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Recurrent Coarctation: Interventional Techniques and Results

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Abstract

Coarctation of the aorta (CoA) accounts for 5% to 8% of all congenital heart defects. With all forms of interventions for native CoA, repeat intervention may be required due to restenosis and/or aneurysm formation. Restenosis rates vary from 5% to 24% and are higher in infants and children and in those with arch hypoplasia. Although repeat surgery can be done for recurrent CoA, guidelines from a number of professional societies have recommended balloon angioplasty with or without stenting as the preferred intervention for patients with isolated recoarctation. For infants and young children with recurrent coarctation, balloon angioplasty has been shown to be safe and effective with low incidence of complications. However, the rates of restenosis and reinterventions are high with balloon angioplasty alone. Endovascular stent placement is indicated, either electively in adults or as a bailout procedure in those who develop a complication such as dissection or intimal tear after balloon angioplasty. Conventionally bare metal stents are used; these can be dilated later if required. Covered stents, introduced more recently, are best reserved for those who have aneurysm at the site of previous repair or who develop a complication such as aortic wall perforation or tear. Stents produce complete abolition of gradients across the coarct segment in a majority of cases with good opening of the lumen on angiography. The long-term results are better than that of balloon angioplasty alone, with very low rates of restenosis. However, endovascular stenting is a technically demanding procedure and can be associated with serious complications rarely.

Keywords

angioplasty, coarctation, stents, interventional catheterization

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Introduction

Coarctation of the aorta (CoA) is an obstructive lesion of the proximal aorta located near the aortic end of the ductus arteriosus or ligamentum arteriosum. Although anatomically the lesion may appear straightforward, it indeed has a complex anatomy, pathophysiology, clinical presentation, treatment options, and outcomes. Morphological spectrum varies from a discrete CoA to hypoplastic aortic arch.

The aortic wall in patients with CoA is often abnormal due to histologic differences in the smooth muscle and extracellular matrix when compared with normal aortic wall. This abnormality may affect local compliance and distensibility.[1,2] Coarctation of the aorta is at times missed on clinical examination, and a high index of suspicion is required to make this diagnosis. Coarctation of the aorta is a relatively common defect that accounts for 5% to 8% of all congenital heart defects. Although the lesion may exist as an isolated defect, the obstruction may also be associated with congenital heart defects, the commonest being a bicuspid aortic valve. Ventricular septal defect and other intracardiac lesions favoring increased pulmonary blood flow at the expense of aortic blood flow are also known associations with CoA.

Since surgical repair of CoA became available in 1944, great progress has been made in the diagnosis and treatment of this condition.[3] Perioperative morbidity and mortality have been greatly reduced, particularly in cases of uncomplicated CoA not associated with complex congenital heart disease.[4–7] Current therapeutic options for native CoA include surgery, intravascular stent placement, and balloon angioplasty. Catheter-based therapies have demonstrated comparable efficacy to surgery in children greater than one year of age.[8,9]

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Recurrent CoA

With all forms of therapy, repeat intervention may be required due to recurrence of obstruction. Restenosis is seen primarily in children, usually due to inadequate aortic wall growth at the site of repair when surgery has been performed before the aorta has reached adult size. The recurrence rate reported in different series varied between 5% and 24%.[10–13] Repeat intervention is at times required for aneurysm formation, which rarely develops at the site of previous intervention.

Risk Factors for Recurrent CoA

Age of the child at the time of intervention. Neonates (<30 days of age) and to a lesser extent older infants (<1 year of age) are at a higher risk for recurrent CoA, especially after balloon dilatation.[11,14–20] In one study, restenosis was more likely when the narrowest segment was <3.5 mm before balloon dilatation and <6 mm after balloon dilatation.[11] Surgical repair at young age is also a risk factor for recurrence.[21] However, some of the studies have not validated this and, in fact, shown that postponing repair can potentially be harmful by lowering the long-term survival and normalization of blood pressure.[10,11,22]

Presence of isthmus hypoplasia. This has been shown to be a potent risk factor for recurrence of CoA.[11,15]

Surgical technique used for repair. Some studies suggest that certain surgical techniques may play a major role in recurrence or persistence of CoA.[23] However, others have not found this to be a significant risk factor. According to one study, the specific surgical method of repair, suture material used, and sewing technique do not appear to be as important as once suggested in determining recurrence.[24]

Catheter Interventional Techniques for Recurrent Coarctation

Although surgery can be undertaken for recurrent CoA with reasonable success and safety, guidelines from a number of professional societies have recommended balloon angioplasty with or without stenting as the preferred intervention of patients with isolated recoarctation.[25–27]

Lock et al have been credited with first balloon angioplasty, which was initially done in the excised segments of human CoA. Since then, balloon angioplasty has become a standard method of treatment in both native and recurrent CoA.[28] Singer et al described a successful balloon dilation of recurrent CoA for the first time. It was performed in a critically ill neonate who developed restenosis after surgery.[29] Since then balloon angioplasty has been shown to be safe and effective for children with recurrent CoA. As it is less invasive than surgery, most centers consider balloon angioplasty as the procedure of choice for recurrent CoA.

Since the early 1990s, use of balloon expandable stents has been added to our armamentarium against coarctation. It has been suggested that stenting after balloon angioplasty lowers the risk for complications and has a beneficial effect on long-term survival. However, children with aortic stent placement are more likely to require a planned reintervention as the stent often needs to be dilated as the child grows.

There has been a recent shift from balloon angioplasty alone to stent implantation. It has been postulated that balloon dilatation can injure the intima and part of the media. Although the vessel diameter increases following balloon angioplasty, fibrous scar tissue can form over a period of months that results in recoil, restenosis, and aneurysm formation.[30] Advantages of using a stent are the radial support to the vessel wall and the apposition of the torn vessel intima to the media with the possibility to perform redilatation (especially in young patients). With this approach, there is no need for oversizing and hence major transmural tears do not occur.[31–33]

Indications for Intervention in Recurrent CoA

The definition of recurrent coarctation is a residual gradient of more than 20 to 30 mm Hg at the coarctation site. For most patients with discrete recurrent/residual coarctation after surgical repair, catheter intervention, either with balloon angioplasty or with endovascular stent placement, has been shown to be a better therapeutic option as compared with surgery. The indications for balloon angioplasty and stent placement are listed in Table 1 per guidelines from American Heart Association (AHA) in 2011.[25]

Diagnostic Workup

All patients should get four limb blood pressure measured, and this will give a fair idea of gradient across the coarct segment. A complete physical examination, an x-ray chest, and an electrocardiogram should be done to confirm the presence of coarctation and to rule out any associated abnormalities.

Echocardiography is able to provide the anatomic details of coarctation segment, especially in small children with good echocardiographic windows. The site, extent of narrow aortic segment, left ventricular size and function, and associated congenital heart disease are all well defined by echocardiography. Aortic arch hypoplasia, if present, should be noted. The diameter of isthmus and descending aorta at the level of diaphragm should be measured to estimate balloon size. Doppler gradients across the coarct segment may not be very reliable, but diastolic drag of the Doppler signal is a very important sign indicative of significant stenosis.

In older children and adults, computed tomography (CT) or cardiovascular magnetic resonance (CMR) imaging should be performed to define the coarctation anatomy better before planning the interventional procedure. The site, extent, the severity of narrowing, and presence or absence of arch hypoplasia is well seen by both these modalities. The diameter of narrowest segment, pre- and poststenotic segment, is measured for determining the balloon and/or stent diameter and length. Complications of previous intervention, such as aneurysm, are best seen on CT or CMR (Figure 1). Computed tomography has

Table 1. Indications for Balloon Angioplasty and Stent Placement in Patients With Recurrent Coarctation of Aorta.

	Class	LOE
Indications for balloon angioplasty		
Transcatheter systolic coarctation gradient of >20 mm Hg and suitable anatomy, irrespective of patient age	I	C
Transcatheter systolic coarctation gradient of <20 mm Hg and in the presence of significant collateral vessels and suitable angiographic anatomy, irrespective of patient age, in patients with univentricular heart or with significant ventricular dysfunction	I	C
Complex coarctation anatomy or systemic conditions such as connective tissue disease or Turner syndrome but each case should be scrutinized on a case-by-case basis	II	C
Indications for stent placement		
Patient of sufficient size for safe stent placement, in whom the stent can be expanded to an adult size and who have a transcatheter systolic coarctation gradient >20 mm Hg	I	B
Placement of stent that can be expanded to an adult size in patients with:	IIa	
a transcatheter systolic coarctation gradient of >20 mm Hg		B
a transcatheter systolic coarctation gradient of <20 mm Hg but with systemic hypertension associated with an anatomic narrowing that explains the hypertension		C
a long-segment coarctation with a transcatheter systolic coarctation gradient >20 mm Hg		B
In patients in whom balloon angioplasty has failed as long as a stent that can be expanded to an adult size can be implanted	IIa	B
Placement of a stent that can be expanded to an adult size for:	IIb	C
a transcoarctation gradient of <20 mm Hg but with an elevated left ventricular end-diastolic pressure and an anatomic narrowing		C
a transcoarctation gradient of <20 mm Hg but in whom significant aortic collaterals exist, which results in an underestimation of the coarctation		

Abbreviation: LOE, level of evidence.

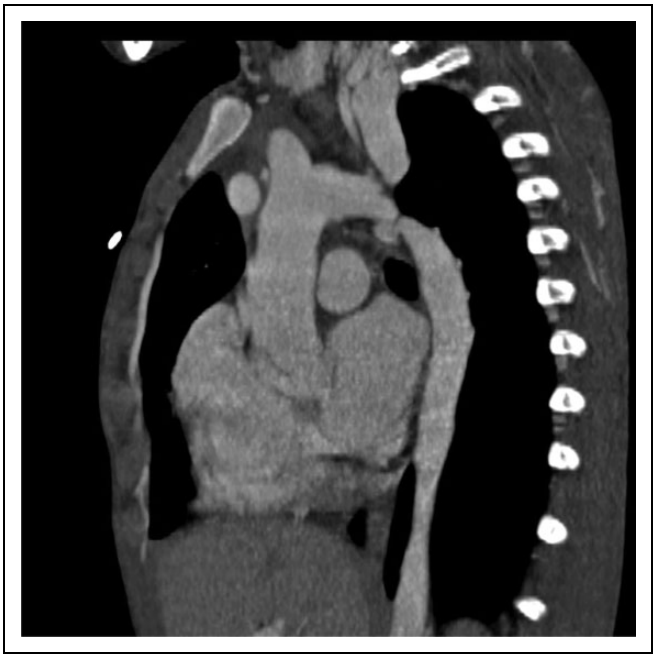

Figure 1. Computed tomography (CT) image of the aortic arch showing coarctation with formation of aneurysm at the site of narrowing.

the disadvantage of radiation exposure but the procedure time is very low when compared with CMR.

Technique of Balloon Angioplasty

The procedure is generally performed under light sedation and local anesthesia. General anesthesia may be required for small children. A percutaneous femoral artery approach is used by most. In case of difficulty or in small infants, axillary approach can be used. Patient is heparinized with 100 units/kg of unfractionated heparin. A pigtail or a multipurpose catheter is passed through the arterial sheath, placed above the coarctation segment, pressure gradient measured, and angiography performed in two views (left anterior oblique and right anterior oblique/anteroposterior views). The angiograms are reviewed for defining the site, severity, and length of the narrow segment (Figure 2A and B). Appropriate sized balloon angioplasty catheter (see subsequently) is selected. Recommended size (0.018-0.035 in) J-shaped guidewire is passed in the catheter and the catheter removed, keeping the wire well above the narrow segment, preferably in the ascending aorta. The balloon catheter is then passed over the guidewire and positioned across the coarctation segment. The balloon is inflated using dilute contrast solution to a pressure of 3 to 5 atm and kept inflated for 5 to 10 seconds. One is able to see the indentation (or waist) on the inflated balloon; this should be in the mid part of the balloon. Monitoring of femoral pressure is recommended during this procedure. One may have to place a small arterial sheath in the other groin. The femoral artery pressure should improve once the balloon is deflated. The systolic pressure rises and the pulse pressure widens. One can repeat the inflation once or twice and note complete disappearance of indentation in the balloon. The balloon is removed keeping the guidewire in position and multipurpose catheter is advanced over the guidewire. Under no circumstances, the dilated segment must be recrossed as that may result in dissection of aorta. Similarly the tip of the catheter or guidewire should not be manipulated over the site of freshly dilated coarctation segment. All catheters, diagnostic or balloon, should be exchanged over the guidewire.

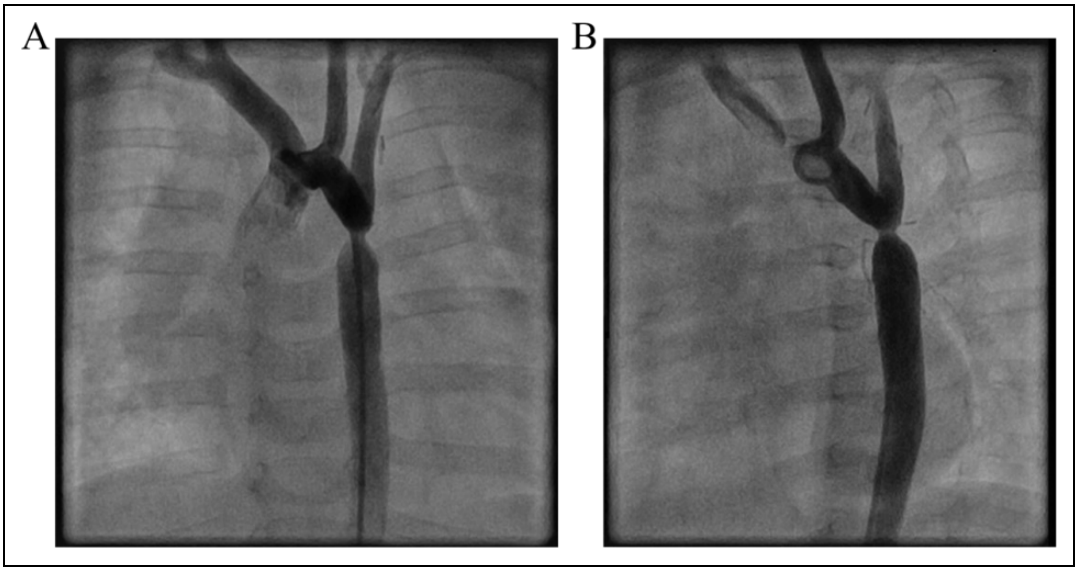

Figure 2. Aortic angiogram in shallow right anterior oblique (A) and left anterior oblique (B) views showing discrete coarctation of aorta.

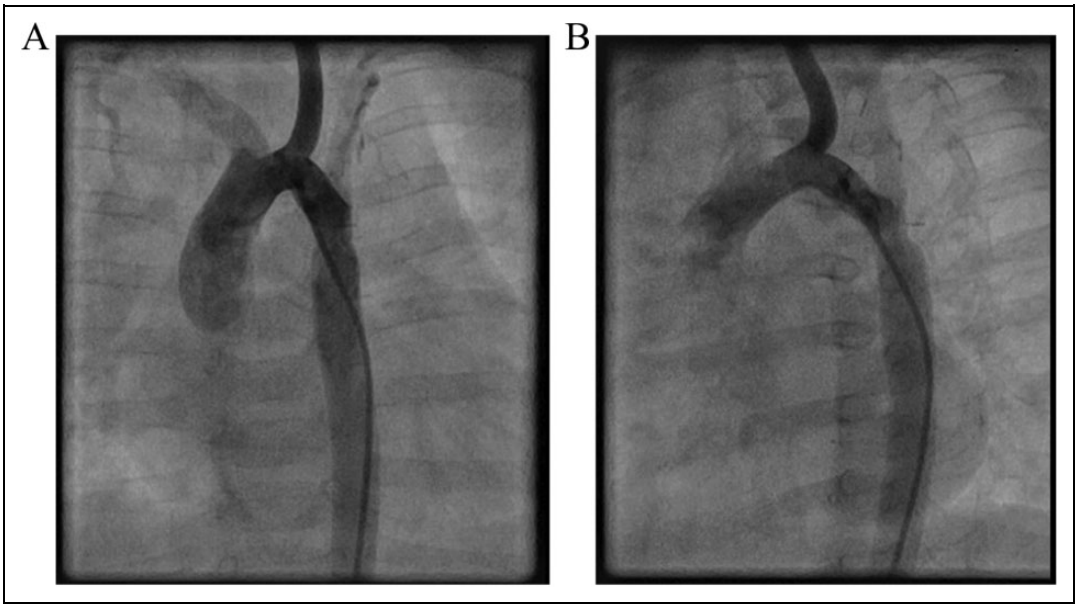

Figure 3. Aortic angiogram in shallow right anterior oblique (A) and left anterior oblique (B) views after balloon angioplasty, showing significant increase in aortic diameter.

The pressure gradient across the dilated segment is measured. In successful cases, the gradient should reduce to less than 10 mm Hg (or <15 mm Hg). The heart rate and blood pressure should be monitored all through the procedure. Repeat angiography is performed in same views, which will show improvement in diameter of the coarctation segment (Figure 3A and B). Small cap dissections are not uncommon at local site, these do not compromise flow. In cases where residual gradients persist, one can go with a slightly larger balloon, not exceeding the recommended size, or consider endovascular stent placement.

Balloon sizing. The size of the balloon should not exceed the size of the descending aorta at the level of diaphragm. At the same time, it should not exceed isthmus diameter by more than 10%. Some operators also consider the size of the narrowest coarctation segment, not exceeding 2 to 3 times the diameter of this segment.

Definition of acute success. A fall in gradient across coarctation to <20 mm Hg (or <15 mm Hg) with an increase in the diameter of the narrow segment in the absence of a serious complication should be labeled as a successful procedure.

Immediate results. Several studies have shown successful opening of discrete recoarctation by using percutaneous balloon angioplasty in children, including infants as young as three

months of age. The immediate success rates have varied from 80% to 93%.[34–42] In the study from Czech Republic of 99 patients aged 36 days to 32.6 years (median 268 days), successful angioplasty could be achieved in 93% of cases.[40] The systolic gradient reduced from 34 (range 26-44.5 mm Hg) to 15 mm Hg (range 8.25-27 mm Hg).

Yetman et al reported that among 90 patients (median age of 1.1 years) who underwent balloon angioplasty for recoarctation, systolic gradient fell from 31 + 21 to 8 + 9 mm Hg ($P = .0001$).[34] Overall success rate (gradient <20 mm Hg) was 88%. Success rate of 91% has been reported in another study of 22 children.[41] In a more recent multicentric study of 36 institutions, 54 patients underwent balloon angioplasty at a mean age of 13.8 + 10.3 years. A gradient of <15 mm Hg was achieved in 80% of cases.[42]

Acute complications. These include aortic wall complications such as dissection, intimal disruption, tear (1%-7%), cerebrovascular accidents (<1%), and death (0%-2%).[34,35,42] Other complications include balloon rupture and local arterial compromise.

Intermediate- and long-term results. Restenosis requiring reintervention, aneurysm formation, and continued hypertension are the important issues in those who undergo balloon angioplasty for recurrent CoA. Reich et al described long-term follow-up data (up to 20.7 years, median 8.1 years) after balloon angioplasty for recoarctation in children, which demonstrated a low complication rate as well as a low risk of aortic aneurysm formation.[40] Survival after 20 years was 91%. However, the reintervention-free survival was only 44%. Yetman et al reported success in a large series of children after balloon angioplasty for aortic recoarctation. However, in 31% of the 90 cases, reintervention was needed.[34] Reintervention rates in the other studies ranged from 6% to 53%.[36,42]

In the multi-institution study reported recently, 27 patients had intermediate term follow-up, and the gradient between upper and lower limb was <10 mm Hg in 94%. Reintervention-free probability was 95% at 1 year and 72% at 5 years.[42]

Therefore, balloon angioplasty seems more suitable for the treatment of restenosis during infancy or early childhood, as it avoids the need for repeat surgery. Balloon angioplasty may be useful to prevent the need for stent implantation entirely or postpone stent implantation until adult age when stenting with adult-sized stents is feasible.[40,42–44]

Endovascular Stents

De Lezo et al published the first experiences of treating severe aortic coarctation with balloon-expandable stents. The premise is that stenting after balloon angioplasty lowers the risk for complications and has a beneficial effect on long-term survival.[45–51] Stents have been shown to abolish gradients across the coarcted segment completely, which is very unlikely to be achieved with balloon angioplasty alone.

The available stents are of two types, bare metal and covered stents. Conventionally bare metal stents are used. Covered stents have a lower radial strength and are much more expensive. These are best reserved for specific indications as detailed subsequently.

Stents support the integrity of aortic wall during balloon dilatation. Endovascular stents for recurrent coarctation can be used for elective stenting, for failed balloon angioplasty, in cases with unsuitable anatomy for balloon angioplasty or as a bailout procedure following complications after balloon angioplasty. There are no randomized trials that compare short- or long-term outcomes following balloon angioplasty with endovascular stent placement in recurrent CoA.

The current AHA guidelines recommend stent implantation for recoarctation therefore in all patients with an aortic recoarctation and a gradient >20 mm Hg, which are of sufficient size for safe stent placement and in whom expansion of stent to an adult size is possible.[25]

Technique

The initial part of the procedure is quite similar to that of balloon angioplasty. However, if elective stenting is being considered and the stenosis is very severe, it is better to dilate with an undersized balloon. An appropriate sized stent is chosen based on angiography images. Generally one uses a size of 4014 stent (14 mm diameter and 40 mm length; Figure 4A and B). The stent is of high profile and one needs to use large-sized long sheaths. To avoid migration of stent during balloon inflation, one can either rapidly pace the right ventricle or use a special balloon-in-balloon catheter. The angiogram is repeated after stent deployment to confirm its position. If necessary, one can redilate the stent using a bigger balloon. The guidewire must stay across the dilated segment during the entire procedure. In some older and elderly patients, one may have to perform staged dilatation.

Covered stents are indicated for aneurysm formation at the coarct site (Figure 5A and B), perforation or rupture of aorta, dissection of intima, or within a fractured previous stent. Covered stents may also be used for postsurgical noncompliant coactation segment.[31,52]

New types of covered stents are of relatively low profile, are balloon mounted, and can be introduced through a 9 or 11F sheath. One need to be very careful when placing a covered stent because if it covers the ostia of a branch, that branch is likely to get occluded. The stent must stay away from left subclavian artery, which is at times very close to the coarct segment.

Results. Immediate success is obtained in >95% of cases. In a large multi-institutional study of 565 procedures, including 269 patients with recurrent CoA, success was achieved in 97.9% of cases.[53] Mean age was 18.1 years. In a more recent series by Holzer et al, the success rate was 96% in 302 patients, including 135 with recurrent CoA.[54] The gradients are generally abolished, almost totally, and the coarct segment diameter increases. Another small study also reported a good immediate and long-term success in 10 patients with recurrent coarctation

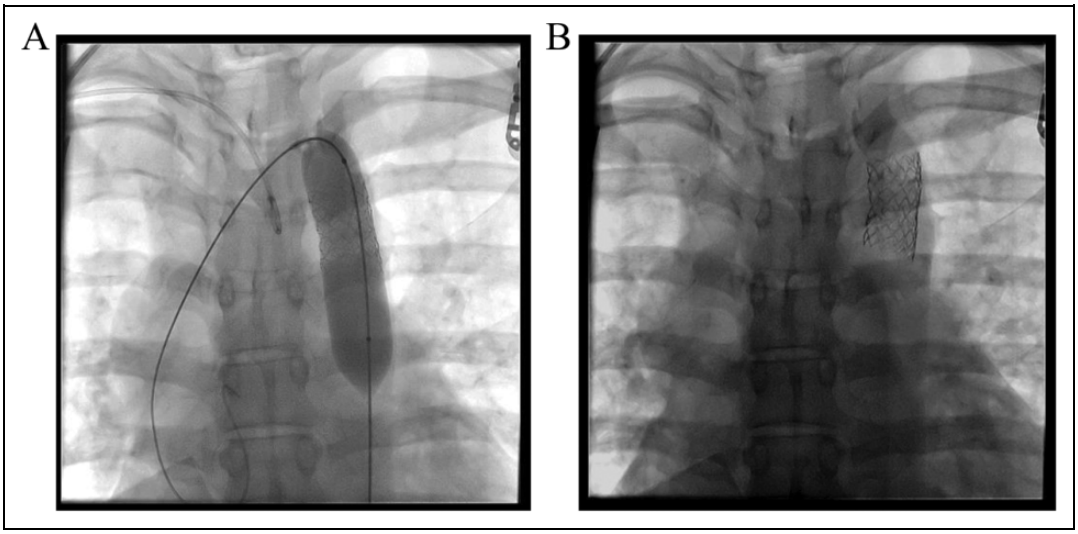

Figure 4. Deployment of endovascular stent across coarctation segment (A). Note the bare metal stent after balloon has been deflated and removed (B).

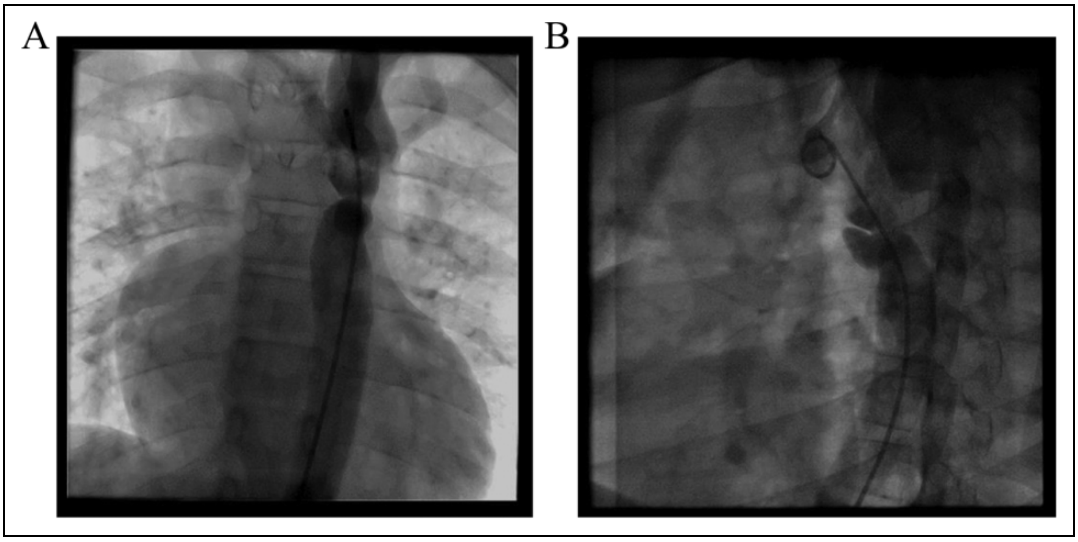

Figure 5. Aortic angiogram showing aneurysm at the site of coarctation segment narrowing in anteroposterior (A) and left anterior oblique (B) views.

beyond 6 years of age.[55] Some of the available stents can be redilated later to a particular size using a bigger balloon.

Complications. Complications following stent placement occur in 10% to 15% of cases. These include aortic wall complications such as intimal tear, dissection, and aneurysm formation (4%-5%). For some of these, covered stent can be deployed inside the bare metal stent as a bailout procedure. Others may need emergency surgery. Technical complications like stent migration and balloon rupture occur in 8% to 10%, cerebrovascular accidents and peripheral emboli in <1%, and local arterial injury in 2% to 3%. Mortality of the procedure is <1%.[53,54,56,57] In one of the studies, the overall complication rate declined from 16.3%, prior to the year 2002 to 6.1%, after the year 2002. Two patients had aortic rupture requiring emergency surgery.[53]

Intermediate and long-term follow-up. Cumulative intermediate success has been reported in 86% and cumulative long-term success in 77%.[54] Restenosis is quite rare if the initial results were good. Aneurysm formation has been reported in 1% to 9%, the incidence depends upon how aggressive one looks for these.[54,58] An integrated arch imaging has been performed in the study by Forbes et al in 144 patients, and the incidence of aneurysm was 9%. Authors recommend this approach for all patients who have been stented.[58] However, majority of these were small and managed conservatively. Stent fracture is also rarely described, which may be responsible for restenosis.

In the Forbes study, a balloon to coarctation ratio of 3.5 or more and present angioplasty increased the likelihood of an abnormal finding on follow-up imaging. Over long term, most patients become normotensive on no or single drug.

Endovascular stenting for CoA is a technically demanding procedure and should be performed in experience centers. These catheterization laboratories should have a vast inventory of stents and balloon and covered stent should be available when the procedure is done in high-risk cases.

Conclusion

Recurrence is seen in a significant proportion of cases of CoA after an initial intervention. Infants and young children are best treated with either balloon angioplasty or surgery. For those presenting in later life, beyond 10 to 12 years of age, endovascular stent placement is likely to provide better long-term outcome as compared to surgery or balloon angioplasty alone. However, stent procedures are technically demanding procedures which should be done in experienced centers. Stents may also be required as a bailout procedure following a complicated balloon angioplasty.

Declaration of Conflicting Interests

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