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STRUCTURAL HEART DISEASE (RJ SIEGEL AND NC WUNDERLICH, SECTION EDITORS)

The Challenges of Redo Aortic Coarctation Repair in Adults

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Abstract

Purpose of Review Aortic coarctation is a common congenital abnormality causing significant morbidity and mortality if not corrected. Re-coarctation or restenosis of the aorta following treatment is a relatively common long-term problem and the optimal therapy has not been elucidated. In this review, we identify the challenges associated with and the optimal management for recurrent aortic coarctation and the most appropriate therapy for different patient cohorts.

Recent Findings Open surgery provides a durable long-term aortic repair, however, given the complex nature of the procedure, has a somewhat higher rate of serious complications. Endovascular repair, although less invasive and relatively safe, has limitations in treated complex anatomy and is more likely to require repeat intervention.

Summary Open surgical repair is more appropriate for infants that have not been intervened on and endovascular therapy should be reserved for older children and adults and those that require repeat intervention.

Keywords Coarctation · Aorta · Pediatric · Congenital · Long-term · Outcomes

Introduction

Aortic coarctation is a common congenital condition characterized by stenosis, most commonly distal to the left subclavian artery, but may be associated with variable severity of hypoplasia of the aorta, including the arch [1••]. It accounts for approximately 5–7% of all congenital cardiovascular defects with an incidence of 3 per 100,000 live births [2, 3]. Although it can be associated with other genetic congenital disease such as Turner syndrome, it is usually sporadic, with predominance in male patients by 1.5:1 [4]. It may cause isolated hypertension, aortic aneurysm or dissection, cerebrovascular aneurysm, coronary artery disease, or heart failure in severe cases [5]. When patients do not undergo repair, there is a decrease in life expectancy; therefore, most patients that require intervention undergo repair early in life or when initial diagnosis is made [1••].

Late complications such as restenosis and aortic aneurysm formation are relatively common [6, 7•, 8]. When late complications occur, intervention such as open surgical or endovascular repair may be utilized. Each modality has associated benefits and drawbacks and must be considered on an individual basis when deciding the optimal treatment plan for a patient. This purpose of this review is to detail the surgical treatment options available for repair as well as decision-making when approaching late complications of re-coarctation in the adult patient. For the purposes of this review, we will discuss treatment options for isolated and discrete aortic coarctations without concomitant aortic arch hypoplasia or intra-cardiac defects that require repair.

Types of Initial Repair

Open surgical repair of aortic coarctation remains the gold standard of care in neonates [9]. For isolated and discrete coarctations, it can usually be approached and repaired via left thoracotomy, excision of the disease aorta (coarctectomy), and reconstruction, typically by extended end-to-end anastomosis. Alternative reconstructive techniques include primary end-to-end anastomosis, use of a

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subclavian artery flap, or repair with interposition graft. In neonates and young children, surgical repair is associated with less risk of reintervention and is therefore the preferred modality of treatment [9].

With the advent of endovascular techniques to treat aortic disease, coarctation of the aorta in older patients is increasingly being treated with minimally invasive catheter-based modalities. Endovascular treatment of aortic coarctation is a preferred method of treatment in many advanced centers and is recommended as an initial therapy for older, adolescent, and adult patients, presenting with native coarctation amenable to such repair [1••, 3, 10]. Balloon angioplasty with or without stent placement can be used to relieve the stenosis. It is recommended that stents are placed only in those patients that have grown to sufficient size to allow for implantation of an adult-sized prosthesis that will not be associated with ongoing stenosis if the patient continues to grow [9, 11–13].

Long-Term Complications Following Repair of Aortic Coarctation

Re-coarctation or restenosis following primary repair is an unfortunate but common long-term complication. Following open surgical repair, the rate of restenosis is approximately 5–15% [6, 7•, 8, 14, 15]. Restenosis does appear to be related to the type of surgical technique utilized to repair the aorta and modified surgical techniques have been created to minimize this complication [16–18]. Specifically, the patch aortoplasty has the highest rates of both restenosis and aneurysm formation, 20–30% and 20–40%, respectively.

Following endovascular balloon angioplasty without stent placement, re-coarctation has a variable incidence and is related to the age at which the original intervention was performed. In neonates and infants, the rate of restenosis is reported up to 50% [19, 20]. In older children, restenosis occurs in 20–30% of patients, and in teenagers and adults rates are as low as 9% [11, 20–23].

Aortic aneurysm following repair of coarctation may occur at the site of previous repair or proximal to it. It is hypothesized that aneurysm formation at the site of previous repair is likely secondary to the damage to the aortic wall including a reduction of its elastic fiber constituent, smooth muscle fibers, and/or overgrowth of scar tissue [24]. Risk factors associated with aneurysm repair include older age at the time of initial repair and the type of repair performed, as discussed below.

The rate of aneurysm formation following open repair ranges from 1 to 9% depending on the series [21, 25–27]. There is a known predilection for late aneurysm formation depending on the type of repair initially performed. Patch angioplasty has been shown to have the highest rate of late aneurysm formation, up to 9% at 24 years of follow-up in one study [25, 26]. In contrast, series evaluating surgical repair with primary anastomosis report aneurysm formation in approximately 1% of patients [25].

Aneurysm formation following endovascular repair of a native coarctation has less consistent results. One study evaluating balloon angioplasty alone in adolescents and adults noted aneurysm formation in 8% of patients at one-year post treatment [21]. Another, evaluating balloon angioplasty noted zero aneurysms with a mean follow-up of 8.5 years [14].

Indications for Repair of Re-coarctation

The indications to intervene on a patient that has re-coarctation are the same as the native disease. They include a trans-coarctation gradient of > 20 mmHg, persistent hypertension not attributed to another cause, radiologic evidence of clinically significant collateral flow, or heart failure attributed to the presence of the coarctation [9, 28, 29]. Criteria to consider repair of a re-coarctation is not solely based on the trans-coarctation pressure gradient because significant collateral flow may substantially underestimate the severity of stenosis. These patients may present with persistent, refractory hypertension, headache, lower extremity claudication, or vascular findings on imaging such as aortic or cerebrovascular aneurysm.

Aneurysms may form at the site of previous coarctation repair or proximal to it and can develop with or without concurrent re-coarctation [25–27, 30–32]. Criteria to repair isolated aortic aneurysms following previous repair of coarctation should follow consensus guidelines of aortic aneurysm repair, however, in consultation with a congenital cardiac surgeon. Indications for repair of aortic aneurysms of the descending aorta include: (a) patients with chronic dissection, particularly if associated with a connective tissue disorder, and a descending thoracic aortic diameter exceeding 5.5 cm; (b) patients with degenerative or traumatic aneurysms of the descending thoracic aorta exceeding 5.5 cm, saccular aneurysms, or postoperative pseudoaneurysms; (c) patients with thoracoabdominal aneurysms, in whom endovascular stent graft options are limited and surgical morbidity is elevated, elective surgery is recommended if the aortic diameter exceeds 6.0 cm or less if a connective tissue disorder is present [33••, 34].

Techniques to Repair Re-coarctation

Both open surgical and endovascular approaches may be considered when faced with re-coarctation and are often age- and patient size-dependent. The decision of the specific technique to utilize depends on several factors, including the patient's overall health, performance status, and ability to tolerate an invasive procedure, anatomic considerations that may preclude endovascular treatment, and concomitant aortopathy or valvulopathy that may require simultaneous intervention.

Endovascular repair is the preferred treatment for re-coarctation, regardless of age [29]. This recommendation is based on data demonstrating a significant increase on mortality for reoperation than for native repair, 3% vs. < 1%, respectively [35, 36]. This number increases when there is associated pre-existing myocardial dysfunction. Both balloon angioplasty and stent grafting are options with good results in the short and long term to support its use [35–40]. Balloon angioplasty has a high rate of restenosis when used as the primary modality to repair a native coarctation; however, the trend toward restenosis is less when it is used to treat aortic re-coarctation, although the data is variable, ranging from 6 to 53%. Procedural morbidity and mortality are significantly less for percutaneous balloon angioplasty with aortic injury, including acute dissection and rupture, occurring in < 2% of cases [9]. Repeat angioplasty is therefore a reasonable approach to the patient with recurrent restenosis. In young children, with a significant amount of aortic growth yet to take place, balloon angioplasty is the preferred treatment modality for re-coarctation [9, 29, 41].

The current use of endovascular stenting and improvement in its technique and graft technology has made it a useful adjunct in the treatment of re-coarctation. Current data supports its use as a safe and effective method of repair [38, 42, 43]. Endovascular stent placement should only be performed as a primary treatment for re-coarctation when the patient is of sufficient size and age that an adult-sized stent graft can be safely placed in order to avoid patient-graft mismatch as additional growth takes place [9]. Successful stent placement is reported up to 99%, and significant reduction in trans-coarctation pressure gradients is consistent in the literature [2]. The general principles of stent graft deployment apply for treatment of coarctation as well. Abnormal vessel tortuosity and inappropriately small proximal and distal landing zones may preclude the use of a stent graft. In some cases, the use of advanced endoluminal revascularization procedures or additional open surgery, such as left carotid-subclavian bypass in the setting of insufficient proximal landing zones may be utilized.

Although safe, stent placement is associated with complications, similar to endovascular graft deployment for other disease processes. These include aortic wall injury, including acute dissection or rupture, endoleak, thromboembolic arterial disease, stent migration, graft infection, paraplegia secondary to spinal cord ischemia, and acute coronary syndrome [2, 37–39, 44, 45].

Surgical Repair

Although endovascular repair is the preferred treatment of re-coarctation, open surgical thoracic aortic repair may be the only option for intervention for complicated anatomy or failed endovascular treatment. This may include coarctation involving the transverse arch or other anatomic variants not amenable to stent grafting or complications of stent grafting including uncontrollable endoleak or graft infection.

In cases of re-coarctation isolated to the descending aorta, repair may be undertaken via left thoracotomy with aortic interposition tube graft implantation [36, 46–50]. Significant adhesive disease may be present, lending to increased complexity and length of the procedure, increasing the likelihood of intraoperative and postoperative morbidity. Further complexity may be added when undertaking repair of the transverse arch or larger segments of diseased aorta that may have concomitant aneurysmal disease or dissection. In these cases, thoracoabdominal aortic repair may be necessary, requiring intervention not only on the aorta, but the visceral arterial system as well [33••].

There are several approaches to repair the descending thoracic aorta. Clamping the aorta proximal and distal to the diseased segment, rapidly sewing the aortic prosthesis into place, known as the “clamp and sew” technique, may be utilized when a long clamp time, and therefore long ischemia time is not anticipated [51–53]. If ischemia is a concern, a method of perfusion of the lower extremities utilizing left atrial to femoral partial cardiopulmonary bypass may be employed. This involves cannulating the left inferior pulmonary vein to shunt oxygenated blood via bypass pump to arterial circulation distal to the clamped aorta [53, 54].

In the setting of complicated repair, hypothermic circulatory arrest can be used. In this case, cardiopulmonary bypass is instituted to maintain distal circulation, while the brain is cooled. The proximal dissection and anastomoses are performed first so that the graft can be cannulated and flow reinstituted to the cerebral vasculature as soon as possible. The distal anastomosis is then performed and the patient weaned from bypass [53].

Finally, in the setting of complex reoperation, with dense scar tissue or anatomic features making reconstruction of the descending thoracic aorta prohibitive, extra-anatomic bypass may be employed. Briefly, this involves rerouting blood flow from normal proximal aorta to normal distal aorta, bypassing the diseased re-coarctation. This may be accomplished via left subclavian to descending aorta bypass via left thoracotomy or less commonly from ascending to descending aorta via median sternotomy [55–57]. Seemingly, an advantage of extra-anatomic bypass through a median sternotomy allows for easy access to the heart to perform concomitant cardiac surgery should it be required.

Complications related to surgical repair of re-coarctation include hemorrhage (2%), thoracic duct injury (4%), nerve injury, including the phrenic and recurrent laryngeal (< 1%),

and spinal cord ischemia (3%), which may lead to paraplegia [58, 59]. Meticulous intraoperative hemostasis including careful attention to suture lines and ligating intraluminal branches of intercostal vessels is critical to avoid unnecessary morbidity. Identification of the left recurrent laryngeal nerve when dissecting around the aortic arch is necessary to avoid injury and subsequent vocal cord dysfunction. Similarly, when working in proximity to the lateral pericardium, the phrenic nerve should be identified and protected to avoid diaphragm paralysis. Although practice patterns may vary among surgical institutions, prophylactic placement of spinal drainage catheters may allow for early identification and prompt intervention at the onset of lower extremity weakness or paraplegia [60]. In addition, careful attention maintaining an elevated arterial blood pressure in the postoperative setting may help avoid the onset of spinal ischemia. Moreover, closed pleural drainage tubes are left in place postoperatively to evacuate hemothorax, effusion, and monitor for changes in quality of the effluent that may raise suspicion for chylothorax and a missed thoracic duct injury.

Conclusion

Aortic coarctation is a common congenital cardiovascular disease that is typically repaired in neonatal period or during infancy. Re-coarctation after primary repair is a late complication that necessitates intervention if the stenosis becomes hemodynamically significant. In the setting of re-coarctation, endovascular repair, specifically, balloon angioplasty is the preferred method of treatment of young children. It has been shown to be safe and effective. However, re-coarctation may be treated using endovascular techniques such as stent graft implantation in addition to balloon angioplasty for older adolescent or adult-sized patients. For cases not amenable to endovascular repair, open surgical repair is necessary. Recent advances in surgical technique, postoperative intensive care. and monitoring strategies have improved outcomes in the modern surgical era. A multidisciplinary approach, including pediatric and/or adult congenital cardiologists working with congenital cardiac surgeons and critical care specialists, is necessary to maximize optimal outcomes for this complicated patient group.

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

Human and Animal Rights and Informed Consent This article does not contain any studies with human or animal subjects performed by any of the authors.

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