follow up.

CONCLUSION: Patients with coarctation of aorta is suffering from delaying in referral to cardiologist and this is reflecting on increasing complication such as left ventricular dysfunction. Overall complication rate of both interventions is low. Balloon angioplasty and stent has equivalent immediate success rates; however, intermediate success rate is much better with stenting.

Key words: Coarctation of aorta, balloon angioplasty, stent, paediatric.

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INTRODUCTION

Coarctation of aorta (COA) is a discrete stenosis of the proximal thoracic aorta just distal to the origin of the left subclavian artery. This stenosis may involve a long segment with aortic arch hypoplasia and it may involve abdominal aorta.¹ It was first described by Prussian anatomist Johann Friederich Meckel in 1760.² It represents 8-10% of all congenital heart diseases. It is more prevalent in males in a ratio of 3:1. Balloon angioplasty has been used for coarctation of the aorta since 1982.³

Previously, COA is classified⁴ into three types

according to the site of coarctation in relation to ductus arteriosus: preductal that commonly associated with turner syndrome,^{5,6} ductal, and post ductal. Coarctation of aorta is considered simple if it is not associated with other congenital heart diseases (CHD) apart from (PDA) and this represents 52% of all cases of coarctation; Otherwise, It is called complex which represents the remaining 48% of the cases.⁷

Patients are usually presented in one of three patterns:⁸

1. Catastrophic illness, heart failure and sudden shock when ductus arteriosus closes in infant at age of 8-10 days.
2. Hypertension presented in later childhood and adolescence.
3. Cardiac murmurs that may be discovered incidentally.

About 20-30% of all patients with COA develop congestive heart failure by 3 months. If it is undetected or untreated, early death may result from congestive heart failure and renal shut down. At age of 46 years, 76% of patients with COA dies. Heart failure is the commonest cause of death (28%), followed by rupture aorta (26%), SBE secondary to stenotic bicuspid aortic valve (18%), and rupture berry's aneurysm (12%).⁸

Patients with coarctation of aorta can be safely engaged in most physical activities; however, heavy isometric exercises such as power weight lifting may be prohibited especially in the presence of pre-stenotic dilatation of the aorta.⁹

In patient with COA, electrocardiography is completely normal in about 20% of patients. Presence of left ventricular hypertrophy and left ventricular strain pattern is highly suggestive of associated aortic stenosis.⁹ chest X ray may show cardiomegaly, rib notching, abnormal contour of the aortic arch, pre-stenotic dilatation that involves the left subclavian artery, and post-stenotic dilation of the descending aorta.² Two dimensional echocardiography provides accurate noninvasive assessment of coarctation.¹⁰ MRI and CT angiography are suitable for patients who needs serial imaging i.e. before and after surgery or balloon. MRI study can even assess the pressure gradient across coarctation.^{11,12}

Adult hypertension or incidental cardiac murmurs are the commonest causes of referral of patients with COA to cardiologist, while only in 14 % of them the diagnosis is established by paediatrician before referral.¹³

COA is one of the treatable congenital heart diseases and one of the rare treatable cause of hypertension. Outcome is dependable on early referral of patients to tertiary centres. This study is designed to review the mode of presentation of patients with COA referred to Ibn Al-Bitar Hospital and to compare the immediate and intermediate outcomes of transcatheter balloon angioplasty with and without stenting.

METHODS

Settings and study design: A descriptive case series study conducted on 45 patients who was newly diagnosed as COA at Ibn Al-Bitar Cardiac Centre in Baghdad from February 2009 to February 2010.

Ethical issues: the study was approved by the administration of the centre. Participants' families were informed about the procedure, its benefits and potential complications, and their written informed consent were requested prior to enrolment in the study.

Definition of the case, inclusion and exclusion criteria: Patients who were below age of 18 years and diagnosed as COA at Ibn Al-Bitar Cardiac Centre were enrolled in this study. The diagnosis was confirmed in all patients by the presence of a pressure gradient of > 20 mm Hg across descending aorta by echocardiography. The total number of patients who fulfilled the above criteria were 45 patients. Three of them have refused to do transcatheter intervention in our centre and were excluded from our calculation of immediate and intermediate outcomes and immediate complications of the intervention.

Outcomes and procedures: Complete medical history was taken from the family with special emphasis on cause of referral to the hospital, family history of COA and or other congenital heart diseases in first-degree relatives. Thorough physical examination was carried out. The diagnosis was made by continuous mode doppler echocardiography by the presence of a pressure gradient of > 20 mm Hg across descending aorta. Demographic features, ways of presentation, family history of COA and other congenital heart diseases, and presence of associated congenital heart diseases were reported by echocardiography for all 45 patients. **Forty-two** of them have undergone catheterization. **Thirty-two** have received balloon dilation and **ten** have received balloon dilation with stenting. Interventions were chosen according to patients' age and their body weights. Generally, we used stent in children age >6 years and/ or body weight > 26 Kg. Pressure gradient across the coarctated segment were measured through catheterization before the intervention and immediately thereafter. Mean change in pressure gradient before and after the intervention of those underwent balloon angioplasty alone were compared to those underwent

balloon dilation with stent. We followed up all patients for 6 month and pressure gradient is measured again by echocardiography and mean pressure gradient were also compared between two groups. We reported the following immediate intra-operative complications: Intimal tear, aneurysm, heart block, death.

Catheterization: Before each procedure, the patient's parents signed a written informed consent. Patients were instructed to cease oral intake a minimum of 4 hours before the scheduled procedure. Maintenance fluids were administered through an intravenous line placed at the time of presentation to the catheterization laboratory, premedication with midazolam afforded sufficient sedation for all patients before transfer to the catheterization laboratory. Under general anaesthesia femoral venous and arterial access are established, after that patients received intravenous heparin in a dose of 150 IU/Kg (maximum 5000 IU) to maintain an activated clotting time >200 seconds; with an additional dose of 75 IU/Kg if the procedure duration exceeded two hours. Baseline haemodynamics including direct measurement of the ventricular end-diastolic pressure through a pig-tail catheter were performed. The peak-to-peak systolic coarctation gradient was first estimated by direct pullback of an end-hole catheter, then confirmed (if possible) by simultaneous ascending and descending aortic pressures, with consideration of any premeasured standing wave. Angiographic lumen diameter was determined by measuring the intraluminal contrast column and correcting for magnification. We used Tyshak balloon and the balloon diameter is chosen to equal the diameter of the aortic isthmus with inflation time of less than 10 seconds; inflations can be repeated two to four times. If necessary for optimal gradient relief a larger balloon may be used but should not exceed the diameter of the aorta at the level of the diaphragm. Gradient measurements were repeated after the intervention.

Stent implantation technique, as dictated by the balloon/stent profile, introduced in the femoral artery was advanced across the lesion over a stiff guide wire. The tip of the wire was positioned in either the ascending aorta or in the right subclavian artery. The sheath and wire then served as a delivery system for the balloon and stent. A side-arm adaptor on the sheath also permitted angiography to refine stent position before deployment. In general, we chose

the shortest stent that could safely straddle the stenotic area in an attempt to avoid creating long, noncompliant segments of aortic wall. If the first stent did not sufficiently span the angiographic area of narrowing a second stent was placed. In several cases the standard implantation technique was modified to minimize the risk of stent migration during balloon inflation. With the stent in position, but still within the sheath, partial retraction of the sheath exposed one end of the balloon and stent. A small volume (1 to 2 mL) inflation caused the exposed portion of the balloon to expand slightly, thereby fixing the stent during subsequent full inflation.

We used two doses of amoxicillin as antimicrobial prophylaxis for all patients in a dose of 50 mg per Kilogram body weight. The first dose was given immediately before placement of the stent and the other dose was 16 hours thereafter. Patient who has a history of sensitivity to penicillin was given vancomycin in a dose of 20 mg per Kilogram body weight.¹³ The patients were observed overnight and received heparin infusions at 15 to 20 IU/kg per hour. Before discharge home, all patients underwent clinical evaluation and echocardiography. Chest X ray was reversed only for those treated by with stents. Aspirin in a dose of 40-80 mg per day was prescribed for 3-6 months for all patients who were treated by balloon dilation with stent.

Definitions: For the sake of this study, the following terms were defined as follows:

1. Significant coarctation means systolic pressure gradient across the coarctated segment of >20 mmHg.
2. Discrete coarctation: the coarctated segment is shelf like and doesn't extend beyond the ligamentum arteriosum.
3. Tubular coarctation: long and diffusely narrowed segment that extend beyond the ligamentum arteriosum.²
4. Successful intervention: peak-to-peak systolic pressure gradient across the coarctated segment < 20 mm Hg after intervention.
5. LV dysfunction: echocardiographic measurement of LVEF of <54% or by catheterization poor global LV contractility.
6. Tear or minor flap: thin mobile membrane extending into the wall over no more than a quarter of the circumference of aorta. If extend over more than a quarter of the circumference of aorta then defined as a major flap.²
7. Restenosis: increase in pressure gradient across the coarctated segment to > 20mm Hg

Table 1: Causes of referral of patients to Ibn Al-Bitar Hospital for Cardiac Surgery.

Cause of referral	Number of patients (%)
Cardiac murmurs	33 (73.33)
Unexplained hypertension	6 (13.33)
LV dysfunction	4 (8.88)
Diagnosis of COA before referral	2 (4.44)

Table 2: Patients with family history of congenital heart disease in first degree relatives.

Congenital heart disease	Number with family history (%)
VSD	2 (4.44)
ASD	1 (2.22)
AS	1 (2.22)
Total	4 (8.88)

VSD: ventricular septal defect, ASD: atrial septal defect, AS: aortic stenosis

after initial successful intervention.

Statistical analysis: All data were coded and entered to computer by using statistical package for social science SPSS-17. Summarization of data done by using numbers, percents and mean±SD. Association between different variables was measured by using Chi-square test. Difference between pressure gradients before and after interventions was measured by using t-test. P value ≤ 0.05 was considered as a level of significance.

RESULTS

The total number of patients included in this study was 45 patients; 32 (71.11%) males and 13 (28.9%) females. The male to female ratio was 2.4:1. The youngest age at diagnosis was one month and the eldest was 18 years (216 months). The mean age of the studied sample at time of diagnoses (in months) was 51.27±65.7.

Table 1 shows the causes of referring patients with COA, while **table 2** reported the prevalence of some congenital heart diseases in the first degree relatives of patients with COA. By echocardiography, 26 patients (57.8%) with simple COA while 19 (22.2) were complex. **Table 3** shows the associated congenital heart diseases.

Angiography of aorta has shown that 34 (80.95%) patients have discrete coarctated segment, while 8 (19.04%) patients has shown tubular segment. **Table 4** shows the haemodynamic data pre and post intervention. Overall immediate intra-operative complications are shown in **table 5**. **Table 6 and 7** show the comparison of immediate and intermediate success rate be-

Table 3: Associated cardiac defects as diagnosed by ECHO.

Cardiac defect	Number of patients (%)
Bicuspid AV	12 (26.66)
VSD	9 (20)
MR	9 (20)
AR	7 (15.55)
ASD	6 (13.33)
AS	5 (11.11)
PDA	3 (6.66)
MS	3 (6.66)

AV: aortic valve, VSD: ventricular septal defect, MR: Mitral regurgitation, AR: Aortic regurgitation, ASD: atrial septal defect, AS: aortic stenosis, PDA: Patent ductus arteriosus, MS: Mitral stenosis

tween balloon dilation a lone and with stent.

DISCUSSION

In this study, male to female ratio was 2.4:1 which is similar to the ratio reported by other worker.¹³ However, an extreme male predominance (8:1) was reported by Ozawa et al.¹⁴ Patients' age at diagnosis ranged from 1 month to 18 years. Ozawa et al¹⁴ reported an age range of 2.5 months to 5 years, and Carline Ovaert et al¹⁵ has reported a range of 6 months to 5.8 years. This clearly shows a delay in the diagnosis and referral of patients with COA, which may be attributed to the lack of awareness about the disease at primary health care facilities.

The most common cause of referral to our hospital is, as expected, cardiac murmurs 33 (73.33%) of cases. While only 2 (4.44 %) of our patients were already diagnosed as COA on referral. It has been stated that diagnosis of COA before referral was 14 %.¹³ Our low percentage of diagnosis before referral may be due to lack of awareness among healthcare providers at primary and secondary healthcare centres, unavailability of diagnostic facility, and lack of expert personnel in congenital heart diseases.

Delay of diagnosis and seeking medical help end with LV dysfunction. Eight patients (8.88 %) were referred as LV dysfunction in our study. Development of LV dysfunction, especially in complex coarctation, masks cardiac murmurs and renders it difficult to diagnose COA unless low threshold for the diagnosis is always maintained.¹⁶

Shunt lesions (VSD and PDA) are the commonest types of left to right shunts associated with COA found in 26% of this study; a finding which is similar to that of Al-Kubaisy¹⁷ and Al-

Table 4: Haemodynamic data of cardiac catheterization before and immediately after the intervention.

Catheterization haemodynamic Data		Systolic	Diastolic	Mean	Systolic PG
Before the intervention	Ascending aortic pressure (mean \pm SD)	135.12 \pm 42.2	71.9 \pm 27.94	90.6 \pm 29.22	53.57 \pm 19.98
	Descending aortic pressure (mean \pm SD)	81.31 \pm 29.26	59.76 \pm 23.00	70.48 \pm 26.66	
Immediately after the intervention	Ascending aortic pressure (mean \pm SD)	91.31 \pm 25.50	60.48 \pm 23.45	76.19 \pm 22.46	7.86 \pm 6.73
	Descending aortic pressure (mean \pm SD)	83.21 \pm 22.97	53.45 \pm 19.11	65.71 \pm 18.27	

PG: Pressure gradient, SD: standard Deviation

Table 5: Complications developed during cardiac catheterization.

Complication	Number of patients (%)
Intimal tear	4 (9.52)
Aneurysm	1 (2.38)
Complete Heart block	1 (2.38)
Death	1 (2.38)
Total	7 (16.66)

Amiry.¹⁸

We found that 26 patients (57.8%) have simple COA, this is similar to other two Iraqi studies that reported simple COA to be 52%¹⁷ and 49 %¹⁸ respectively. In our series of cases, bicuspid aortic valve was the commonest associated cardiac lesion with COA seen in 12 patients (26.66%). Although Moss¹³ has also shown that bicuspid aortic valve is the commonest associated lesion in COA but he reported it in 80 % of his sample. This huge difference in detecting Bicuspid aortic valve may be attributed to the fact that it is easily missed by echocardiographer unless high level of expectation is maintained. Mitral valve lesions (stenosis and regurgitation) are seen in 12 (26.66%) patients of our sample. Perloff has reported nearly similar result.²

Angiographically discrete coarctation seen in 34 patients (80.95%). the same finding was reported by Moss.¹³

With regards to immediate operative complications, intimal tear has happened in 2 (9.52%) patients. Both of them were seen exclusively in balloon dilation and never happened with

stenting. In Al-Amiry study,¹⁸ intimal tears was reported in 34 % of the patients with balloon dilation and 28% of patients underwent stenting. The obvious difference can be referred to the type of balloon used in both studies. We have used Tyshak balloon while Al-Amiry has used Zemid balloon and it is well-known that Tyshak balloon has a low bursting pressure than Zemid resulting in low incidence of intimal tear.

Only one patient (2.38%) has developed aneurysm after balloon dilation. This is less than that reported by Masood Sadiq.¹⁹ This difference can be explained by the fact that Sadiq has operated on older patients than ours, his range of age was 20-40 years. In addition, many of his patients had risk factors for atherosclerosis such as diabetes mellitus and smoking.

Transient complete heart block has developed in one patient who has severe uncontrolled hypertension and managed by stenting. Up to our effort in searching literature about this complication, we could not find a case of complete heart block in patients with COA who underwent balloon dilation with or without stenting. In our study, one patient has died on the table due to rupture dissecting aorta. This unfortunate and dreadful incident has occurred during stent placement. Many studies have shown similar sinister complication; Swan²⁰ has reported death in 0.7 % of his series, while Ovaert¹⁵ has reported it in 1%.

We defined success as peak-to-peak systolic pressure gradient across the coarctated seg-

Table 6: Response of haemodynamic parameters to interventions during cardiac catheterization

Intervention	Initial pressure gradient (mean \pm SD)	Post-interventional pressure gradient (mean \pm SD)	Success rate	P value
Balloon dilation	48.59 \pm 16.96	6.09 \pm 5.50	100%	0.003
Stenting	69.50 \pm 21.40	13.5 \pm 7.47	90%	0.002

Table 7: Intermediate outcome by ECHO follow up .

Intervention	Pressure gradient on follow up after 6 months (mean \pm SD)	Success rate	P value
Balloon dilation	31.41 \pm 11.59	62.5%	0.007
Stenting	15.5 \pm 3	88.88 %	0.0001

ment < 20mm Hg after intervention. Accordingly, we achieved initial success rate in 100 percent of those treated by balloon angioplasty, and in 90 % (9/10) of those treated by stent (one patient has died due to rupture aortic aneurysm). These results are better in comparison to Al-Amiri¹⁸ who reported 84 % and 88 % respectively, and they are comparable to the results of other studies.^{21,22,23,24,25}

On intermediate (six months after intervention follow up) follow up by echocardiography, restenosis was observed in 12 (37.5%) patients with balloon angioplasty. While restenosis was reported in one patient (11.11%) treated by stent. This observation is contradicting Al-Amiry¹⁸ who found no significant difference between balloon dilation and stenting on intermediate follow up. The superiority of the stent over balloon dilation can be explained by the fact that the stent are designed to oppose the elastic recoil of the stenotic vessel.

Despite the general agreement that stenting in paediatric age groups is indicated for children > 6 years of age and/or > 26 kg in weight, Hamdan et al had used stents for children 4 years old and/or 18 kg in weight and made advantage from the superiority of stents over balloon.²⁶

CONCLUSIONS

Patients with COA is suffering from delaying in referral to cardiologist and this is reflecting on increasing complication such as LV dysfunction. Overall complication rate of both interventions is low. Balloon angioplasty and stent has equivalent immediate success rates; however, intermediate success rate is much better with stenting.

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Abbreviation list: Aortic regurgitation (AR), atrial septal defect (ASD), aortic stenosis (AS), aortic valve (AV), Coarctation of aorta (COA), Congenital heart diseases (CHD), International unite (IU), Left ventricle (LV), Left ventricular ejection fraction (LVEF), Magnetic resonance image (MRI), Mitral regurgitation (MR), Mitral stenosis (MS), Patent ductus arteriosus (PDA), ventricular septal defect (VSD)

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