



Early Results of Balloon Angioplasty of Native Coarctation of Aorta Under 2 Years Old

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Abstract

Objective: The balloon angioplasty is a controversial procedure for the treatment of coarctation of the aorta (COA). This study determines the results of balloon angioplasty of native COA in infants.

Materials and Methods: Twenty-five subjects had undergone balloon angioplasty under drug-induced sedation using the retrograde technique through the femoral artery. In all the cases a Tyshak Mini balloon had been used. The patients had all been followed by thoracic echocardiography. Data were analyzed with SPSS 16.

Results: Twenty-five patients under 2 years of age with native COA, had undergone balloon angioplasty. The median age and weight of the subjects were 55 (12-700) days and 4860 ± 192 g respectively. Mean stenotic site diameter was 2.31 ± 0.58 mm before procedure and 5.41 ± 1.09 mm after procedure ($P < 0.001$). Mean pressure gradient before and after procedure was 34.48 ± 15.39 mm Hg and 5.84 ± 3.79 mm Hg respectively ($P < 0.001$). Early minor and major complications were seen at 16% and 24% of the cases respectively. The only short come complication was recurrent COA in 3 cases (12%). The final outcome of patients was recovery in 20 subjects (80%).

Conclusion: Based on the results of this study and reviewed studies, balloon angioplasty might be an alternative procedure in infants with native COA. However timely diagnosis and improvement in angioplasty techniques are necessary to improve the outcome.

Keywords: Angioplasty, Aortic Coarctation, Balloon, Infant

Introduction

Coarctation of aorta (COA) is seen in 6%-8% of patients suffering from congenital heart diseases. COA has been reported to be the fourth most prevalent heart disease in infants, requiring catheterization and surgery during the first year of life (1). Similar to other obstructive conditions of the left side, COA is more prevalent in males compared to females, with a male-to-female ratio ranging from 1.23 to 1.74 (1,2).

COA might occur in different parts of the aorta, including the ascending aorta, aortic arch or descending thoracic or abdominal aorta. In terms of anatomical, pathophysiologic and clinical manifestations, their treatment and prognosis are different. In the simple form, COA is usually seen as a discrete stenosis in the proximal portion of thoracic aorta in the vicinity of the arterial duct and might be accompanied by patent ductus arteriosus (PDA) or ventricular septal defect (VSD). In the complex type, it is usually associated with complex intra-cardiac anomalies and may affect a long segment of aorta and can be associated with aortic arch hypoplasia (the infantile type). Extra-cardiac vascular anomalies, such as innominate artery defects, collateral arterial circulation and aneurysm of the Circle of Willis are seen in a large number of patients with

COA. The pathophysiology of COA varies depending on the severity of the lesion and concurrence of other defects such as VSD/PDA and obstruction of the left ventricular outlet. Bicuspid aortic valve (BAV) is seen in up to 85% of patients with COA. Clinical manifestations of COA range from heart failure during infancy to asymptomatic systemic hypertension or cardiac murmur in older children and adults that are accidentally discovered during routine examination. Treatment options include surgery with thoracotomy and correction of stenosis using different techniques, in most cases by removing the constricted area and anastomosis of the 2 ends of the constricted area (end-to-end) and balloon angioplasty and percutaneous stenting. The long-term prognosis of this anomaly and its clinical outcomes vary considerably after treatment and are not always benign. In the majority of patients, the long-term prognosis is affected by the residual stenosis in the area, recurrence of stenosis, associated intracardiac defects and chronic hypertension. If COA is diagnosed during infancy, which is associated with the manifestations of heart failure, prompt treatment is necessary. After a short period of medical therapy, in order to stabilize the patient's hemodynamic status, definitive treatment with balloon angioplasty or surgery is necessary.

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Surgical repair of COA was reported for the first time in 1945 (3). The mortality rate after surgery in these infants ranges from 2% to 10% and the mortality rate is higher in cases associated with intra-cardiac defects (4-8). The postoperative complications in COA patients include paradoxical hypertensions, arteritis of mesenteric artery and intestinal ischemia (9), ischemia of the spinal cord and paralysis of extremities (10), injury to laryngeal and diaphragmatic nerves, chylothorax, hemorrhage and infection. Percutaneous balloon angioplasty is a less invasive option in comparison to the surgical repair of COA, which is used in patients with discrete COA. This procedure is an effective technique in recurrent COA after surgery; however, there is controversy over it as an initial treatment modality in native COA, which is attributed to the high success rate of the surgical technique in isolated native COA.

Balloon angioplasty for the treatment of COA has been used since 1982 (11) and different results have been reported in relation to its safety and effectiveness in patients with native COA (12-18).

The mid-term outcomes of balloon angioplasty are better in older children and adolescents compared to that in infants, similar to the surgical technique (14-17,19). A number of complications have been reported after balloon angioplasty for native COA. Mortality after infancy is rare but a mortality rate of 0.7% has been reported during infancy (12). The most frequent acute complication is the injury to the femoral artery, which is more common in infants under 12 months of age; however, its incidence has decreased after low-profile angioplasty catheters became available (20). Less common complications include hemorrhage from the femoral artery and cerebrovascular accidents. Paradoxical hypertension is uncommon after balloon angioplasty of COA (21).

Considering the results of different previous studies and the overall controversy over balloon angioplasty for native COA, the present retrospective study was undertaken to evaluate the outcomes of the technique in infants with native COA in Madani Heart Center in Tabriz.

Materials and Methods

The aim of this retrospective cohort study was to determine the recovery rate of vascular stenosis and pressure gradient and also the short and mid-term complications and recurrent stenosis and aneurysm of the aorta after balloon angioplasty of native COA in infants less than 2 years of age. The samples consisted of 25 consecutive patients which underwent percutaneous balloon angioplasty between May 2011 and December 2014 in Madani Heart Center in Tabriz, Iran. Balloon angioplasty was administered to all the patients, under drug-induced sedation, using the retrograde technique through the femoral artery. Subjects in a critical condition before the procedure underwent intra-tracheal intubation. In all the subjects, a Tyshak Mini balloon (Numed Company, Canada), measuring 6-10 mm in diameter and 20 mm in length, was used. Before and after the procedure, the pressure gradient

and the minimum diameter at the stenotic area was determined by aortography and recorded. After the procedure, aortography was repeated for the evaluation of aneurysm or dissection. The subjects were followed by thoracic echocardiography in an outpatient setting after discharge from the hospital and in case of recurrent stenosis of the aorta and an increase in the pressure gradient to over 40 mm Hg as determined by echocardiography or the presence of dilatation and/or decrease in the function of the left ventricle with any pressure gradient, the patient again underwent angiography and angioplasty.

The patients' data were extracted from their hospital files and analyzed with Kolmogorov-Smirnov, sample paired t test and Wilcoxon signed-rank tests, using SPSS 16.

Results

A total of 25 patients with COA, who had undergone balloon angioplasty, were evaluated; 20 patients (80%) were male and 5 (20%) were female. The median age of the subjects was 52 days, with minimum and maximum ages of 12 and 700 days, respectively. The mean weight of the infants was 4860 ± 192 g.

The most frequent chief complaint of the patients was respiratory distress along with poor feeding in 8 (32%) cases. Clinical examination of the patients revealed 16 cases (64%) of weak pulse along with cardiac murmur and pulmonary rales and 9 cases (36%) of weak pulse associated by cardiac murmur (Table 1).

Clinically 10 (40%) of the cases had cardiorespiratory failure who underwent endotracheal intubation and the other 15 (60%) were hemodynamically stable and had spontaneous breathing.

The COA was associated by VSD in 15 (60%), PDA in 12 (48%), atrial septal defect in 10 (40%), and shone complex in one (4%) of the case. Only 6 (24%) of the cases had isolated COA (Table 2). 18 (72%) had juxtaductal, 6 (24%) had preductal and 1 (4%) had postductal type COA.

Mean stenotic site diameter was 2.31 ± 0.58 mm before procedure and 5.41 ± 1.09 mm after procedure ($P < 0.001$). Mean pressure gradient before and after procedure was 34.48 ± 15.39 and 5.84 ± 3.79 mm Hg, respectively ($P < 0.001$).

The complications of balloon angioplasty were divided

Table 1. The Baseline Characteristics of 25 Infants With Coarctation of Aorta, Receiving Treatment With Balloon Angioplasty

Variable		
Sex, n(%)	Male	20 (80)
	Female	5 (20)
Age (day)	Min	12
	Median	55
	Max	700
Physical examination n(%)	Weak pulse + murmur	9 (36)
	Weak pulse + murmur + pulmonary rales	16 (64)
	Preductal	6 (24)
COA type, n(%)	Juxtaductal	18 (72)
	Postductal	1 (4)

Table 2. Associated Anomalies With COA

Anomalies	Number	%
ASD-PDA-COA	4	16.0
ASD-PDA-COA-VSD	3	12.0
ASD-VSD-COA	3	12.0
COA	6	24.0
SHONE-VSD-PDA	1	4.0
VSD-COA	4	16.0
VSD-PDA-COA	4	16.0
Total	25	100.0

Abbreviation: COA, coarctation of the aorta; PDA, patent ductus arteriosus; VSD, ventricular septal defect.

into early and late. In 3 cases (12%), the early complication was the thrombosis of femoral artery, 2 of which were treated by anticoagulative agents.

In 5 cases (20%) hemorrhage from the femoral artery necessitated transfusion of blood. Paradoxical hypertension was seen in 2 cases (8%). None of the patients developed aortic dissection or cerebrovascular accident. In 3 cases (12%) the late complication of recurrence of coarctation of aorta was detected after 7, 9 and 14 months; in 2 of these cases balloon angioplasty was repeated successfully, but in one case angioplasty was carried out surgically due to the bilateral obstruction of the femoral artery. Both patients who received antihypertensive therapy after the procedure received the drug during the follow-up period. None of the subjects cursed aneurysm, aortic dissection and endarteritis during the follow-up.

The patients were followed for a mean 16 months, ranging from a minimum of 7 days to a maximum of 45 months. The final outcomes of the patients were as follows: 20 cases (80%) were completely cured and were followed clinically; 4 cases (16%) died a few days after the procedure and one case (4%) suffered from bilateral obstruction of the femoral artery and needed surgical repair.

About correlation of the complications with the clinical status of the patients both early and late complications and deaths occurred in patients that were hemodynamically unstable and had been intubated before procedure. None of the deaths was related to the procedure technique directly, but had been due to severe hemodynamic instability and multi-organ failure.

Mean left ventricle ejection fraction in patients with and without complication was 29 ± 0.14 and $25 \pm 0.13\%$, respectively ($P=0.66$). None of the patients with stable hemodynamic cursed complication.

Discussion

Coarctation of aorta is one of the relatively common congenital anomalies of the cardiovascular system that might be manifested during infancy as heart failure and even cardiogenic shock. Unfortunately the initial clinical manifestations in infants are non-specific and mainly consist of tachypnea, poor feeding and restlessness, which result in delays in the correct diagnosis and therapeutic interventions. Such a situation was noted in 32% of the patients evaluated in the present study, resulting in the clinical misdiagnosis in 76% of the cases. All of the subjects had

weak pulse in the lower extremities along with heart murmur which can help in early diagnosis of this condition during clinical examination. In the majority of patients in this study, COA was accompanied by other cardiovascular anomalies.

For patients present with cardiopulmonary insufficiency and hemodynamic disturbances, the first therapeutic procedure is supportive, including use of oxygen, inotropic agents, diuretics and if necessary prostaglandin E1 and correction of disorders such as hypoglycemia, hypothermia, acidosis and anemia. After stabilizing the patient's hemodynamic status, definitive repair should be undertaken. Surgical repair during infancy is associated with a high mortality rate, especially when the condition is associated with intra-cardiac anomalies (4-8,22,23). However, in isolated cases surgical repair is associated with a low mortality rate (24-27).

Percutaneous balloon angioplasty is usual in re-COA after an initial surgical operation; however, there is controversy over it as an initial therapeutic strategy for native COA. Some studies have shown the safety and efficacy of balloon angioplasty in native COA (12-18), but according to other studies balloon angioplasty is not appropriate for native COA because of residual and recurrent stenosis and development of aneurysm at the angioplasty location, especially during infancy (28-35).

The rate of residual pressure gradient over 20 mm Hg after balloon angioplasty is 8%-27% based on available data (12,16,18) and is more common in infants under 6 months of age compared to older children (14-17,19).

In the present study none of subjects had residual pressure gradient over 20 mm Hg; however, 3 subjects (12%) exhibited recurrent stenosis, 2 of whom again underwent successful angioplasty and one underwent surgical repair. This rate of recurrent stenosis is similar to the surgical technique and other studies (27-35).

The prevalence of aneurysm after angioplasty has been reported to be 5%-15% (16-18,27-33). In the present study, none of the subjects exhibited aneurysm during the follow-up period.

After balloon angioplasty of native COA, acute complications might arise. Mortality has not been reported after infancy but death might occur during infancy (12). In the present study, none of the patients died in the catheterization laboratory and due to angioplasty. After successful angioplasty – without acute complications – 4 (16%) of the cases died in the intensive care unit (ICU) due to severe cardiopulmonary insufficiency and multi-organ failure. All these patients were less than 60 days of age and presented with cardiogenic shock in the pediatric emergency unit and had undergone cardiopulmonary resuscitation and intra-tracheal intubation, followed by angioplasty in an emergency setting.

The most common acute complication is injury to the femoral artery (6%-17%), which is more common in infants under 1 year of age (27-29); however, introduction of low-diameter balloon catheters has resulted in a decrease in such complications (20).

In the present study, 3 patients (12%) developed thrombosis of the femoral artery; two patients underwent anticoagulant therapy but in one subject (4%) femoral artery thrombosis resulted in the obstruction of the artery. In this patient there was sufficient distal run-off through collateral vessels. Other acute complications included hemorrhage from the femoral artery, cerebrovascular accidents and paradoxical hypertension (21).

Five subjects (20%) in this case series required transfusion of blood and 2 (8%) received antihypertensive agents due to an increase in blood pressure after the procedure and continued receiving such medication throughout the follow-up period.

In relation to the selection of the treatment modality, a study compared the surgical and balloon angioplasty techniques in infants less than 3 months of age, demonstrating similar results but the complications were more numerous and duration of intubation and hospitalization was longer in the surgery group (35). Therefore, it has been concluded that balloon angioplasty is more effective in severely ill infants compared to surgery and has many advantages. In addition, in cases in which none of the treatment modalities is definitely superior, it appears trans-catheter technique is preferable. However, selection of a correct treatment modality predominantly depends on the patient's age at the time of presentation, the anatomy of the stenotic site and the adjacent structures (36).

Conclusion

Based on the results of the present study and review of other relevant studies, it appears balloon angioplasty can be considered as an alternative technique in infants with native coarctation of aorta; however consulting the following significantly improve the outcomes.

First, attention to the symptoms and signs, comprehensive examination of the patient, use of simultaneous pulse oximetry of the lower and upper extremities before discharging the infants from the maternity ward/hospital in order to prevent delay in correct diagnosis of the condition; second, use of low-profile catheters during angioplasty, frequent irrigation of the catheter and judicious use of heparin to prevent femoral artery thrombosis.

Ethical Issues

The ethical committee of Tabriz University of Medical Sciences approved the study.

Conflicts of Interests

Authors declare that they have no competing interests.

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References

1. American Academy of Pediatrics. Report of the New England regional infant cardiac program. *Pediatrics*. 1980;65(2):377-461.
2. Campbell M, Polani PE. The aetiology of coarctation of the aorta. *Lancet*. 1961;1(7175):463-468.
3. Crafoord C. Congenital coarctation of the aorta and its surgical treatment. *J Thorac Surg*. 1945;14:347-352.
4. van Son JA, Daniels O, Vincent JG, van Lier HJ, Lacquet LK. Appraisal of resection and end-to-end anastomosis for repair of coarctation of the aorta in infancy: preference for resection. *Ann Thorac Surg*. 1989;48(4):496-502.
5. Rubay JE, Sluysmans T, Alexandrescu V, et al. Surgical repair of coarctation of the aorta in infants under one year of age: long-term results in 146 patients comparing subclavian flap angioplasty and modified end-to-end anastomosis. *J Cardiovasc Surg (Torino)*. 1992;33(2):216-222.
6. Merrill WH, Hoff SJ, Stewart JR, Elkins CC, Graham TP Jr, Bender HW Jr. Operative risk factors and durability of repair of coarctation of the aorta in the neonate. *Ann Thorac Surg*. 1994;58(2):399-402.
7. Quaegebeur JM, Jonas RA, Weinberg AD, Blackstone EH, Kirklin JW. Outcomes in seriously ill neonates with coarctation of the aorta: a multi institutional study. *J Thorac Cardiovasc Surg*. 1994;108(5):841-851.
8. Wood AE, Javadpour H, Duff D, Oslizlok P, Walsh K. Is extended arch aortoplasty the operation of choice for infant aortic coarctation? Results of 15 years' experience in 181 patients. *Ann Thorac Surg*. 2004;77(4):1353-1357.
9. Ho EC, Moss AJ. The syndrome of mesenteric arteritis following surgical repair of aortic coarctation. *Pediatrics*. 1972;49(1):40-45.
10. Brewer LA 3rd, Fosburg RG, Mulder GA, Verska JJ. Spinal cord complications following surgery for coarctation of the aorta. *J Thorac Cardiovasc Surg*. 1979;64(3):368-381.
11. Singer MI, Rowen M, Dorsey TJ. Transluminal aortic balloon angioplasty for coarctation of the aorta in the newborn. *Am Heart J*. 1982;103(1):131-132.
12. Tynan M, Finley JP, Fontes V, Hess J, Kan J. Balloon angioplasty for the treatment of native coarctation: results of valvuloplasty and angioplasty of congenital anomalies registry. *Am J Cardiol*. 1990;65(11):790-792.
13. Lock JE, Bass JC, Amplatz K, Fuhrman BP, Castaneda-Zuniga W. Balloon dilation angioplasty of aortic coarctations in infants and children. *Circulation*. 1983;68(1):109-116.
14. Beekman RH, Rocchini AP, Dick M 2nd, et al. Percutaneous balloon angioplasty for native coarctation of the aorta. *J Am Coll Cardiol*. 1987;10(5):1078-1084.
15. Morrow WR, Vick GW 3rd, Nihill MR, et al. Balloon dilatation of unoperated coarctation of the aorta: short and intermediate term results. *J Am Coll Cardiol*. 1988;11(1):133-138.
16. Mendelshon AM, Lloyd TR, Crowley DC, Sandhu SK, Kocis KC, Beekman RH 3rd. Late follow-up of balloon angioplasty in children with a native coarctation of the aorta. *Am J Cardiol*. 1994;74(7):696-700.
17. Fletcher SE, Nihill MR, Grifka RG, O'Laughlin MP, Mullins CE. Balloon angioplasty of native coarctation of the aorta: mid-term follow-up and prognostic factors. *J Am Coll Cardiol*. 1995;25(3):730-734.
18. Fawzy ME, Fathala A, Osman A, Badr A, Mostafa MA, Mohamed G, et al. Twenty-two years of follow-up results of balloon angioplasty for discrete native coarctation of the aorta in adolescents and adults. *Am Heart J*. 2008;156(5):910-917. doi: 10.1016/j.ahj.2008.06.037.
19. Fiore AC, Fischer LK, Schwartz T, et al. Comparison of angioplasty and surgery for neonatal aortic coarctation.

- Ann Thorac Surg. 2005;80(5):1659-1664.
20. Burrows PE, Benson LN, Williams WG, et al. Iliofemoral arterial complications of balloon angioplasty for systemic obstructions in infants and children. *Circulation*. 1990;82(5):1697-1704.
 21. Choy M, Rocchini AP, Beekman RH, et al. Paradoxical hypertension after repair of coarctation of the aorta in children: balloon angioplasty versus surgical repair. *Circulation*. 1987;75(6):1186-1191.
 22. Zehr KJ, Gillinov AM, Redmond JM, et al. Repair of coarctation of the aorta in neonates and infants: a thirty-year experience. *Ann Thorac Surg*. 1995;59(1):33-41.
 23. Alsoufi B, Cai S, Coles JG, Williams WG, Van Arsdel GS, Caldarone CA. Outcomes of different surgical strategies in the treatment of neonates with aortic coarctation and associated ventricular septal defects. *Ann Thorac Surg*. 2007;84(4):1331-1336.
 24. Wright GE, Nowak CA, Goldberg CS, Ohye RG, Bove EL, Rocchini AP. Extended resection and end-to-end anastomosis for aortic coarctation in infants: results of a tailored surgical approach. *Ann Thorac Surg*. 2005;80(4):1453-1459.
 25. Burch PT, Cowley CG, Holubkov R, et al. Coarctation repair in neonates and young infants: is small size or low weight still a risk factor? *J Thorac Cardiovasc Surg*. 2009;138(3):547-552. doi: 10.1016/j.jtcvs.2009.04.046.
 26. Meng BY1, Wang T, Zhang Q, Ma C, Peng L, Wang YX, Pan XL. One-stage complete correction of 52 cases infantile aortic coarctation or interrupted aortic arch associated with intracardiac anomalies (Chinese). *Zhonghua Wai Ke ZaZhi*. 2011;49(1):66-69.
 27. Rao PS, Galal O, Smith PA, Wilson AD. Five- to nine-year follow-up results of balloon angioplasty of native coarctation in infants and children. *J Am Coll Cardiol*. 1996;27(2):462-470.
 28. Ammar RI. Balloon angioplasty for native aortic coarctation in children and infants younger than 12 months: immediate and medium-term follow-up. *J Invasive Cardiol*. 2012;24(12):662-666.
 29. Lee CL, Lin JF, Hsieh KS, Lin CC, Huang TC. Balloon angioplasty of native coarctation and comparison of patients younger and older than 3 months. *Circ J*. 2007;71(11):1781-1784.
 30. Adjagba PM, Hanna B, Miró J, et al. Percutaneous angioplasty used to manage native and recurrent coarctation of the aorta in infants younger than 1 year: immediate and midterm results. *Pediatr Cardiol*. 2014;35(7):1155-1161. doi: 10.1007/s00246-014-0909-3.
 31. Rao PS, Jureidini SB, Balfour IC, Singh GK, Chen SC. Severe aortic coarctation in infants less than 3 months: Successful palliation by balloon angioplasty. *J Invasive Cardiol*. 2003;15(4):202-208.
 32. Suarez de Lezo J, Pan M, Romero M, et al. Percutaneous interventions on severe coarctation of the aorta: A 21-year experience. *Pediatr Cardiol*. 2005; 26(2):176-189.
 33. Patel HT, Madani A, Paris YM, Warner KG, Hijazi ZM. Balloon angioplasty of native coarctation of the aorta in infants and neonates: Is it worth the hassle? *Pediatr Cardiol*. 2001;22(1):53-57.
 34. Truong DT, Tani LY, Minich LL, Burch PT, Bardsley TR, Menon SC. Factors associated with recoarctation after surgical repair of coarctation of the aorta by way of thoracotomy in young infants. *Pediatr Cardiol*. 2014;35(1):164-170. doi:10.1007/s00246-013-0757-6.
 35. Rao PS, Chopra PS, Kosciak R, Smith PA, Wilson AD. Surgical versus balloon therapy for aortic coarctation in infants <3 months old. *J Am Coll Cardiol*. 1994;23(6):1479-1483.
 36. Doshi AR, Rao PS. Coarctation of aorta-management options and decision making. *Pediatr Therapeut S* 2012;5:2161-0665.

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