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## Case Reports

# Aortic rupture during aortoplasty in Takayasu arteritis – A rare complication: Case report and review of literature



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## ABSTRACT

Balloon angioplasty of the stenosed aorta is usually a relatively simple, yet potentially a catastrophic procedure. Aortic rupture during aortoplasty, though uncommon, carries a high mortality. We report case of a 39-year-old female with aortoarteritis with multiple arterial stenoses whose infra-renal abdominal aorta ruptured during balloon dilatation of the stent deployed in that segment. The site of aortic rupture was temporarily occluded by low-pressure inflation of the same balloon and then was sealed using a stent-graft introduced by contra-lateral femoral arterial access.

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## 1. Introduction

Aortic rupture is a catastrophic complication during angioplasty and stenting of the aorta.<sup>1</sup> Surgical salvage is possible only in minority, due to inherent procedural delay.<sup>2–4</sup> Of late, endovascular grafts have been tried successfully<sup>5–7</sup> but at times the patient may collapse due to torrential blood loss during the time such a life-saving procedure is being instituted.<sup>8</sup> Here we present the case of a young female with aortoarteritis (Takayasu arteritis) with multiple arterial

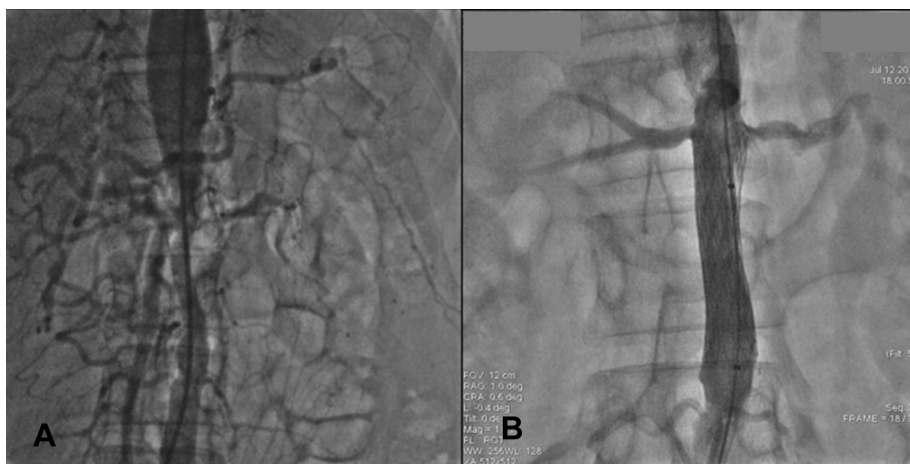
occlusions/stenoses, including that of the descending thoracic aorta and abdominal aorta. Soon after balloon-dilatation of the infra-renal aortic stent, there was a haemodynamic collapse due to aortic rupture which was managed by temporary balloon occlusion followed by covered stent deployment from another vascular access; this lifesaving strategy drastically reduced blood loss and quickly restored haemodynamic stability permitting unhurried deployment of aortic stent graft. The technique has not been reported to be used so far in the above setting.

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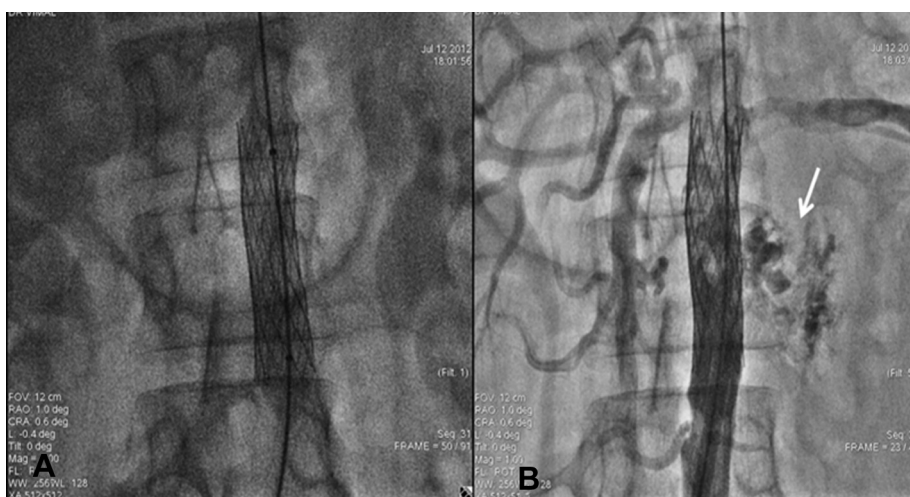


**Fig. 1 – A. Aortogram showing stenosis of the infra-renal abdominal aorta. B. Aortogram showing the deployed Palmaz peripheral stent in the infra-renal aorta with residual waist.**

## 2. Case report

A 39-year-old female presented with resistant hypertension and lower limb claudication. She had feeble femoral pulses and an abdominal bruit. Her blood pressure was 210/110 mm Hg in the right arm and 120/70 mm Hg in the lower limbs in supine position. The laboratory investigations showed hemoglobin of 12.3 g%, ESR was 15 mm in the first hour and C reactive protein was negative suggesting no activity. Angiographically, she had 99% stenosis of right carotid artery, 100% stenosis of left carotid artery, 90% stenosis of left renal artery, and significant stenosis of aorta at thoracic (T<sub>10–11</sub>) and at infra-renal levels (L<sub>1–2</sub>) (Fig. 1A). At catheterization, peak gradient across thoracic aortic stenosis was 45 mm Hg while across the abdominal aortic stenosis was 40 mm Hg. After a written and informed consent, balloon angioplasty of the left renal artery was performed with a good angiographic result without stenting. A week later, she was taken up for aortoplasty. Percutaneous right femoral arterial access was obtained and an 8F sheath was inserted and a 20G cannula was

inserted in the left radial artery for blood pressure monitoring. 5000 U of unfractionated heparin was used for anticoagulation. Aortogram was done with 7F pigtail catheter in antero-posterior and lateral views to delineate the stenotic and the reference segments via the femoral route. The thoracic aortic lesion was dilated with a 12 × 40 mm balloon (Maxi LD, Cordis, The Netherlands). A 14 × 40 mm Palmaz stent (Cordis Corporation, Miami, USA) manually crimped on a Maxi LD balloon (14 × 40 mm) was taken through an 11F sheath and deployed at 4 ATM pressure. The stent was further dilated with the same balloon at 8 ATM pressure. After this the lesion in the infra-renal abdominal aorta was dilated with a 12 × 40 mm balloon at 4 ATM pressure and subsequently, 14 × 50 mm Palmaz stent manually crimped on a 14 × 40 mm Maxi LD balloon was deployed across the lesion at 4 ATM pressure (Fig. 1B). The shorter length balloon was used as 50 mm or longer length balloon was not available. The longer stent on a shorter balloon could be deployed by the method already described.<sup>9</sup> The stent was further dilated with the same balloon at 8 ATM pressure (Fig. 2A). The patient



**Fig. 2 – A. Aortogram showing further dilatation of stent after deployment with 14 × 40 mm Maxi LD balloon. B. Aortogram showing aortic rupture with extravasation of blood from aorta (arrow).**

complained of back pain during the balloon inflation. On deflating the balloon, the systolic blood pressure fell dramatically from 200 to 100 mm Hg. Angiogram showed a torrential leak from the left side of the stent (Fig. 2B). The same angioplasty balloon was immediately positioned at site of the leak and inflated at 3 ATM so as to achieve balloon tamponade. Intravenous protamine 50 mg was given. Blood pressure was seen to stabilize at around 120 mm Hg systolic. As there was a risk of hemodynamic collapse if the balloon was deflated and removed to allow introduction of a stent-graft from the same vascular access, 8F left femoral artery access was obtained and a 300-cm 0.035" guidewire was advanced up to the inflated balloon. The balloon was transiently partially deflated to permit further rapid cranial advancement of the 0.035" wire till the aortic arch, after which the balloon was quickly re-inflated. Then, the left femoral artery sheath was exchanged with an 11F Mullins sheath (Cook Inc., Bloomington, USA) and its distal end was positioned just below the lower end of inflated balloon (Fig. 3A). Then a 12 × 61 mm balloon-expandable stent-graft (Advanta V12, Atrium Medical Corporation, New Hampshire, USA) was positioned at the site of aortic rupture via the Mullins sheath using fluoroscopic guidance alone, after partial deflation of the occluding balloon. After confirming the position of the stent-graft by matching its proximal end with the deployed stent, the tamponading balloon was fully deflated and pulled out along with its guidewire (Fig. 3B) and the stent-graft was rapidly deployed at 6 ATM pressure. The check angiogram confirmed complete sealing of the leak (Fig. 4A and B). There was no gradient at any level in the thoracic or abdominal aorta. Patient did not have any significant fall in hemoglobin with serial hemoglobin levels remaining around 11.5 g%. Her further hospital course was uneventful. At six months follow up she continues to be asymptomatic and her hypertension is controlled with two anti-hypertensive drugs.

### 3. Discussion

The concept of balloon aortoplasty was first reported for coarctation of the aorta in 1982 by Singer et al.<sup>10</sup> In

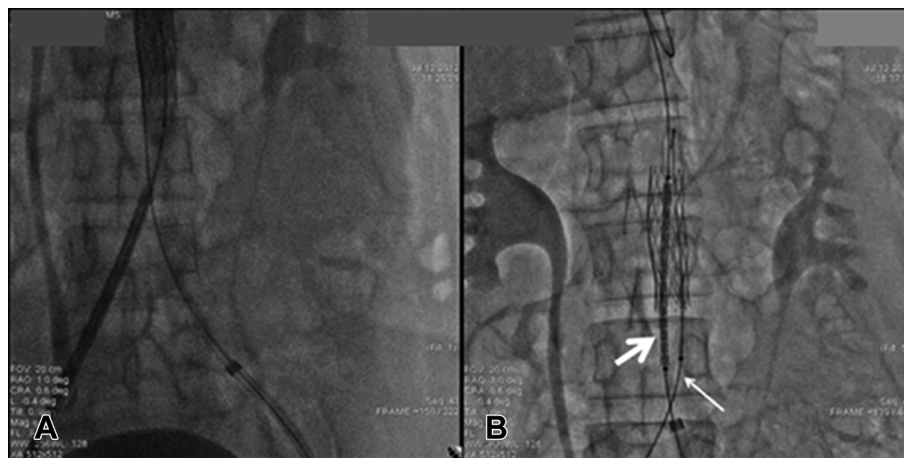
aortoarteritis, it was first performed by Yagura et al in 1984.<sup>11</sup> It is generally considered to be a safe and effective procedure with low incidence of aortic rupture which if it occurs is attended with high mortality.

After extensive search, we could find 13 published cases of aortic rupture during aortoplasty; 10 cases in coarctation (Table 1) and 3 cases in aortoarteritis all of whom had undergone an intervention in the past (Table 2). Ours is probably the first reported case of aortic rupture in a patient with aortoarteritis while undergoing first intervention. Of the three cases of aortic rupture in aortoarteritis, two died and one survived following stent-graft repair, emphasizing the grave prognosis of this dreaded complication.<sup>5</sup>

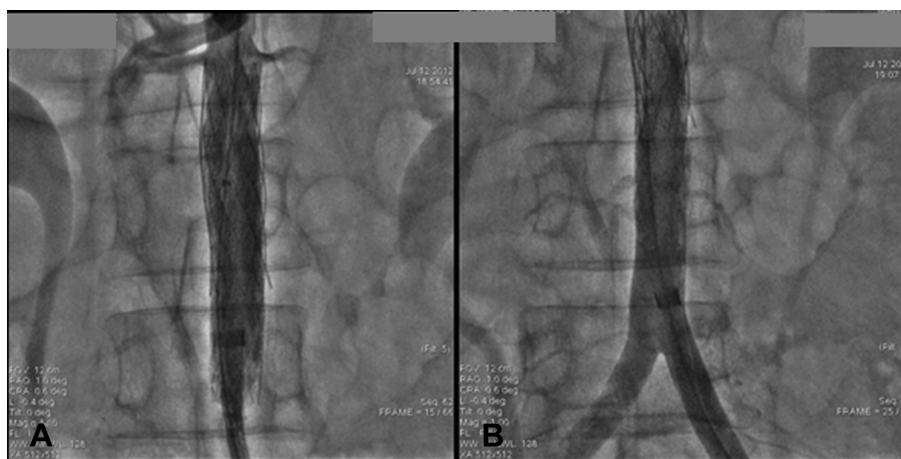
In aortoarteritis, the vessel is tough, noncompliant & rigid due typically to involvement of all the three layers of the vessel wall and requires prolonged, repeated balloon dilatation at a higher pressure (as compared to coarctation).<sup>17</sup> Conversely, due to more fibrosis and thickening of aortic wall in aortoarteritis, it is less likely to rupture unlike congenital coarctation of aorta. However in aortoarteritis much higher inflation pressures are typically required for adequate lesion dilatation and hence the rupture rate may in fact be higher. Unless managed immediately and appropriately, aortic rupture is more likely to be fatal.<sup>1–8,12–15</sup>

In coarctation, there is medial thickening and intimal hyperplasia forming a ridge that encircles mostly posteriorly and laterally within the lumen of the descending thoracic aorta.<sup>18</sup> Chances of aortic rupture are more with coarctation since there is cystic medial necrosis in the thickened media. The medial layer in the coarcted segment contains fragmented elastic fibers, increased connective tissue and decreased smooth muscle cells which predispose to rupture.<sup>1,19</sup>

Since aortic rupture results in rapid exsanguination, immediate, systematic and planned management is of paramount importance to prevent death. As already stated in literature<sup>18</sup> and reemphasized by us, aortoplasty should be undertaken only after appropriate size stent-grafts are available on shelf. This is important since patient may have fatal outcome even if surgery is undertaken.<sup>4</sup> Our case demonstrates that dual vascular access can be very effectively used to prevent blood loss from the rupture site, while stent-graft is



**Fig. 3 – A. Mullins sheath being advanced from left femoral access. B. Showing the stent-graft (thick arrow) being taken across the stent while deflated balloon is being withdrawn (thin arrow).**



**Fig. 4 – A. Aortogram showing the sealing of aortic leak. B. Final aortogram showing well deployed stent-graft with no obstruction in aorta and no extravasation.**

**Table 1 – Reported cases of aortic rupture in coarctation of aorta.**

S. no.	Author	Year	Age (yrs)/sex	Previous procedure	Modality to treat rupture	Result
1	Kulick et al <sup>12</sup>	1990	30/F	None	Surgery	Saved
2	Balaji et al <sup>2</sup>	1991	8/M	Patch aortoplasty	Surgery	Died
3	Roberts et al <sup>13</sup>	1993	25/M	End to end anastomoses	Surgery	Died
4	Varma et al <sup>1</sup>	2001	65/F	None	Surgery planned	Died
5	Korkala et al <sup>8</sup>	2002	44/F	None	Stent-graft attempted followed by surgery	Saved
6	Tan et al <sup>7</sup>	2005	33/F	End to end anastomoses	Stent-graft	Saved
7	Collins et al <sup>14</sup>	2006	51/F	None	Covered stent over covered stent	Saved
8	Alcibar et al <sup>5</sup>	2007	62/F	End to end anastomoses	Stent-graft	Saved
9	Wu et al <sup>15</sup>	2012	9/F	Patch aortoplasty	Iliac limb graft	Saved
10	Alcibar J et al <sup>16</sup>	2013	-/F	None	Covered stent	Saved

**Table 2 – Reported cases of aortic rupture in Takayasu arteritis.**

S. No.	Author	Year	Age (yrs)/sex	Previous procedure	Modality to treat rupture	Result
1	Sharma et al <sup>3</sup>	1995	32/F	Patch aortoplasty	Surgery planned	Died
2	Tyagi et al <sup>5</sup>	2003	32/F	Aortic stenting	Stent-graft	Saved
3	Deshmukh et al <sup>4</sup>	2003	30/F	Patch aortoplasty	Surgery	Died

being deployed. In our case, no blood transfusion was required. Finally, the aim of aortoplasty should not be cosmetic result, but should be guided by reduction in gradient. In fact a small waist in the deployed stent will prevent stent migration. Aggressive high pressure post-stent balloon dilatation to achieve a cylindrical contour of stent is best avoided.

Balloon angioplasty for treatment of native coarctation has been reported to have relatively high rate of recoarctation and aneurysm formation.<sup>18</sup> In contrast, aneurysm at the site of angioplasty after balloon dilatation of the aorta in patients with aortoarteritis is rare, which could be due to marked thickening aortic wall due to fibrosis of all three layers.<sup>17</sup> Balloon oversizing, higher inflation pressure, balloon rupture and wire manipulation, have all been implicated in the possible mechanisms of aortic rupture. Endovascular stent-grafts have been used as a less invasive alternative to surgery for managing aortic aneurysms, pseudoaneurysms, dissection of aorta, blunt thoracic aortic injury, and peripheral arterial aneurysms.<sup>5</sup> These endovascular stent-grafts are a

must in the inventory of any cardiac catheterization laboratory doing aortoplasty. Although the use covered stents or stent-grafts have been reported in aortoplasty to prevent aortic dissection, rupture and late aneurysm formation, its use as a routine strategy has the disadvantage of larger vascular access requirement for deployment and the risk of obstructing important branch vessels. Further accurate sizing of stent-graft is important as they cannot be significantly dilated beyond their nominal diameter.<sup>16</sup> Nevertheless stent-grafts may be used electively in patients with complex aortic anatomy.<sup>16</sup>

#### 4. Conclusion

Aortic rupture during aortoplasty is a very rare catastrophic complication of aortoplasty which was successfully managed in our case by dual vascular access technique, wherein another vascular access is used to deliver the stent-graft while



the perforation site is kept temporarily sealed by a balloon introduced through the initial vascular access. This technique has not been reported in the management of a catastrophic aortic leak previously.

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### Conflicts of interest

All authors have none to declare.

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