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Acute Type A Aortic Dissection and Coarctation: Single-Stage Repair Using a Clamshell Incision and a Systematic Literature Review

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ABSTRACT

Background: Aortic coarctation (CoAo) may be discovered only when complicated by acute type A aortic dissection (ATAAD). We present a case with a one-stage repair of this pathologic association and review the relevant literature focusing on the surgical choices.

Case report: A 43-year-old man presented with acute thoracic pain. Computed tomography and echocardiography demonstrated CoAo, ATAAD type II, an ascending aorta aneurysm, and moderate regurgitation of a bicuspid aortic valve. Emergency surgery was performed. A clamshell incision, cardiopulmonary bypass with dual arterial cannulation (axillo-femoral), CoAo repair (by resection-interposition), and supracoronary aorta replacement were performed. Four years later, the patient was healthy and asymptomatic.

Review: Thirty surgical cases of ATAAD with CoAo repair after the dissection onset were included. Iatrogenic dissections and formerly repaired CoAo without surgical indication were excluded.

Results: The mean patient age was 27.8 ± 12 years; there was a male predominance (76.7%). The patients frequently presented with ascending aorta aneurysm (86.2%), bicuspid aortic valve (69%), and type II dissection (79.3%); dissection never extended below the CoAo. The one-stage treatment (15 patients; 55.5%) included 12 surgical repairs of CoAo (mostly by ascending-to-descending aorta extra-anatomic bypass; 58.3%) and three balloon angioplasties. In patients with uncorrected CoAo at the onset of cardiopulmonary bypass, double arterial perfusion was used in 55.5%.

Conclusions: One-stage repair (hybrid or surgical), double arterial perfusion, and extra-anatomic ascending-to-descending aorta bypass are the most common options for treating ATAAD-CoAo. The clamshell incision provides excellent access for an extended arch procedure and facilitates anatomic isthmus repair.

INTRODUCTION

Coarctation of the aorta (CoAo) is one of the risk factors for developing acute type A aortic dissection (ATAAD) [Erbel 2014]. This may be due to (persistent) hypertension or a consequence of the high prevalence of bicuspid aortic valve (BAV) in CoAo patients or even the effect of a diffuse arteriopathy possibly present in CoAo [Warnes 2003]; each of these mechanisms probably brings its pathogenic contribution. ATAAD associated with unrepaired (or recurrent) CoAo (ATAAD-CoAo) is rare. It represents a surgical challenge and also a matter of debate regarding the best operative options. Here, we describe a case of a successful one-stage repair of a neglected CoAo with bicuspid aortic valve (BAV) complicated by an ascending aorta aneurysm (AAA) and ATAAD. Based on a brief literature review, we further discuss the strategies used for the surgical management of ATAAD-CoAo.

CASE REPORT

A 43-year-old man without previously known heart disease was admitted for acute thoracic pain, polypnea, and moderate hypoxemia. The patient had a normal brachial artery pressure but weak femoral pulses. Contrast-enhanced computed tomography (CT) revealed an ATAAD (DeBakey type II) with massive false lumen thrombosis complicating a 6-cm AAA and CoAo. A diffuse mediastinal hematoma suggested a contained rupture of the false lumen. (Figure 1)

Transthoracic echocardiography (TTE) revealed moderate aortic regurgitation (AR) of the BAV, hypertrophy of the left ventricle, and a medium pericardial collection. The aortic annulus was normal (24 mm), and the aortic root diameter was 40 mm. Emergency surgery was performed. A bilateral anterior thoracotomy (in the fourth intercostal space) with transverse sternotomy (clamshell incision) was performed. Cardiopulmonary bypass (CPB) employed a bifurcated arterial line (right axillary and right femoral cannulation). Arterial

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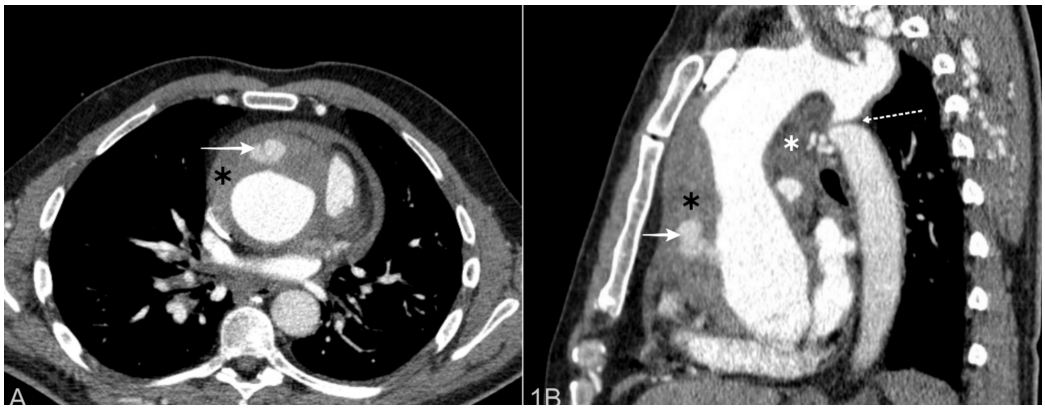


Figure 1. Preoperative computed contrast-enhanced tomography: Acute type A aortic dissection of ascending aorta and aortic coarctation: A. Axial section; B. Oblique sagittal section. White full arrow – circulated false lumen; Black asterisk – false lumen thrombosis; Dotted arrow – Coarctation; White asterisk – mediastinal hematoma

pressure was monitored simultaneously in the left radial and left femoral arteries. The patient became unstable during the aortic isthmus exposure. Thus, CoAo repair was performed on-pump through the resection and interposition of a size 24 collagen-coated Dacron graft. The resection included the frail distal aortic arch and the left subclavian artery (LSA) origin. The AAA was opened after aortic clamping below the origin of the innominate artery, and an intimal tear was found 2 cm above the right coronary ostium. A recent thrombus occupying most of the false lumen was removed. The heart was arrested using Custodiol® (Dr. Franz Köhler Chemie GmbH, Bensheim, Germany) cardioplegia (1800 ml at 4°C, in antegrade intraostial administration, supplemented retrogradely with half the initial dose, after 90 min of cross-clamping). The AR was due to incomplete fusion of the conjoint coronary cusps of the BAV; this small cleft was sutured. The aortic arch was certified intact during a short circulatory arrest (2 min) at 30°C. The sinotubular junction was reconstructed using a continuous horizontal suture of the dissected layers between Teflon strips. The supracoronary ascending aorta was replaced with a size 30 collagen-coated Dacron graft. LSA flow was restored using a 9-mm Dacron graft from the ascending aortic prosthesis. CPB and myocardial ischemia times were 311 and 113 min, respectively. CPB weaning was uneventful.

Postoperative echocardiography showed good biventricular function and mild AR. The patient was hemodynamically stable under low doses of noradrenaline. The patient was extubated on the second postoperative day (POD); a Klebsiella pneumoniae lung infection developed after that. It required three more days of mechanical ventilation and meropenem treatment. The patient was transferred to the ward on the 11th POD. Five days later, he had a minor stroke that resolved without sequelae. Staphylococcus epidermidis superficial wound infection (of the right thoracotomy) cleared after vacuum therapy and antibiotics. The patient was discharged on the 48th POD. The patient is now healthy and asymptomatic at four years postoperative. (Figure 2)

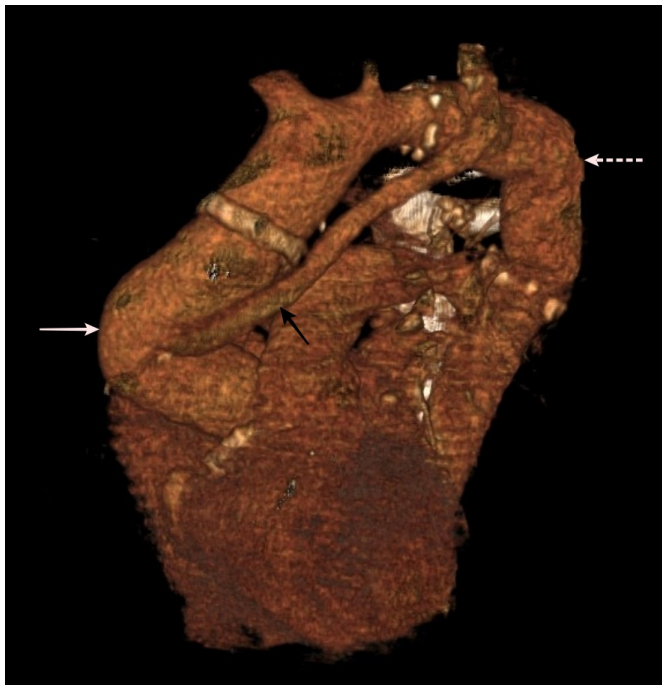
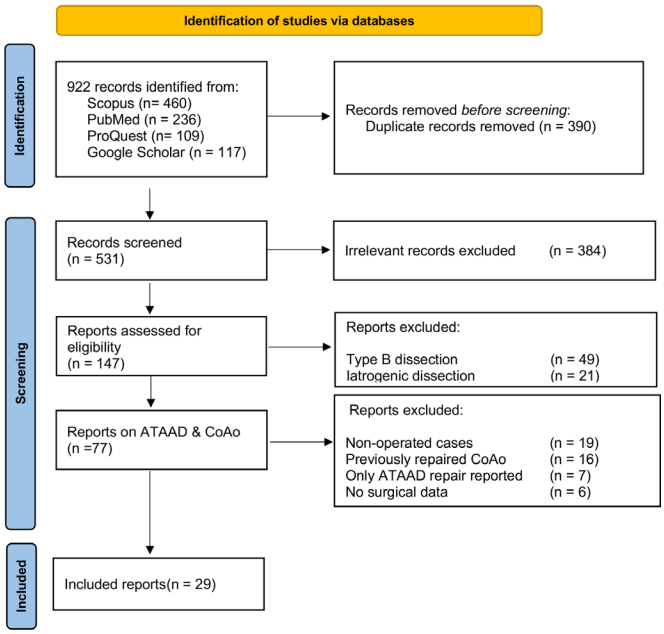


Figure 2. Three-dimensional rendering of postoperative computed tomography (10 months after repair): Ascending aorta graft – white arrow; Aortic arch to descending aorta graft – dotted white arrow; Graft from ascending aorta graft to left subclavian artery - black arrow

MATERIAL AND METHODS

The literature on the management of ATAAD-CoAo was reviewed. The PubMed, Scopus, ProQuest, and Google Scholar databases were searched. Cases of patients who underwent ATAAD and CoAo repair after the onset of dissection were included. The following exclusion criteria were applied: unreported repair of both diseases, CoAo repair without significant recurrent stenosis preceding ATAAD, and iatrogenic

Table 1. PRISMA flow diagram



dissection (mostly occurring after percutaneous CoAo treatment); reports (usually of genetics) lacking relevant details on the applied surgery. (Table 1) Including the presented case, 30 cases were gathered.

RESULTS

The mean patient age was 27.8 ± 12 years, ranging between 12 and 54 years. (Table 2) A net predominance of male patients was observed (76.7%). In the 29 cases with known data, ATAAD mainly developed in patients with preexisting AAA (86.2%) and BAV (69%). Dissection was confined to the ascending aorta (type II) in 23 out of 29 patients (79.3%) and never extended to the aorta below the CoAo. Hemopericardium was noted in 48.2% of cases. Two patients were diagnosed with Turner syndrome [Imamura 1995; Belov 2007].

The strategy of the treatment was simultaneous repair, one-stage treatment (OST), in 17 patients (56.7%) or sequential correction, dual-stage treatment (DST) of ATAAD-CoAo, in 13 patients (43.3%). The repair of ATAAD-CoAo was entirely surgical in 25 cases (83.3%); in the other five patients, a hybrid approach was applied: CoAo treatment by balloon angioplasty (BA) ± stenting. In three patients, BA was performed just before CPB for ATAAD repair [Imamura 1995; Heper 2005; Siromakha 2021], and this was considered hybrid OST; alternatively, BA performed following ATAAD repair [Bakker 2007; Dominguez 2022] was considered hybrid DST.

The arterial cannulation site could be retrieved in 20 out of the 25 cases having an uncorrected CoAo at the beginning

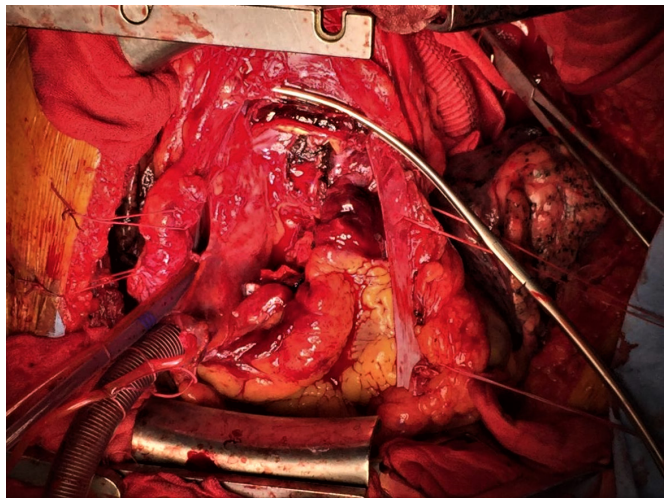


Figure 3. An intraoperative view shows the excellent access to the whole thoracic aorta offered by a clamshell incision (picture taken after the coarctation repair). The distal ascending aorta and the LSA are clamped. Subserous thrombus remains on the distal ascending aorta and pulmonary artery following the contained rupture of the false lumen.

of CPB. In 11 of them (55.5%), double arterial cannulation was used (usually in the axillary and femoral). If a single arterial cannula was used, it was more frequently inserted above the CoAo (axillary or aortic arch); reportedly, if cannulation was performed only in the femoral artery, the radial arterial pressure was maintained above 40 mmHg throughout the CPB [Hovaguimian 1990].

ATAAD repair included a conduit replacement of the ascending aorta in all patients. The aortic valve was replaced in 14 cases (46.7%). Use of a composite graft (Bentall or Chabrol procedures) was the most frequent option (in 11 patients); alternatively, a separate valve replacement (Wheat procedure) was performed (three cases). A valve-sparing root replacement was used twice. The decision to replace the aortic valve seemed unrelated to the presence of a BAV nor the type of repair (OST, DST). The valve replacement rate was 45% in the 20 BAV patients vs. 55.5% in the nine non-BAV patients; it was higher in OST than in DST (52.9% vs. 38.5%). Both differences were not significant. In four cases, complete replacement of the aortic arch was observed (13.3%).

The preferred technique for CoAo repair in the surgical OST group (14 patients) was an extra-anatomic ascending-to-descending aorta bypass (nine cases; 64.3%); more frequently, the distal anastomosis was performed on the descending thoracic aorta (five patients). Alternatively, the anatomic repair was performed by connecting the arch replacing graft to the aorta below the CoAo (in four patients) or by graft interposition (in the current case). In 12 of the surgical OST cases, the type of surgical incision could be retrieved; in half of them, the procedure was performed using only a sternotomy; an additional incision (laparotomy or left thoracotomy) was used in five, while a clamshell incision was used in the present case. Only one case of mortality was reported [Hovaguimian 1990].

Table 2. Review of surgical management of acute type A aortic dissection with coarctation of the aorta

Author, year	Age	Sex	BAV	AAA	ATAAD type	Repair type	ATAAD repair	CoAo repair	Art. Cann.	Incision
Lawson et al. 1979	21	m	-	-	I	DST	Conduit	Res-Int	F	Th; St
Bricker et al. 1980	20	m	?	?	?	DST	Conduit	?	?	St; Th
Sampath et al. 1982	37	m	+	+	II	DST	V. Cond.	Res-Int	F	Th; St
Hovaguimian et al. 1990	34	m	-	-	I	DST	V. Cond.	?	F	St; Th
Svensson 1994	30	m	+	+	II	OST	V. Cond.	EAAbAo	Arch-AbA	St+Lap
Imamura et al. 1995	19	f	+	+	II	OST-H	Wheat	BA	?	St
Tesler et al. 1996	54	m	+	-	II	DST	Conduit	Res-Int	Ax-F	St; Th
Paparella et al. 1999	14	m	-	+	II	OST	Conduit+Arch	Ax-F	St+Th	
Plunkett et al. 2000	29	f	-	+	I	DST	Conduit	Res-Int	F	St; Th
Buket et al. 2001	16	m	-	+	II	OST	V. Cond.	EAThAo	Ax-F	St
Bonvini et al. 2002	13	m	+	+	II	OST	V. Cond.	EAAbAo	?	St+Lap
Rampoldi et al. 2002	52	f	-	+	I	DST	V. Cond.	LSA-Ao	Ax-F	St; Th
Heper et al. 2005	20	m	+	+	II	OST-H	Wheat	BA	F	St
Belov et al. 2007	-	f	+	+	II	OST	Conduit post AVR	Redo EAThAo	Ax	St
Horai et al. 2007	36	m	+	+	II	OST	VSRR+arch	AscAo-F	St	
Bakker et al. 2007	16	f	+	+	II	DST-H	Conduit	BA	F	St
Biricinoglu et al. 2008	17	m	+	+	II	OST	V. Cond.	EAThAo	Ax-F	St
Cranfield et al. 2008	29	m	+	+	I	OST	Conduit	EAAbAo	Ax-F	St+Lap
Belov et al. 2011	26	m	+	+	II	OST	Wheat	EAThAo	Ax-F	St+Th
Boyacıoğlu et al. 2013	21	m	-	+	II	DST	Conduit	LSA-Ao	Ax	St; Th
Bezgin et al. 2013	16	m	+	+	II	DST	Conduit	?	Ax	St; Th
Okita et al. 2014	37	m	+	+	II	OST	VSRR+arch	Arch-F	St	
Sabzi et al. 2015	45	M	-	+	II	DST	V. Cond.	Res-Int	Arch	St; Th
Milovanović et al. 2017	12	m	+	+	II	DST	Conduit	Res-Int	Arch	St; Th
Khaja et al. 2017	25	m	+	+	I	OST	Conduit+arch	?	?	
Patel et al. 2021	20	m	-	+	II	OST	V. Cond.	EAThAo	?	St
Siromakha et al. 2021	28	f	+	-	II	OST-H	Conduit	BA	?	St
Dominguez et al. 2022	28	m	+	+	II	DST-H	V. Cond.	BA	Ax	St
Jiang et al. 2022	47	m	+	+	II	OST	V. Cond.	EAThAo	Ax-F	St
Iosifescu et al. 2022	43	m	+	+	II	OST	Conduit	ResInt	Ax-F	BilatTh

AbA, abdominal aorta; Arch - aortic arch; Art.Cann., arterial cannulation site; AscAo, ascending aorta; Ax, axillary; BA, balloon angioplasty; BilatTh, bilateral thoracotomy + transverse sternotomy; Conduit, supracoronary replacement of the ascending aorta; EAAbAo, extra-anatomic bypass to abdominal aorta; EAThAo, extra-anatomic bypass to descending thoracic aorta; F, femoral; H, Hybride procedure; LSA-Ao, left subclavian-aortic bypass; Lap, laparotomy; Res-Int, CoAo repair by resection interposition; St, median sternotomy; Th, thoracotomy; V. Cond., ascending aorta replacement with a valved conduit (Bentall or Chabrol procedure); VSSR, valve-sparing root repair (David or Yacoub procedure); Wheat, aortic valve replacement + supracoronary replacement of the ascending aorta

DISCUSSION

Campbell's classical study demonstrated the grim natural history of CoAo, with severely reduced life expectancy (to only 34 years for the one-year-old patients); 21% of the recorded deaths were due to aortic rupture [Campbell 1970]. The catastrophic aortic complication is explained by CoAo-induced hypertension, BAV-related arteriopathy (the prevalence of BAV in patients with CoAo is between 50 and 75%) [Erbel 2014] or by a Co-Ao-related arteriopathy [Warnes 2003]. The propensity of CoAo with BAV toward ascending aorta aneurysm, dissection, and rupture was demonstrated by Maude Abbott, who found BAV in over 50% of CoAo with ascending aorta rupture, but in only 16% of cases

with other thanatogenesis [Abbott 1928]. Reciprocally, CoAo was demonstrated to increase ascending aorta complications in BAV patients [Oliver 2009].

The literature review demonstrated that ATAAD-CoAo most frequently occurred in a previously developed AAA (> 85%) and was associated with BAV in over two-thirds of the cases. The dissection was usually of type II (~ 80%). In the reviewed cases, it never extended below the CoAo; however, this extension was once described in an autopsy case [Saunders 2009]. Expectedly, ATAAD-CoAo showed a high male prevalence (3.3:1) since CoAo, ATAAD, and BAV were all associated with the male sex (2.5:1, 1.9:1, and 3.7:1, respectively) [Michalski 2015; Huckaby 2022; Kong 2020].

ATAAD-CoAo is probably the most severe form of complex CoAo; due to the high risk of false lumen rupture, its emergent surgical treatment is indisputable. However, the best strategy and tactics for the treatment of this entity is not clearly established. The literature review demonstrated non-uniform practices, with various options of management. The first cases were sequentially performed through a DST approach [Lawson 1979; Bricker 1980; Sampath 1982; Hovaguimian 1990]. In two of them [Lawson 1979; Sampath 1982], CoAo was treated first, but this sequence was never repeated afterward, due to the risks implied by postponing ATAAD repair. In addition to avoiding lengthy surgery, the main argument for DST is the hypothetical prevention of coronary flow imbalance after CPB, due to the sudden drop in diastolic aortic pressure generated by the CoAo repair. This pathological sequence that usually responds to vasopressors was described long ago after CoAo repair with ongoing severe aortic regurgitation [Rufilanchas 1977] or stenosis [Pethig 1996]. However, there is insufficient evidence regarding its frequency or occurrence without an associated aortic valve pathology. After OST was first performed for ATAAD-CoAo [Svensson 1994], it quickly became the leading therapeutic choice. In addition to the advantage of a single procedure, supplementary arguments favor OST. Its use avoids increased afterload after CPB and may prevent postoperative bleeding, reducing the exposure of aortic sutures to hypertension [Svensson 1994; Büket 2001]. OST also precludes malperfusion below the CoAo in cases with underdeveloped collaterals or inadequate postoperative cardiac output [Cranfield 2008].

A hybrid procedure with percutaneous CoAo repair immediately followed by ATAAD correction is probably the most attractive variant of OST under ideal conditions. However, this strategy rarely is feasible on an emergency basis. Instead, it is usually intended to perform a surgical OST of ATAAD and CoAo, adapting one's experience and strategy for complex CoAo. Before opening the chest, the incision and cannulation site(s) must be chosen. When aortic arch replacement is not required, a median sternotomy followed by ATAAD repair and an intrapericardial extra-anatomic ascending-to-descending aorta bypass seems a straightforward option [Büket 2001; Patel 2021]. It showed good early and late results in complex CoAo cases, despite some reported bleeding complications [Said 2014], persistent effusions [Brink 2013], and concerns related to the use of a lengthy prosthetic conduit. However, this technique is unusual for many cardiac surgeons. Alternatively, one can perform another extra-anatomic bypass to the descending aorta superior to the left hilum [Birincioglu 2008; Belov 2011] or the abdominal aorta [Svensson 1994; Bonvini 2002; Cranfield 2008]. If aortic arch replacement is required (intimal tear in the arch or distal arch hypoplasia), the CoAo usually is repaired by the end-to-end connection of the aortic arch graft to the infrastenotic aorta [Paparella 1999; Horai 2007]. Most of these options would require an additional incision, such as a left thoracotomy or laparotomy.

In the present case, before the procedure, it was unclear whether the aortic arch surrounded by the hematoma was to be conserved or replaced. Therefore, easy access to the entire thoracic aorta was required. The clamshell incision fulfills this goal [Kouchoukos 2008]. It offered an excellent approach to the aortic root, arch, and descending thoracic aorta. (Figure 3) Thus, it facilitated anatomical isthmus repair and the partial resection of the distal arch. Based on this experience, we highly recommend it for the OST of ATAAD-CoAo, especially in cases requiring the extension of repair to the aortic arch.

Preservation of the aortic valve is one of the surgeon's goals in ATAAD repair, especially in young patients. In the reviewed group, the aortic valve could be preserved in 16 cases (53.3%) in patients with or without BAV. Resuspension of commissures, with sinotubular junction reconstruction, and cuspal repair or a valve sparing root replacement (VSRR) are the possible means to achieve this goal. In the present case, reductive reinforcement of the sinotubular junction and conjoint cuspal cleft suture successfully were used.

The reported experience with ATAAD-CoAo repair reveals two main options regarding arterial perfusion: single or double cannulation. Most patients already have well-developed collateral circulation owing to long-standing significant CoAo. Thus, adequate perfusion of the entire body is usually obtained, even if single cannulation is used. However, inadequate perfusion in the territory across the CoAo has been reported [Horai 2007]. Besides, in an ATAAD patient, one may not have time to assess the development of the collaterals. Their correct function may be questionable in the event of hemodynamic instability. Furthermore, the heat exchange efficiency in the underperfused territory has not yet been studied. To avoid these potential drawbacks, double arterial cannulation should be preferred since it provides good perfusion to the territories above and below the CoAo and, if required, facilitates the extension of surgery to the aortic arch and descending aorta [Czerny 2014; Kreuzer 2018].

Study limitations: There is substantial publication bias toward the overrepresentation of successfully treated cases, limiting the actual operative risk evaluation. We failed to obtain access to the full text of three reports [Bricker 1980; Imamura 1995; Belov 2007]. Some of the cases, specifically those published as image reports, included only a brief descriptive text; thus, the incidence of BAV in ATAAD-CoAo may be underestimated. Incidence of the Turner syndrome among patients with ATAAD-CoAo also may be underestimated, as some genetics reports on the subject were excluded due to lack of surgical relevance.

CONCLUSIONS

ATAAD complicating unrepaired CoAo is most often a type II dissection originating in a previously developed AAA; it most frequently occurs in BAV patients and (almost) never extends below the CoAo. The timely treatment of CoAo, early detection, and treatment of AAA are essential for ATAAD prevention. For the treatment of ATAAD-CoAo, OST (hybrid or surgical) and dual arterial cannulation are advantageous options. The use of a clamshell incision facilitates the anatomic OST of ATAAD-CoAo.

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