

Stenting therapy for aortic coarctation: a tunisian center experience.

Stenting de la coartation de l'aorte : experience d'un centre tunisien.

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Résumé

Introduction ; Le stenting constitue désormais le traitement de choix de la coarctation native ou récurrente de l'aorte (CoA) avec une anatomie favorable dans la plupart des centres.

Méthodes ; Nous avons rapporté dans cette série monocentrique menée au service des Explorations Fonctionnelles et de Réanimation Cardiologique à l'hôpital La Rabta entre Octobre 2017 et Avril 2018 notre expérience de stenting des CoA.

Resultats : Durant la période de l'étude, sept patients ont été stenté pour une CoA. L'âge moyen des patients était de 32 ans avec un sex ratio de 0.14. Tous les patients étaient hypertendus. La CoA était native chez 5 patients et récurrente chez les 2 autres. Le gradient trans-sténotique moyen était de 73mmHg avec un rapport moyen entre le diamètre de l'isthme sténosé et celui de l'aorte saine de 29%. Six patients ont bénéficié d'un stent couvert devant une CoA sub-atrétique ou d'un pseudo-aneurysme associé et le dernier a eu un stent non couvert devant un risque d'atteinte de l'artère Adamkiewicz. Un succès procédural immédiat a été observé dans tous les cas avec un gradient final de moins 10 mmHg et un ratio final du diamètre isthmique par rapport à l'aorte saine de plus de 50% chez tous les patients. On n'a pas noté de complications aortiques ni fémorales. Après un suivi moyen de 13 mois, tous les patients étaient vivants avec une hypertension artérielle résiduelle chez trois d'entre eux.

Conclusion : Le stenting des CoA représente une modalité thérapeutique efficace et sûre. Sa place comme traitement de choix peut être actuellement suggérée.

Mots-clés

Coarctation de l'aorte ;
angioplastie ; stenting ;
stent couvert.

Summary

Background : In native or recurrent coarctation of the aorta (CoA) with appropriate anatomy, stenting has become the treatment of first choice over surgery repair and balloon angioplasty.

Methods : We reported in this single center case series led in the Functional Explorations and Cardiac Intensive Care Department at La Rabta Hospital between October 2017 to April 2018 our experience of stenting of CoA.

Results : In the study period, seven patient with CoA underwent aortic stenting. The mean age was 32 years, with a sex ratio of 0.14. Arterial hypertension was present in all patients. CoA was native in five cases and recurrent in two. The mean baseline trans-stenotic gradient was of 73mmHg, and the mean coarctated to normal aorta ratio was of 29%. Six patients were treated with a covered stent placement because of pre-atretic CoA or associated pseudo-aneurysm and one patient benefited from a bare metal stent implantation because of a risk of Adamkiewicz artery damage. Immediate procedural success was achieved in all cases with a final gradient of less than 10mmHg and a final coarctated to normal aorta ratio more than 50% in all patients. There were no aortic or femoral complications. After mean follow-up of 13 months, all patients were still alive with a residual arterial hypertension remaining in three of them.

Conclusion : Stenting of CoA represent an efficacious and secure therapeutic modality supporting its potential indication as a first line treatment of native and recurrent coarctation.

Keywords

Aortic coarctation;
angioplasty; stenting;
covered stent.

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INTRODUCTION

Coarctation of the aorta (CoA) is defined as a narrowing in the aorta, most commonly at the site of insertion of the ductus arteriosus, just distal to the left subclavian artery. It accounts for 5-8% of all congenital heart defects (1). In Tunisia, the prevalence is about 4.3% among births with diagnosed congenital heart diseases (2). Untreated, patients will die before 50 years of age with a median of 31 years old, mainly because of heart failure, aortic rupture, infective endocarditis and intracranial hemorrhage (3). As a result, every diagnosed severe CoA should be treated since the childhood even before becoming symptomatic.

The main goal of treatment is complete repair of the coarctated segment with neither complications nor residual gradient across the narrowed segment. The type of repair depends on the age of the patient, the severity of the coarctation, as well as the anatomic characteristics. There is no clear evidence to support the superiority of either surgical or catheter-based treatments for coarctation of the aorta (4,5). Historically, Crafoord surgical repair was described in 1945 and it had stilled the unique treatment until the first balloon angioplasty reported in 1982. Then, the first attempts of stent placement in the beginning of the 1990s were performed. Recently, percutaneous approaches have been more and more refined due to increased operators' experience and improved balloon and stent technology which have translated to increased success and safety of trans-catheter therapy. Therefore, angioplasty has become the treatment of choice in many centers in CoA with appropriate anatomy in patients aged of more than 1 years old (1).

The aim of this case series is to report our emerging experience in percutaneous treatment of aortic coarctation.

METHODS

Patients selection

We analyzed data from seven consecutive patients who were treated percutaneously for CoA from October 2017 to April 2018 in La Rabta Hospital in Tunisia.

A pre-interventional assessment was performed in order to evaluate the severity of the coarctation and its appropriateness to a percutaneous treatment. This included a clinical examination with a measure of blood pressure in the upper and lower limbs, an echocardiography and a computed tomography (CT) or a magnetic resonance imaging (MRI). The CT or MRI were performed in all patients in order to assess non-invasively the entire aorta. Both exams depict site, extent, and degree of the aortic narrowing, the aortic arch, the pre- and post-stenotic aorta, and collaterals.

Treatment was indicated, as recommended in the last guidelines of the European society of cardiology (1), in front of a significant native or recurrent coarctation with a narrowing of more than 50%, arterial hypertension with pressure difference >20 mmHg between upper and lower limbs, or left ventricular hypertrophy.

Only patients with a localized membrane shaped coarctation at the level of the aortic isthmus were included. Patients with isthmus or arch hypoplasia were not eligible to percutaneous therapy. Moreover, neonatal critical coarctation and severe associated congenital heart defects were excluded. All these forms were managed surgically.

Technical aspects

For all patients, a similar approach was adopted as showed in (figure 1): (1) The procedure was performed under general anesthesia. (2) We used the retrograde trans-femoral approach with a 12F or 14F sheath for non covered or covered stents respectively. (3) An antibioprophylaxis (Cephapirine) and an intravenous heparin at 70 to 100 mg/kg were administrated. (4) The coarctation segment was crossed using a straight hydrophilic coated 0.035'' wire or a 0.014'' coronary guidewire in sub-atretic CoA. This wire was secondary advanced distally meaning a Multipurpose or Judkins Right catheter in the right subclavian artery. (5) An exchange to a 0.035'', 2.60m long, stiff guidewire was performed (Lunderquist Extra-Stiff Wire Guide, Cook, Bloomington, USA) for better support. (6) We measured the peak-to-peak systolic pressures gradient across the coarctated segment (7) We performed a detailed angiogram, in profile and right anterior oblique projections, using either a pigtail or preferably a multitrack catheter which has the advantage of allowing a dye injection while the wire remained in place. This angiogram provided important evaluation of the diameter and the length of the aorta at the level of the narrowing and the adjacent segments as well as the distance between the origin of the left subclavian artery and the coarctation site. (8) The diameter of bare metal (BMS) or covered stent (CS) was selected based on aortic diameter proximal to the stenosis. This diameter should not have exceeded the size of the aorta at the level of the diaphragm and balloon to coarctation ratio should have been less than 3.5. (9) The stent was positioned in a manner to cover the stenotic segment while avoiding the origin of the left subclavian artery. (10) The stent was deployed by a 2 step-Ballon-in-Ballon technique (BIB, Numed, NY, USA) (Figure 2). (11) Angioplasty was considered successful if a gradient < 10 mmHg was achieved without any aortic wall injury (rupture or dissection). (12) An immediate or delayed post-dilation was indicated in case of residual gradient >10 mmHg by the mean of a non-compliant balloon under temporary cardiac rapid pacing. (13) Patients were discharged 48h

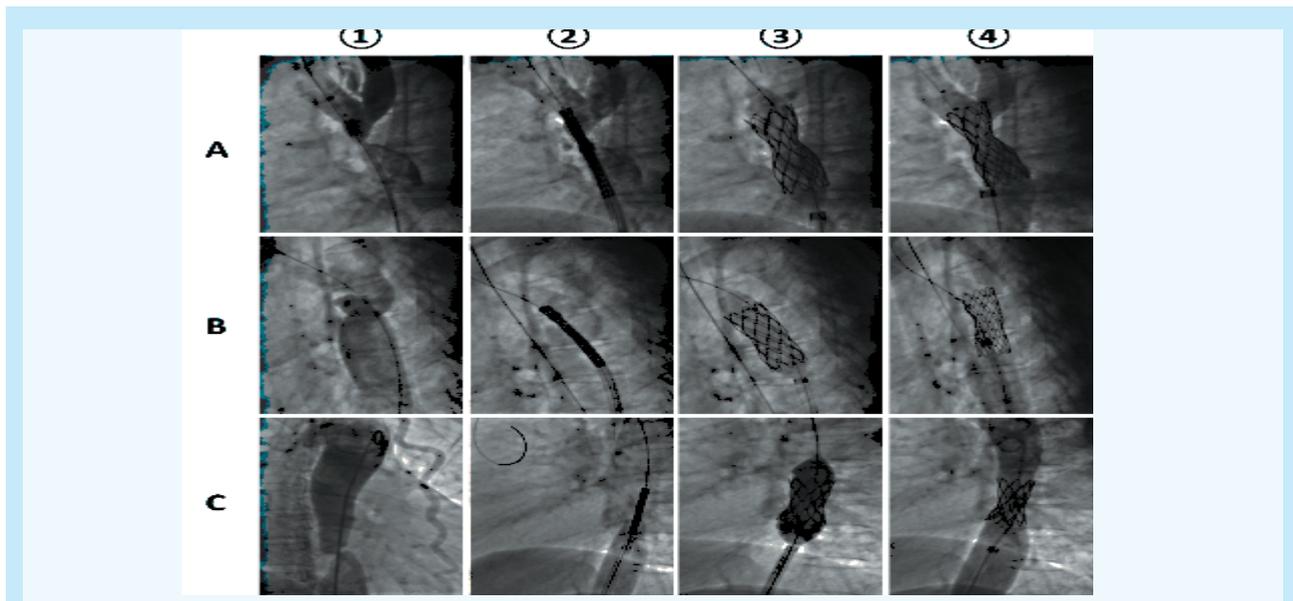


Figure 1 : Procedural steps of stenting of aortic coarctation

Native CoA (Panel A), Recurrent CoA with pseudo-aneurysm (Panel B) and recurrent CoA distant from the aortic isthmus (Panel C).

① Angiography of thoracic aorta using a pigtail or a multitrack catheter showing CoA.

② Positioning of covered (A and B) or bare metal stent (C) delivered by a balloon-in balloon catheter to cover the narrowed segment while avoiding the left subclavian artery ostium.

③ Inflating the two balloons to impact the stent to the aortic wall.

④ Final angiographic result showing a well apposed stents without obstruction of the left subclavian artery nor rupture or dissection of the aortic wall. The pseudo-aneurysm in Panel B was excluded.

CoA= Aortic coarctation.

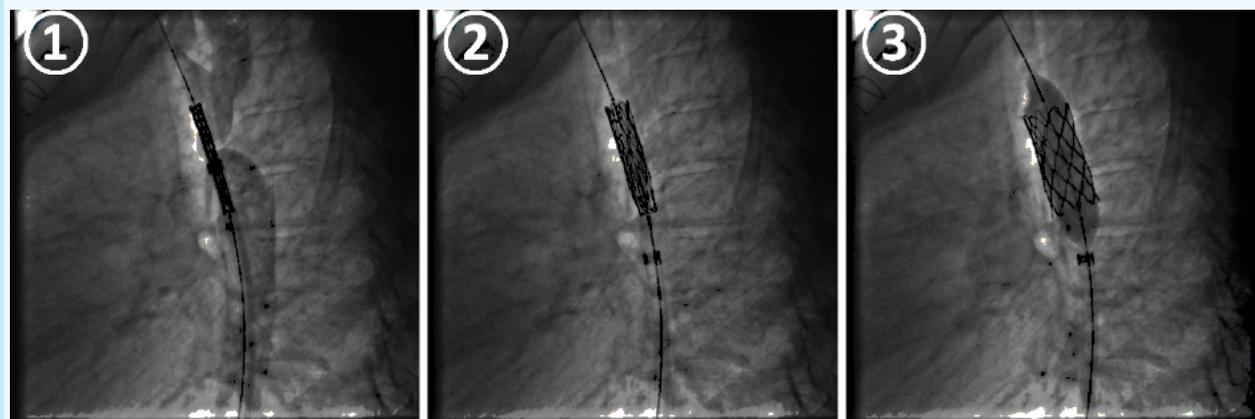


Figure 2: The balloon-in-balloon inflation technique.

Step ①: Stent positioning.

Step ②: Inner balloon inflation to the half of target diameter avoiding “dog bone” formation in order to trap the stent and decrease the risk of slippage.

Step ③: Outer balloon inflation achieving full expansion of the stent.

after the procedure. Aspirin and endocarditis prophylaxis were prescribed for 6 months. (14) Residual arterial hypertension (HTN), and long term aortic wall

complications (recoarctation, aneurysm formation) were screened clinically and by the mean of CT or MRI performed one year later.

Table 1: Patients and aortic coarctation characteristics.

Case n°	age	Sex	Native/ Recurrent coarctation	Anatomic particularities	Pressure difference between upper / lower limbs (mmHg)	HTN	Number of anti- hypertensive drugs	LVH	Bicuspi d aortic valve	Trans-stenotic gradient (mmHg)	Stenotic segment diameter / normal aortic segment diameter (mm)
1	31	female	native	-	>20	Yes	4	Yes	No	64	3/14 (21%)
2	43	female	recurrent	7 cm distant from left subclavian artery	>20	Yes	3	Yes	No	100	5/19(26%)
3	34	female	native	-	>20	Yes	3	Yes	Yes	60	5/14(36%)
4	16	male	native	-	>20	Yes	3	Yes	No	76	3/13 (23%)
5	21	female	native	-	>20	Yes	3	Yes	Yes	64	4/15(27%)
6	45	female	recurrent	Pseudo-aneurysm adjacent to the coarctation site	>20	Yes	2	No	No	70	6/15 (40%)
7	36	female	native	-	>20	Yes	3	Yes	Yes	80	4/14(28%)

HTN= Arterial Hypertension; LVH= Left Ventricular Hypertrophy

RESULTS

Epidemiologic characteristics as well as anatomical and hemodynamic features of CoA of each patient were summarized in Table 1. The mean age of our patients was 32 years, with a majority of females (sex ratio = 0.14). Pre-treatment HTN was found in all cases. Patients were receiving a mean of 3 anti-hypertensive drugs that failed to achieve optimal control in all cases. All patients had a significant blood pressure difference between upper and lower limbs >20 mmHg. Echocardiography revealed left ventricular hypertrophy in all cases except one. Bicuspid aortic valve was observed in three patients without any associated congenital heart defect nor systolic left ventricular dysfunction. Two patients had diagnosed recoarctation (reCoA) at twelve and fifteen years after surgical repair with a pre-stenotic pseudo-aneurysm identified in one of them. The mean baseline trans-aortic gradient was of 73 mmHg and the mean coarctated to normal aorta ratio was of 29%. All patients

weighted more than 35 Kg, allowing a stent placement instead of balloon angioplasty (BA). All patients suffering from native or recurrent CoA were treated with a CS (Covered CP Stent™, NUMED, NY, USA) except one patient treated with a BMS (CP Stent™, NUMED, NY, USA) as coarctation was not isthmic and was 7cm distant from the left subclavian artery with a risk of Adamkiewiczartery damage. Immediate post-procedural result was satisfactory in all cases with a final peak-to-peak trans-aortic gradient of less than 10 mmHg (figure 3). Final coarctated to normal aorta ratio was >50% in all patients with a mean of 73%. There were no aortic complications, nor dissection neither rupture. As well, no local femoral complications were noted despite using large sheath diameters and the unavailability of vascular closure devices. After a mean follow-up of 13 months, all patients were still alive. Residual, but controlled HTN was found in three patients with a mean of one anti-hypertensive drug withdrawal following the procedure. Immediate and long-term follow-up outcomes were summarized in Table 2.

Table 2: Immediate results and long-term follow-up after stenting.

Case n°	balloon diameter (mm)	CS / BMS	Final coarctated to normal aorta ratio	Final transisthmic gradient (mmHg)	Immediate complications (aortic / femoral)	Follow-up dudation (months)	Residual arterial hypertension	Number of anti- hypertensive drugs	Alive
1	15	CS	78%	5	No	24	Yes	3	yes
2	15	BMS	86%	0	No	24	No	0	Yes
3	15	CS	93%	0	no	24	No	0	Yes
4	18	CS	77%	0	No	8	Yes	2	Yes
5	18	CS	69%	0	No	6	No	0	Yes
6	15	CS	68%	5	No	2	No	0	Yes
			+ Exclusion of the pseudo-aneurysm						
7	18	CS	77%	0	No	1	Yes	2	Yes

CS= Covered Stent; BMS= Bare Metal Stent.

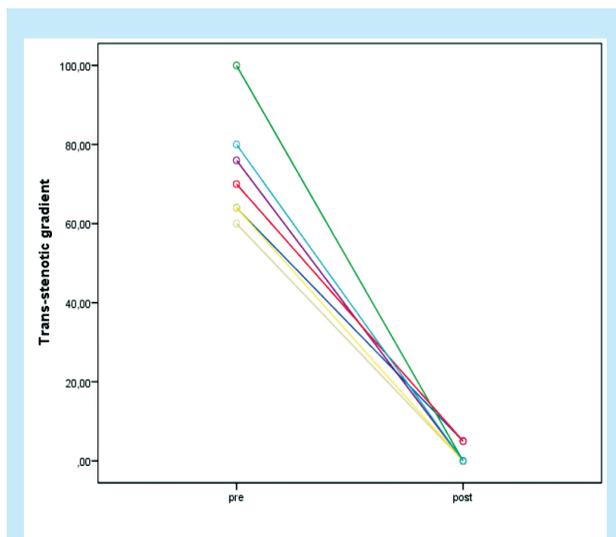


Figure 3: Trans-stenotic gradient evolution before and after aortic stenting.

DISCUSSION

CoA angioplasty is the third common congenital percutaneous intervention (16%) after closure of patent duct arteriosus and atrial septal defect (6). In our experience, all patients undergoing trans-catheter treatment were treated with stent placement. In fact, the long-term of stenting results are better than that of BA alone, with comparatively very lower rates of restenosis and pseudo-aneurysms formation (7). In reported series, there was a tendency to this strategy: 62% in the Congenital Cardiovascular Interventional Study Consortium registry (CCISC) (8) and 54% in a recent meta-analysis (9). Recent American guidelines supported this approach by enlarging the indications of CoA stenting to all cases where a stent can be expanded to an adult size in patients with a trans-catheter systolic coarctation gradient of >20 mmHg or with systemic hypertension associated with an anatomic significant narrowing or in case of balloon angioplasty failure (7). This was based on several recent data are encouraging stent angioplasty over balloon angioplasty (BA) or surgical treatment. CCISC, a multicenter observational registry, showed that stenting was superior to BA in achieving lower post procedural gradient immediately and in the short term follow up (5). According to data from the same registry, stenting of CoA was associated with 96% acute success, 86% intermediate success and 77% long term success (10). Greater immediate success was showed in more recent series approximating 100% (11,12). Likewise, in our case series, the procedure was successful with a gradient less than 10 mmHg in all patients.

Despite the actual agreement that stenting could be indicated as a first line treatment for CoA, whether to use BMS or CS is still a debate issue. A recent randomized controlled trial aimed to compare these two attitudes but it provided no firm answer. In fact the procedure was successful in 100% of cases with no statistically significant difference in terms of recoarctation and pseudo-aneurysms between the two arms (12). Many authors are suggesting the use of CS in particular cases to treat (bail-out) or to prevent aortic wall injury instead of their systematic elective use, and to treat other patients using BMS (13,14). To evaluate this attitude, a recent multicenter trial (COAST II) was designed (15). In COAST II, patients received covered stents for native or recurrent CoA either because of acute or chronic aortic complications or to prevent their occurrence in high risk patients having: nearly atretic aorta, genetic syndrome associated with aortic wall weakening like Turner syndrome or advanced age >60 years. This triage was associated with excellent outcome as trans-aortic gradient fell in all cases with a complete coverage of aortic wall injury in 93% with no reported acute aortic complications, repeat interventions or deaths. Therefore, this selective approach for CS seems to be more reasonable. In our series, CS was implanted in five patients suffering from a quasi-atretic CoA and in one patient presenting a pseudo-aneurysm. The last patient had a CoA located at the descending thoracic aorta far from the isthmus with a risk of Adamkiewicz artery damage, justifying, consequently, the use of a BMS. CoA stenting could be associated with adverse effects. However, when compared to other therapeutic modalities, stented patients had fewer complications than surgical and BA ones: 2.3%, 8.1% and 9.8% respectively in the CCISC registry. In addition, hospitalization was longer in operated patients, and aortic wall injury was more likely in BA group (5). Immediate complications are mainly represented by aortic wall (1.3%) and femoral injuries (1%) (5). Aortic wall complications including dissection or rupture are the most worrying. Our patients did not experience any of these complications. Fortunately the incidence is rare and risk factors are: pre-stent angioplasty, balloon to coarctation ratio >3.5, abdominal CoA and age >40 years (16). Importantly, the vital risk justifies the immediate availability of surgical standby or covered stents which were used successfully to manage reported cases of aortic wall rupture (17). Moreover, intermediate and long-term complications are mainly represented by the development of aneurysms and recurrent CoA. Aortic aneurysms are usually diagnosed at intermediate follow-up in approximately 6% of patients (18) justifying an imaging screening using CT or MRI (19). Balloon to coarctation ratio >3.5 and pre-stent angioplasty were found to be risk factors (5). Most aneurysms are small, only requiring conservative management. In case of size

progression, covered stent placement is required (20). Recurrent CoA is defined as more than 50% narrowing at the site of diaphragm compared to descending aorta (19). Recurrent CoA requiring reintervention are reported to be 4% following stenting (10). Severe intimal stenosis, stent fracture, stent recoil, and adjacent somatic growth are the potential factors requiring reintervention (20).

Controlling blood pressure and the related cardiovascular complications is among the main goals of coarctation repair. However, in our case series, stenting succeeded to achieve normal blood pressure in only four out of seven patients. Likewise, a recent systematic review reported that residual HTN was identified in nearly a third of patients undergoing surgical or catheter-based CoA repair (21) without significant difference between the different therapeutic modalities (5,22). The identification of HTN after CoA repair should prompt imaging assessment for seeking restenosis. Nevertheless, most patients with residual HTN do not have reCoA. Several mechanisms have been proposed to explain this finding including stiffening of central and peripheral arterial walls, impaired endothelial function, altered baroreceptor sensitivity, abnormal pressure waveform reflection and alterations of the rennin-angiotensin system (21).

Finally, in our experience, two patients suffered from recurrent CoA following surgical repair. In this setting, balloon angioplasty has been shown to be the best therapeutic option in case of discrete recurrent coarctation (7). The length and the degree of stenosis in our first case dictated the need of stenting. In the second case, covered-stenting was indicated in front of the development of pseudo-aneurysm at the site of previous repair. In fact recurrent CoA stenting was reported to be effective in >96% in the largest reported series (10,16), and when compared to stenting of native CoA, the outcome was not significantly different in the intermediate and long-term (23).

CONCLUSION

Excellent outcomes support that treating CoA with stent placement represent currently an emerging alternative to classic therapeutic modalities: surgery and BA. The use of CS provides good results, but a reasonable selection of high risk patient seems to be wiser. A long term follow-up after CoA angioplasty is mandatory to screen complications like reCoA, aneurysms and residual hypertension with its cardiovascular impact in order to manage them properly and improve patients' prognosis.

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