CASE REPORT

Treatment of a recurrent renal artery stenosis and stent fracture using a drug eluting stent in a pediatric patient

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Abstract Renal artery obstruction is uncommon in the pediatric population and therefore represents a challenging diagnosis to ascertain and treat. While angioplasty and surgery have been used to treat it, stent implantation has been left mainly for adult use. In the following paper, we expose the case of an 8-year-old female who presented with severe arterial hypertension. Initially, the patient was evaluated and up to 5 antihypertensive medications were started. After complete evaluation, she was found to have severe right renal artery distal and proximal stenosis. She underwent recurrent angioplasties and then a bare metal stent (BMS) implantation, due to recurrence of stenosis. The original BMS developed a circumferential fracture, leading to a second stent implant, this time using a drug eluting stent (DES). She has been stable for 8 years since the last intervention, although using 2 antihypertensive medications. The use of BMS and DES may add another tool in the armamentarium of the pediatric interventionist to treat these children with recurrent stenosis that fail angioplasty.

Keywords Stent · Pediatrics · Renal artery · Stenosis · Hypertension

Introduction

In children, renal artery stenosis (RAS) is most commonly caused by fibromuscular dysplasia, although it may also appear in association with neurofibromatosis, Takayasu's arteritis, Moyamoya disease, thrombosis, and vascular malformations [1]. Rarely, the most common cause of RAS in adults, which is atherosclerosis [2] may also be observed in the pediatric population [3]. The physician's goal with these patients must be to control blood pressure and avoid future complications as is usually seen with long standing hypertension and renal failure. Treatments include medications, surgical procedures, and, for approximately the last 2 decades, percutaneous interventions [4–6].

Case presentation

An 8-year-old female, presented to the Emergency Department with severe arterial hypertension. The blood pressure on admission was 200/110, heart rate 89, she was afebrile and in no respiratory distress. Her weight was 100 pounds (47.7 kg) and her height 4'8" (140 cm). Her only complaint was headache. A total of 5 antihypertensive medications (labetalol and enalapril orally, clonidine patch, and hydralazine and nitroprusside IV) had to be added over several days to finally control the blood pressure while evaluation was carried out. A magnetic resonance angiogram (MRA) performed revealed right RAS. Initially, the complete blood count was normal; a complete metabolic panel showed no abnormal values. Electrolytes,

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including blood urea nitrogen and creatinine levels were also normal.

An arteriogram was performed under intravenous sedation and with biplane fluoroscopic guidance. The right femoral artery was cannulated with a 4Fr introducer sheath (Cordis Co. Johnson & Johnson; Miami, FL, USA) using the modified Seldinger technique. With a 4Fr Pig-Tail catheter (Cordis Co, Johnson & Johnson; Miami, FL, USA) and a hydrophilic 0.035" guide wire (Terumo Cardiovascular, USA), abdominal aortograms, by power injection of contrast, were carried out. The right RAS, which included the ostium, was confirmed. The aorto-renal gradients were measured using a 4Fr Cobra-1 catheter (Cordis Co, Johnson & Johnson; FL, USA). The right-sided gradient was approximately 100 mmHg. The left renal artery had no gradient. Then, selective bilateral renal arteriograms, by hand injection of an equal amount of contrast and normal saline 0.9 % mixture (1:1), were done using the same catheter. Approximately, 2–3 ml of the contrast solution was used for each one of the injections. After the stenosis was fully examined, the angioplasty was performed. For this a 4 mm × 2 cm, Mini Ghost balloon catheter (B. Braun, Bethlehem, PA, USA) was advanced over a 0.014" Whisper guide wire (Abbott Vascular, USA) that had been pre-positioned in the right renal arterial lumen via the Cobra-1 catheter. Inflations were done up to 20 atmospheres of pressure. Post-angioplasty angiograms were done with the Cobra-1 catheter to evaluate the result. It showed an excellent result. When finalized the study, the catheters and introducer were removed and pressure applied over the puncture area to achieve hemostasis. Following the procedure there was significant improvement of the gradient from the aorta to the right kidney. This leads to a reduction in blood pressure. Medication requirements consequently decreased as well, with dosage adjustments done according to the blood pressure on follow-up visits.

Unfortunately, the stenosis recurred twice in less than 2 months, elevating her blood pressure as high as 190/110 mmHg, requiring 2 additional angioplasties. Meanwhile, her renal function tests remained normal, whatsoever. On the 3rd occasion, about 6 weeks later, it became clear that alternative treatments would have to be considered. Of importance is to say, that in all the interventions the same materials were used, except for those that will be described. An intravascular stent became the preferred option in our clinical judgment. So, at this time, and following the same previously described technique, a 6 mm × 18 mm pre-mounted, Herculink BMS (Abbott Vascular, Santa Clara, CA, USA) was implanted, and dilated to 4 mm (Fig. 1). Approximately, 2 months after the initial implantation of the BMS (5.5 months after presentation), elevation in the blood pressure to 190s/100s, suggested recurrence of the obstruction; this was confirmed by

renal duplex study. She was taken back to the catheterization suite. Arteriograms were repeated and it was found that the patient had severe intimal proliferation within the lumen of the stent. Once again the stent was redilated with a high-pressure, 4 mm × 2 cm Mini Ghost balloon catheter (B. Braun, Bethlehem, PA, USA) after pre-dilatation with a 4 mm × 2 cm cutting balloon catheter (Boston Scientific, Natick, MA, USA). The final lumen was adequate and the pressure gradient had decreased to less than 10 mmHg.

Eight months after initial presentation, a second dilatation of the BMS had to be done. This time, it was noted that the stent had a circumferential fracture in the middle. It had become totally separated and imbedded into the vessel wall (Fig. 2). The re-dilatation of the vessel was performed using another cutting balloon catheter and a high-pressure catheter. The lumen of the right renal artery and pressure gradient improved with this intervention. However, before 6 months had passed, the patient returned with a 3rd recurrence of RAS. After discussing the case with the parents and their wish to avoid surgery, the proper informed and written consent, for off-label use, to implant a drug eluting stent (DES) was obtained. Following the same catheterization technique, a 3.75 mm × 18 mm Taxus stent (Boston Scientific, Natick, MA, USA) was chosen and implanted in the right renal artery. It was easily over-dilated to 4 mm with a high-pressure Mini Ghost balloon and excellent results were appreciated immediately. The gradient was zero. She was already on an 81 mg aspirin regimen at this point, and now clopidogrel 37.5 mg orally was added. After 6 months, another angiogram was performed to evaluate the DES. Only minimal intimal proliferation was observed (Fig. 3). The gradient had increased slightly to a maximum of 10 mmHg. No other intervention was performed at that time. The patient was continued on aspirin 81 mg and clopidogrel 37.5 mg orally daily, and antihypertensive medications: labetalol 100 mg daily and nifedipine XL 60 mg daily. Eight years have passed since the implantation of the DES. Currently she has become obese for her age, with a body mass index of 32/m². Now on only 2 antihypertensives at lower doses: (labetalol 50 mg orally daily and nifedipine XL 30 mg daily), she has a controlled blood pressure. Follow-up of the renal artery gradient has been done routinely with duplex ultrasound once every 6 months for the first 4 years and then yearly, thereafter.

Discussion

Renovascular hypertension (RVHtn) may be caused by renal artery stenosis (RAS), which can be unilateral or bilateral. It causes a decrease in renal blood flow, activating

Fig. 1 Pre (a) and post (b) photos showing the implantation of the 1st bare metal stent in the right renal artery

Fig. 2 Details of the fractured bare metal stent in the right renal artery. See how the stent is separated *right in the middle* (arrows)

Fig. 3 Aorto-renal arteriogram performed 6 months after the implantation of the drug eluting stent, inside the bare metal stent. See that the intimal proliferation is essentially inexistent

the renin–angiotensin system and leading to systemic hypertension. RVHtn has an aggressive course leading to progressive malignant hypertension resulting in ischemia of the affected kidney [7]. Patients with RVHtn may be asymptomatic or they may present with nonspecific

symptoms related to the end-organ effects. Common symptoms found among patients are headache, change in mental status, seizures, hyperirritability, hyperexcitability, fatigue or lethargy. The presence of the following four factors must trigger a high index of suspicion: first, when

diastolic hypertension (>100 mmHg) develops in a patient younger than 30 years or older than 55 years; second, when a pre-existing stable hypertension suddenly accelerates; third, the presence of an abdominal bruit found during a physical exam; and fourth, a rapid progression to malignant hypertension within a period of 6 months [7–9].

Treatment for RAS is controversial. The starting treatment is pharmacological, composed of antihypertensive drugs and diuretics. Its management is based on the control of high blood pressure and the preservation of kidney function. In those patients, where the hypertension cannot be controlled or there is presence of a progressive renal dysfunction, revascularization of the artery is considered. This can be achieved through percutaneous angioplasty and surgical interventions. In severe cases, where none of the previous treatments is successful, a nephrectomy is an alternative [1–9].

Percutaneous transluminal renal angioplasty has been considered the treatment of choice for revascularization [1, 4–6]. In cases, angioplasty had been reported as very useful, but other authors have reported poor long-term results needing the use of stents [1, 10–15]. In our patient, the angioplasties performed obtained an excellent immediate result, but really poor intermediate or long-term results. The use of stents has proven to be of great benefit for the arterial revascularization, improving the renal function and controlling the hypertension. It has become the recommended treatment for severe stenosis and difficult lesions such as ostial and restenotic lesions [1, 4–7]. We took this line of treatment for our patient, since the preferred route was less invasive, and the angioplasty alone was not successful at long term as desired. However, authors have reported that the use of BMS may have a high incidence of restenosis [5]. DES or surgery has been used for such cases. Surgery is considered when the angioplasty has failed to revascularize the obstruction or for patients with occluded renal artery and functional but ischemic kidneys [7]. The reconstitution of blood flow through surgery has a high risk of morbidity and mortality. These patients have a 3–6 % risk of mortality, which increases with the presence of other comorbid conditions [8].

Drug eluting stents are normal metallic stents coated with a pharmacological agent that interferes with the process of restenosis [10–12]. The use of DES is controversial, and there is not much information on the use of these stents especially in the pediatric population [1, 4]. The use of the DES was chosen since the angioplasties and even the use of a BMS were not successful. In our patient the DES reduced the restenosis rate and at 6 months there was still excellent flow with minimal intimal proliferation. Even though, there is controversy of the high risk of thrombosis of these stents, there is information that recommends the use of these DES with dual antiplatelet therapy decreasing the chance of a thrombotic event [12]. The use of DES has been confined

to the coronary arteries. Very limited number of cases have been reported describing their use in other anatomical places. However, more clinicians have been using them to treat recurrent stenosis of the renal arteries in adults [13]. Other physicians have also used polytetrafluoroethylene (PTFE) covered stents for the treatment of recurrent stenosis [14]. There is no indication for the use of a DES outside the coronary system. However, there have been reports of their use in the peripheral-vascular vessels and even renal arteries in adults. So, this experience was used and applied to our case for its use. In our case, given the recurrent stenosis in spite of repeated angioplasties, BMS placement, and redilatations, it was determined that when the DES was implanted she would remain on dual antiplatelet medications for at least 1 year.

Another problem encountered with our patient was the mid-stent circumferential fracture, which was not related to the cutting balloons used to dilate the stenosis caused by the intimal proliferation. Fracture of stents have been reported by other authors as well, [14, 15] although there have been only a few reports of stent fractures in the renal arteries. Chua and Hung [15] suggested as the renal artery entrapment by the diaphragmatic crus, and a mobile kidney with an acute angulation at the proximal segment of the renal artery, as the 2 possible reasons for stent fracture. The reason mentioned by Guzzetta and his co-workers, [16] also dealt with the motion of the renal artery. Robertson et al. [17], found stent fractures more common in the left than the right, possibly due to the structures associated to each kidney. For example, the right kidney is less mobile due to its localization in between the right-sided inferior vena cava and posterior to the liver.

Conclusion

We present the unique case of a pediatric patient with recurrence of severe stenosis of the right renal artery, even though it was treated with angioplasty and a stent placement. The use of a DES improved the stenosis providing better control of the hypertension and the renal function. The use of DES could be considered as another tool in the treatment of recurring RAS.

Conflict of interest The authors have declared that no conflict of interest exists.

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