

Brief Report

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Abstract

We report a neonate with aortic arch hypoplasia and coarctation, in whom patency of the arterial duct could not be re-established and who was too ill to undergo primary surgical correction safely. This patient was treated in two stages: 1) angioplasty and stenting, 2) surgical correction. The safe period for surgical correction may be 2–3 weeks after angioplasty and stenting.

Neonates with aortic arch hypoplasia and coarctation are usually offered a primary surgical correction. However, when such patients are critically ill and patency of the arterial duct cannot be re-established to achieve pre-operative stability, primary repair may be hazardous. Such cases may be treated more safely in two stages^{1–4}:

1. Angioplasty and stenting to stabilise the patient
2. Elective surgery for removal of stents and arch repair when the patient is clinically stable

This two-stage approach raises the following question: when should the second procedure be done? If done very soon, stent removal would be easy, but the aortic wall may be fragile due to inflammation caused by angioplasty such that it may be torn by the act of suturing.^{3–6} If done late, the aortic wall will not be inflamed anymore, but the stents may be difficult to remove because these become covered with neo-intima; stent removal may tear the arch. Alternatively, the second stage may be done safely much later if the stented segment is not too long such that it may be excised (together with its stent) without compromising the repair. Either way, the stent(s) should be removed completely in order not to interfere with the growth of the aorta.

When the stented segment is too long to be excised, the second procedure should be done soon so that the stent(s) may be removed safely, but not so soon that the aortic wall would still be friable. This dilemma is highlighted in various publications, but none specifies when is too soon and when is too late. This question cannot be fully answered yet, but, with the approval of our ethics committee, we report a case that may help in this decision.

Case report

A 3-week-old girl weighing 3.4 kg presented with circulatory collapse, multiorgan failure, and episodes of seizures. She had severe aortic coarctation, distal aortic arch hypoplasia (Fig 1A), and severe biventricular dysfunction (left ventricular ejection fraction <15% and Tricuspid Annular Plane Systolic Excursion of 3 mm). There was a common origin of the right brachiocephalic and left carotid arteries. The diameters of the distal arch and isthmus were 3 and 1 mm, respectively. There were no other malformations. Her arterial duct was very small and did not respond to prostaglandin infusion. Her pupillary reflex and electroencephalogram were normal during periods between seizures. She was treated with emergency percutaneous angioplasty. Two metallic bare coronary stents (Resolute OnyxTM, Medtronic), 18 and 15 mm long, were implanted in series covering the entire stenotic zone and were dilated up to their maximum diameter of 4 mm. The baby's condition improved rapidly, and her cardiac function became normal. She was extubated the following day and discharged from the ICU a day later.

In planning the second stage, the option of excision of the stented segment was ruled out because this was too long (Fig 1B and C). Therefore, the second stage had to be done soon. However, considering the duration of an acute inflammatory reaction as seen typically in wound healing (about 2 weeks), and based on our experience with stents in other sites in older patients, we decided to wait 2–3 weeks for the aortic inflammation to subside.

The operation was done 18 days after stenting. The aortic wall was in excellent condition. Also, the stents were not covered with neointima and could be removed with gentle traction

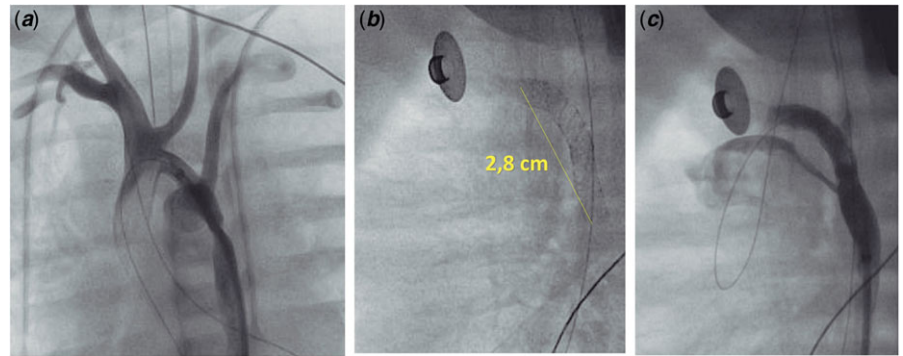


Figure 1. (a) Angiogram showing hypoplasia of the distal aortic arch and coarctation. (b) Stents covering the long stenotic zone. (c) Final result after angioplasty and stenting.

without tearing the arch. The aortic segment affected by coarctation (the isthmus) was excised and the arch was reconstructed with an extended end-to-end anastomosis combined with augmentation of its undersurface with a pericardial patch (Curved No-React® Porcine Patch, NRPP-11m09, BioIntegral Surgical). The outcome was excellent: no residual stenosis, normal blood pressure, good biventricular function, and no neurologic deficit.

Comment

We do not know the exact safe time interval for this procedure; there is very little information published about this.^{1–6} In these studies, most cases were managed in one of two ways:

1. The entire stented segments were removed followed by end-to-end anastomosis. This is a very good solution without any time pressure to proceed to the second stage. However, this is not always possible, specifically when the stented segment is too long, which was the problem in our patient.
2. When the stented segments were too long to be excised, these were incised longitudinally and laid open without removing the stents (because these were firmly embedded in neointima) followed by patch augmentation of this zone. This is suboptimal because stent material left in situ may possibly interfere with growth of the aorta. Ideally, stents should be removed completely, either alone or en-block with the stented aortic segment.

In the few cases where the stents were removed completely without excising the aortic wall, these were done at no more than 2 months following stenting, but stent removal was often not easy. In one case, after 38 days following stenting, the stent could not be removed such that the “incise and patch” repair technique was used.⁴

These reports present excellent results but have not specifically tried to identify the time frame for safe stent removal. Nevertheless, one of these reports states that stents may be removed very easily if the procedure is done only a few days after stenting.³ Therefore, we estimated that the safe period may be between a few days and 1 month. We also drew from our own unpublished experience in other circumstances where we had seen that stents may be removed easily 2 weeks following stenting. These circumstances include

stents in the pulmonary arteries and the right ventricular outflow tract. Overall, we estimated that 2–3 weeks may possibly be the ideal period. Although this empirical estimate was based on small numbers, we trusted it and did the procedure on the 18th day following stenting; the stents were removed very easily. We report this to add to the existing information on this matter, which is very little at present, hoping that the body of information will gradually grow such that one day the safe time interval may be identified with precision.

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Conflicts of interest. None.

Ethical standards. The authors assert that all procedures contributing to this work comply with the ethical standards that are shared between all the ethics committees in the state of Andalucía in Spain and with the Helsinki Declaration of 1975, as revised in 2008, and has been approved by the ethics committee of the state of Andalucía, Spain.

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