Kommerell's diverticulum and right-sided aortic arch: A cohort study and review of the literature

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We report four consecutive cases of Kommerell's aneurysm of an aberrant left subclavian artery in patients with a right-sided aortic arch and the results of a systematic review of the literature. In our cohort of patients, three had an aneurysm limited to the origin of the aberrant subclavian artery, causing dysphagia and cough, and one had an aneurysm involving also the distal arch and the entire descending thoracic aorta, causing compression of the right main-stem bronchus. A left subclavian-to-carotid transposition was performed in association with the intrathoracic procedure, and a right thoracotomy was used in all patients. One of the patients underwent surgery with deep hypothermia and circulatory arrest, and the others with the adjunct of a left-heart bypass. The repair was accomplished with an interposition graft in two patients and with endoaneurysmorrhaphy in the others. The postoperative course was complicated by respiratory failure and prolonged ventilation in one patient, and one patient died because of severe pulmonary emboli. The survivors are alive and well at a follow-up of 1 to 3 years. Only 32 cases of right-sided aortic arch with an aneurysm of the aberrant subclavian artery have been reported: 12 were associated with aortic dissection, and 2 presented with rupture. Surgical repair was accomplished in 29 patients. A number of operative strategies were described: right thoracotomy, bilateral thoracotomy, left thoracotomy with sternotomy, sternotomy with right thoracotomy, and left thoracotomy. In only 12 cases was the subclavian artery reconstructed. We believe that a right thoracotomy provides good exposure and avoids the morbidity associated with bilateral thoracotomy or sternotomy and thoracotomy. We feel that a left subclavian-to-carotid transposition completed before the thoracic approach revascularizes the subclavian distribution without increasing the complexity of the intrathoracic procedure. (J Vasc Surg 2004;39:131-9.)

A right-sided aortic arch is an uncommon congenital defect of the aorta, and it is rare in the setting of an otherwise normal heart. Fioratti and Aglietti[1] described a right-sided aortic arch more than two centuries ago. Several classifications of these anomalies have been proposed on the basis of the arrangement of the arch vessels, relationships with the esophagus, or the presence of congenital heart anomalies.[2-7]

In the adult population a right-sided aortic arch is often asymptomatic unless aneurysmal disease develops. This usually occurs at the level of the take-off of an aberrant left subclavian artery and is known as a Kommerell's aneurysm. In spite of its rarity, this condition is clinically relevant because of the mortality associated with rupture, the morbidity caused by compression of mediastinal structures, and the complexity of surgery. The purpose of this study is to review the literature and report our experience.

MATERIAL AND METHODS

Review of the literature. Reports of thoracic aortic aneurysms associated with a right-sided aortic arch were identified through a Medline database search from 1966 to July 2002 using Ovid software (Ovid Technologies, Inc., New York, N.Y.) and the search strategy "Aneurysm and thoracic aorta (Exploded MeSH headings)" and "right-sided arch or Kommerell or arch or aberrant left subclavian text word" or "thoracic aorta (abstract word)." Reference lists of all relevant articles and reference lists of review articles were also examined. Four personal cases were included and the surgical management described.

Cohort study. Data regarding demographics, symptoms, imaging results, and outcome from four patients operated on by the senior author (C.S.C.) from June 1999 to August 2002 at a tertiary vascular center were collected from hospital charts, physician's office files, and outpatient records. Long-term follow up was done contacting all survivors by telephone.

The case of one of the four patients with a right-sided arch and a large Kommerell's aneurysm associated with an aneurysm of the entire descending thoracic aorta has been previously reported.[8]

RESULTS

Case reports

Case 1. A 21-year-old man was referred for assessment of claudication of the left arm and dysphagia associated with a vascular anomaly of the aortic arch. At birth he was diagnosed with congenital laryngeal stridor and treated conservatively. At age 5 years he was investigated for recurrent respiratory problems. A chest radiograph and a barium swallow indicated the presence of an arch anomaly with posterior compression of the esophagus, and an arch angiogram demonstrated a right-sided aortic arch and aberrant left subclavian artery (Fig 1, *A*). Symptoms were relieved after surgery, which consisted of division of the *ligamentum arteriosus* and of the aberrant left subclavian artery through a left thoracot-

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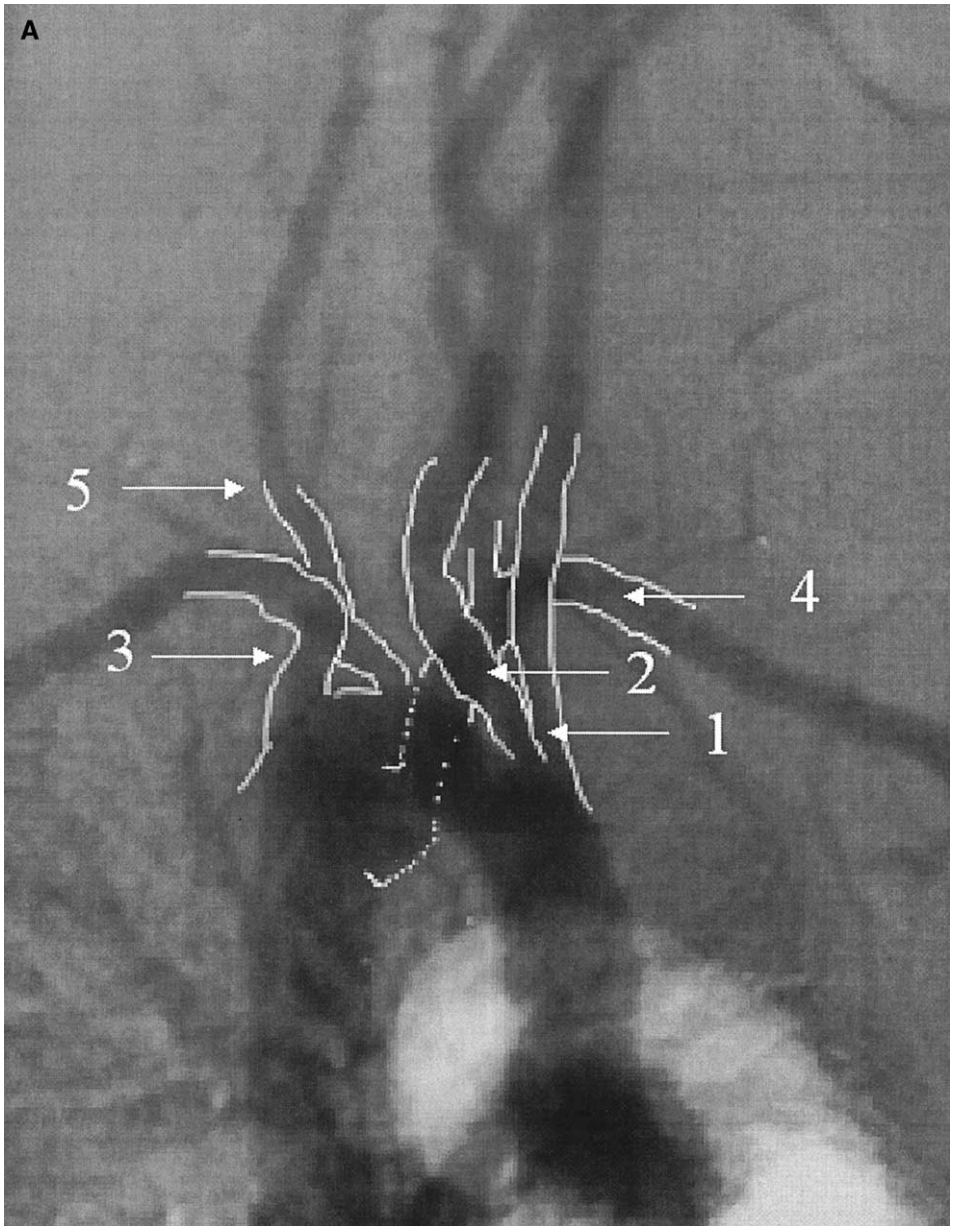

Fig 1. Case 1. Angiography demonstrates the right-sided aortic arch and aberrant left subclavian artery with a Kommerell's diverticulum at age 5 years (**A**) and the occluded subclavian artery and the Kommerell's aneurysm at age 21 years (**B**). *1*, Left common carotid artery; *2*, right common carotid artery; *3*, right subclavian artery; *4*, left subclavian artery; *5*, right vertebral artery originating from the aortic arch; *6*, Kommerell's aneurysm.

omy. At age 21 years, the patient presented with increasing arm claudication, left shoulder pain, and odynophagia. His blood pressure was 125/58 mm Hg in the right arm and 80 mm Hg in the left arm. The remainder of the physical examination was unremarkable. A Doppler ultrasound scan showed a retrograde flow in the left vertebral artery. Magnetic resonance angiography and an arch angiogram confirmed the presence of a right-sided aortic arch, an occluded aberrant left subclavian artery, and the presence of a Kommerell's aneurysm measuring 3 cm (Fig 1, *B*).

Surgery was performed in two stages. In the first, a right posterolateral thoracotomy was performed through the fourth intercostal space with removal of the fifth rib. A left atrial to thoracic aorta partial bypass was used for distal perfusion. An aortic clamp was placed between the right carotid and the subclavian artery, and a second clamp was placed in the descending thoracic aorta. A longitudinal aortotomy was used to enter the lumen of the aorta and to expose the neck of the Kommerell's aneurysm. An endoaneurysmorrhaphy was performed

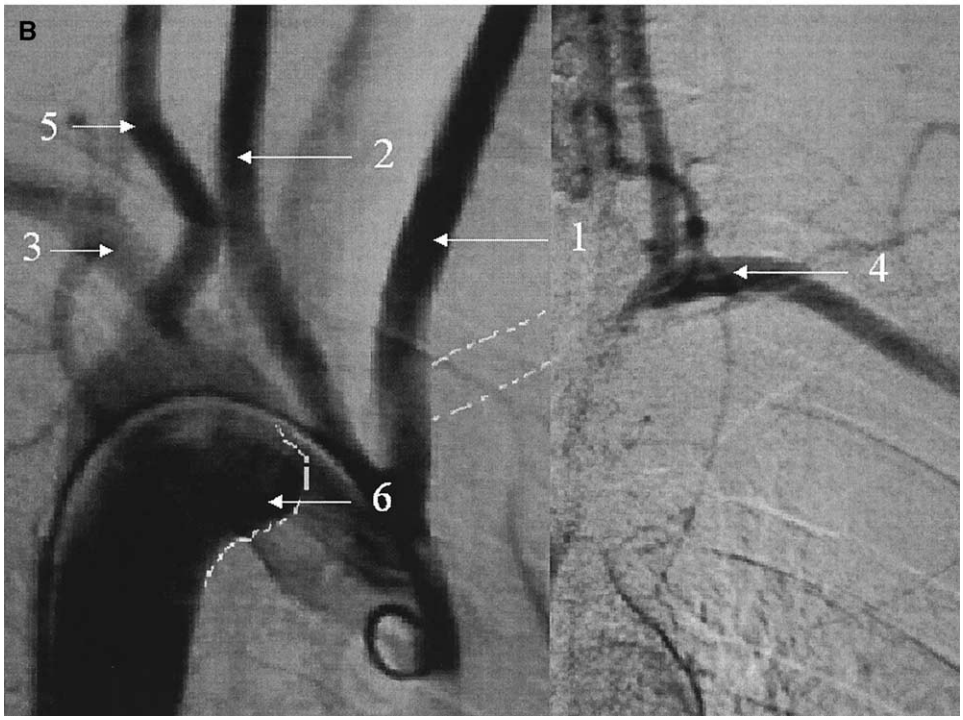

Fig 1. (Continued.)

by using two continuous sutures and two pledgeted stitches of 3-0 prolene. The aortotomy was closed with 3-0 prolene in a running fashion. The left atrium and the aorta were decannulated, and the native circulation was re-established. In a second stage, a left subclavian-to-carotid transposition was done by using the technique described elsewhere.[9] This patient remained symptom-free at the 14 month follow-up.

Case 2. A 53-year-old female was referred because of symptoms of dysphagia, epigastric discomfort, and palpitations. In the past she received treatment for *Helicobacter pylori* gastritis and underwent cardiac investigations including a stress test and echocardiography, which were reported as normal. She presented to the emergency department complaining of retrosternal epigastric pain radiating through the back and associated with tingling in the left hand. Blood pressure was 125/65 mm Hg in the right arm and 110/65 in the left arm. A chest radiograph showed an aortic knuckle in the right mediastinal profile in addition to the one on the left and a smooth indentation in the trachea. Cardiac catheterization disclosed a normal coronary circulation and a right-sided aortic arch with an aberrant left subclavian artery (Fig 2, *A*). Computerized axial tomography confirmed the presence of an aneurysm of the origin of the aberrant left subclavian artery measuring 3 cm in diameter (Fig 2, *B*), and a barium swallow demonstrated the esophageal compression.

Surgery was performed in two stages during the same operative procedure. With the patient in the supine position, a left subclavian-to-carotid transposition was done first. In the second stage, the patient was positioned in the left lateral decubitus with the chest at a 45° angle. Thoracotomy, partial left-heart bypass, and endoaneurysmorrhaphy were done, as in the previous case. The patient remained symptom-free at a 1-year follow-up.

Case 3. A 67-year-old man presented with dry cough and occasional dysphagia. His past medical history was significant for angina treated with coronary angioplasty and stenting of the left anterior descending and right coronary artery. His blood pressure was asymmetrical, with the left brachial pressure 15 mm Hg lower than the right, and his heart rate was regular. Angiography disclosed the same arrangement of the aortic arch branches described in the other patients. Computerized axial tomography confirmed the presence of a large Kommerell's aneurysm, measuring 6.4 cm, at the isthmus of the aorta from which the left subclavian artery originates (Fig 3).

At the time of surgery, a left subclavian-to-carotid transposition was done first with the patient in the supine position. The patient was then turned on the left lateral decubitus, and a thoracotomy performed with removal of the fifth rib. A left atrial-to-femoral bypass was initiated. The repair of the distal arch aneurysm and Kommerell's aneurysm was done with interposition of a 26-mm Dacron graft. The patient was extubated the first postoperative day, transferred to the step-down unit on the fifth day, and subsequently transferred to the floor. On the sixteenth postoperative day, he became hypotensive and dyspneic and suffered a cardiac arrest. An autopsy disclosed the presence of massive pulmonary emboli.

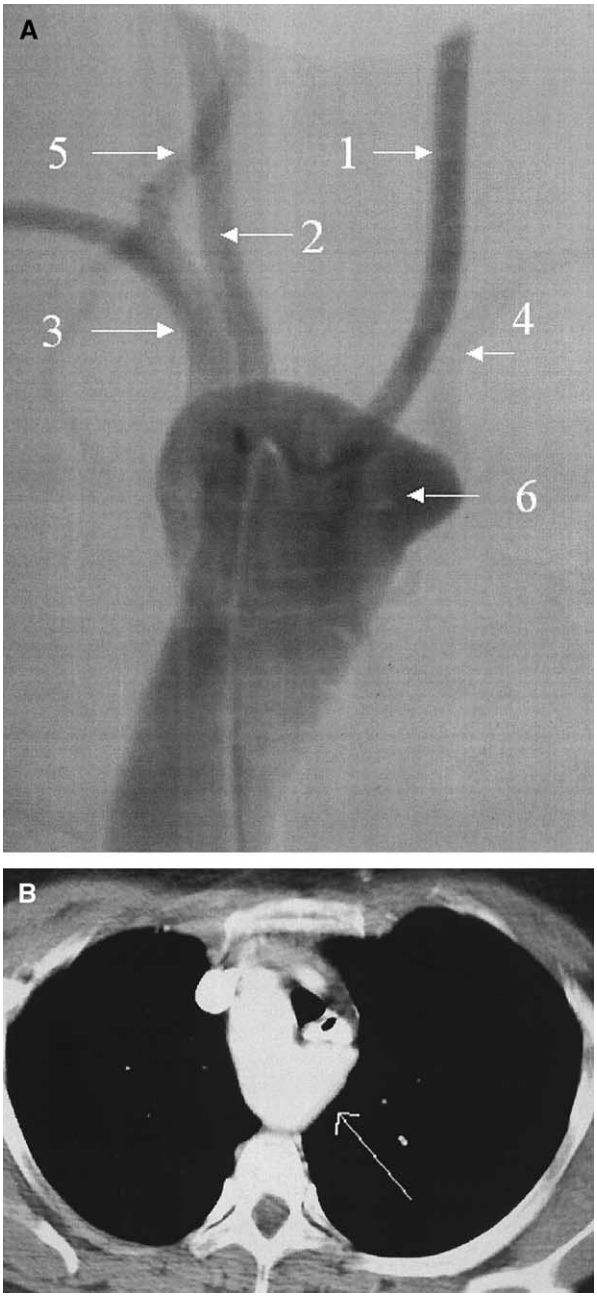

Fig 2. Case 2. Angiography (**A**) and computerized tomography (**B**) confirm the presence of the Kommerell's aneurysm.

Review of the literature

Thirty-two adult cases of thoracic aortic aneurysms with right-sided aortic arch have been reported (Table). Of these, 12 presented with dysphagia,[10-16] 13 with dissection,[15,17-27] 8 with respiratory symptoms,[9-12,16,28] and two with rupture.[23,29] Their ages ranged from 24 to 77 years, with a median of 51 years. Fourteen patients were male, 10 were female, and in 8 the sex was not specified.

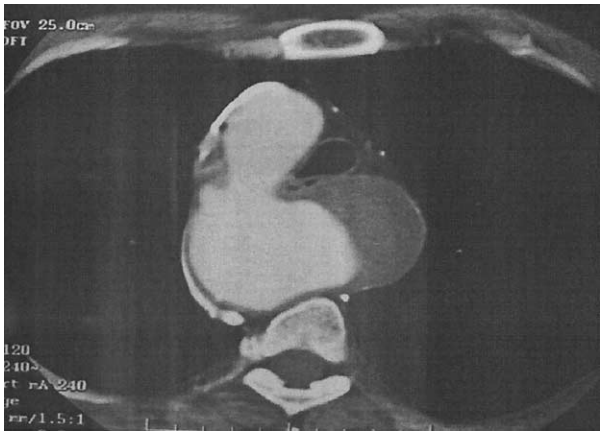

Fig 3. Computerized axial tomography demonstrated a 6.4-cm aneurysm.

The median size of the Kommerell's aneurysm was 6 cm (range, 2-12cm).

Of the 32 cases reported, 29 (90%) were treated surgically. Three did not undergo surgery, either because they died before surgery or because they were deemed to be at too high a risk. All three died of rupture.[20,29-31] In 27 patients, surgery was described in detail. A number of operative strategies were used and are summarized as follows: right thoracotomy in 16 patients,[10,11,13-15,17,19,23,24,26,27,32] median sternotomy with thoracotomy (either right or left) in 4 patients,[14,18,22,32] left thoracotomy in 2 patients,[16] median sternotomy in 2 patients,[28,33] bilateral thoracotomy in 1 patient,[21] bilateral thoracotomy and transverse sternotomy in 1 patient,[12] endovascular stent-graft repair in 1 patient,[34] and extra-anatomical bypass in 1 patient.[18] The outcome of surgery was not reported in 4 patients,[11,16] and in the remaining 25, the mortality was 12%. One patient developed paraplegia after surgery (4%).[30]

The aberrant subclavian artery was ligated or occluded in 13 cases[12-15,17,18,21,24,27,28,33,34] and was reconstructed with an aortosubclavian bypass or with an inclusion technique in 7[16,19,32,35] and with a subclavian-carotid transposition in 5.[8,11] In two patients the description of the management of the aberrant subclavian artery was unclear.[26,30]

A partial cardiopulmonary bypass was used in 8 patients,[10,17,19,22,24,27,30,32] a temporary brachiofemoral bypass in 1,[23] deep hypothermia and circulatory arrest in 6,[8,13,15,26,32] a clamp-and-go technique in 6,[12,14,16,21,35] and endovascular repair in one.[34] In seven patients the operative description did not specify if circulatory support was used.[11,18,28,33]

DISCUSSION

A right-sided aortic arch is a rare congenital defect of the aorta. It is present in 0.05% to 0.1% of radiology series[7] and in 0.04%-0.1% of autopsy series.[36] In autopsy series, 50% of right-sided arched are associated with an aberrant left subclavian.[36]

Kommerell[37] in 1936 reported an aberrant right subclavian artery originating from the descending thoracic aorta of a left-sided arch and associated with persistence of a remnant of the right dorsal aorta. The latter appeared as a diverticulum from which the aberrant right subclavian artery originated. This is known as the diverticulum of Kommerell.[37] An aberrant right subclavian artery with a left-sided aortic arch is the most common of the anomalies involving the subclavian artery.

In the embryo, six pairs of aortic arches develop at different stages of the organogenesis. Details of the embryology of this region have been described elsewhere.[8] The fourth primitive left aortic arch forms the adult aortic arch. The right fourth generally disappears, producing the normal course of the aorta arching to the left and descending to the left of the spine. If the left fourth arch disappears and the right persists, a right aortic arch develops. If both arches persist, they form a double arch or a vascular ring encircling the trachea and esophagus.

A number of classifications of these anomalies have been described, based on the arrangement of the aortic arch vessels, the relationships with the esophagus, or the presence of congenital heart anomalies.[5-7] Edwards[38] described three main types of right-sided aortic arch: type I, with mirror-image branching of the major arteries (Fig 4, *A*); type II, with an aberrant subclavian artery (Fig 4, *B*); and type III, with isolation of the subclavian artery (where the subclavian artery is connected to the pulmonary artery through the *ductus arteriosus*; Fig 4, *C*). In each of these three major types of right aortic arch, the ductus arteriosus may be on the left, on the right, or bilateral. Type I represents 59% of all right aortic arches, type II 39.5%, and type III 0.8%.[6]

Congenital heart anomalies (including Tetralogy of Fallot, pulmonary stenosis with ventricular septal defects, tricuspid atresia, and *truncus arteriosus*) are present in 75% to 85% of type I and type III and in 5% to 10% of type II right-sided arches.

The association of a right aortic arch with Tetralogy of Fallot (Corvisart's disease) is well described. It ranges from 13% to 34% in different series.[36] Tetralogy of Fallot is the most common heart anomaly in type I (93%), and it is present in 6.5% of type II and 0.5% of type III right-sided arches.[6]

The etiology of right-sided arch anomalies is unknown. A deletion in chromosome 22q11 is known to be associated with a 24% incidence of isolated anomalies of laterality of branching of the aortic arch.[39]

A right-sided aortic arch may be asymptomatic. In infancy, symptoms are related to congenital heart anomalies or to compression of mediastinal structures such as the trachea or the esophagus. In adulthood, symptoms are more often the result of early atherosclerotic changes of the anomalous vessels, dissection, or aneurysmal dilatation with compression of surrounding structures causing dysphagia (*dysphagia lusoria*—dysphagia by a trick of nature), dyspnea, stridor, wheezing, cough, choking spells, recurrent pneumonia, obstructive emphysema, or chest pain.[5,13,40]

An aberrant subclavian artery may be located behind the esophagus (80%), between the esophagus and the trachea (15%), or in front of the trachea (5%)[41] and can cause symptoms even in the absence of an aneurysm. The literature on right or left aberrant subclavian arteries indicates that division of the artery alone is often associated with relief of symptoms. In the pediatric age group, Roberts[42] reported 20 patients with right or left aberrant subclavian arteries. The treatment included ligation of the ductus arteriosus alone in 17 patients and of the subclavian artery in seven. Although no symptoms of subclavian steal or arm claudication were reported over a follow-up of 16 years, the authors acknowledged that revascularization of this artery may prevent later symptoms from occurring. This is exemplified by our first patient, in whom a debilitating left arm claudication appeared 16 years after his pediatric surgery. In adults, Hallman and Cooley[43] advocated re-establishment of flow to the upper extremity because simple ligation was associated with ischemic symptoms. Bailey[44] described re-establishment of flow by reimplanting the aberrant vessel to the ascending aorta. Orvaldt[45] described the subclavian carotid artery transposition to revascularize the upper extremity, and Campbell[46] used an inclusion technique, placing a Dacron graft within an aneurysmal aberrant subclavian artery and, therefore, maintaining flow in a retroesophageal position.

In a review of the literature of Kommerell's aneurysms associated with right or left aberrant subclavian arteries, Austin reported that 19% of affected patients presented with rupture, and all of them died.[47] In our review of aneurysm associated with right-sided arch, 6% of affected patients presented with rupture and 53% with either rupture or dissection. The size at which these aneurysms will rupture cannot be predicted because of the relative rarity of the condition and the limited information available. Of the 32 cases collected in the review of the literature by Austin, the size was reported in 20 (mean diameter = 7 ± 3 cm). Six of these were associated with rupture, and the size was described in four (range, 4-10 cm in diameter). In our review, the size was reported in 12 of 32 patients (mean diameter, 5.7 ± 2.2 cm; range, 2-12 cm). The average size at which these aneurysms ruptured was 5.8 ± 2 cm (2-10 cm).

Austin[47] reported an operative mortality for elective treatment of Kommerell's aneurysm of 16.6%. In our review, the mortality for elective aneurysm with right-sided arch was 8.3%, and for aneurysms associated with dissection 18%.

Surgical management of large aneurysms is more complex than for smaller ones. Therefore, in good-risk patients we suggest aggressive treatment for aneurysms of 3 cm or greater in diameter.

A left subclavian-to-carotid transposition completed before the thoracic approach revascularizes the subclavian distribution without increasing the complexity of the intrathoracic procedure. We believe that reconstruction of the left subclavian artery should be done in all patients; in young individuals this may prevent arm claudication, and

Characteristics of studies included in the systematic review

Authors, Year	*n*	*Size of aneurysm (cm)*	*Age (y)*	*Gender*	*Symptoms*	*Surgical approach*
Dikman SH, 1974[29]	1	8	77	Female	Rupture	—
Roan P, 1979[17]	1	—	48	Female	Dissection	Right thoracotomy
Bodine JA, 1982[20]	1	—	61	Female	Dissection	—
Floten HS, 1984[21]	1	—	64	Male	Dissection	Bilateral thoracotomy
Ohteki H, 1987[22]	1	—	62	Male	Dissection	Left thoracotomy and median sternotomy
Sugita T, 1990[23]	1	—	64	Male	Dissection with rupture	Right thoracotomy
Macda M, 1990[24]	1	—	49	Male	Dissection	Right thoracotomy
Fukushima K, 1991[30]	1		53	Male	Dissection	Medical treatment
	1		47	Male	Dissection	Unclear
Takano H, 1993[33]	1	4.3	24	Female	Asymptomatic	Median sternotomy
Caus T, 1994[10]	1	12	44	Man	Dysphagia, cough	Right thoracotomy
Kieffer E, 1994[11]	1	Unknown	—	—	Respiratory symptoms and dysphagia	Right thoracotomy and subclavian carotid transposition
	1	Unknown				
	1	Unknown				
	1	Unknown				
Kanoh M, 1995[18]	1	—	69	Female	Dissection	Median sternotomy and left thoracotomy
Baev B, 1995[12]	1	—	33	Female	Dysphagia, cough and dysphonia	Bilateral thoracotomy and transverse sternotomy
Patiniotis TC, 1995[13]	1	4.5	60	Female	Dysphagia	Right thoracotomy
Osako M, 1996[19]	1	7	70	Female	Dissection, dysphagia	Right thoracotomy
Imagawa H, 1997[28]	1	5.8	49	Female	Dyspnea	Median sternotomy
Donatelli F, 1997[14]	1	3	45	Female	Dysphagia	Right thoracotomy
Moizumi Y, 1999[15]	1	5	39	Male	Dissection, dysphagia	Median sternotomy and right thoracotomy
Muraoka M, 1999[35]	1	—	41	Male	Vertebro-basilar insufficiency	Unclear
Minato N, 1999[27]	1	2	50	Male	Dissection	Right thoracotomy
Cinà CS, 2000[8]	1	9.3	68	Male	Dysphagia	Right thoracotomy
Okada K, 2001[34]	1	6	76	Male	Chest pain	—
Tsukube T, 2001[32]	1	—	—	—	—	Right thoracotomy
	1					Median sternotomy and thoracotomy
	1					Right thoracotomy
Tsunemi K, 2001[26]	1	—	51	Male	Dissection	Right thoracotomy
Mossad E, 2002[16]	1	2	50	Male	Dysphagia, dyspnea	Left thoracotomy
	1	Unknown	28	Unknown	Unknown	Left thoracotomy

older patients may avoid the possibility of a subclavian steal syndrome. Subclavian-to-carotid transposition before intrathoracic repair is a practical way to achieve this without increasing the overall morbidity of the procedure. Our

(Continued)

Type of repair	*Type of extracorporeal circulation used*	*Outcome*	*Follow up (mo)*
—	—	Died before surgery	—
Interposition graft; ligation left subclavian artery	Partial cardiopulmonary bypass	Died	—
—	—	Died before surgery	—
Extra-anatomical bypass; ligation left subclavian artery	No	Alive	16
Surgery aborted	Partial cardiopulmonary bypass	Alive	5
Aneurysmorrhaphy and repair of dissection	Temporary brachiofemoral bypass	Alive	Unknown
Interposition graft; ligation left subclavian artery	Partial cardiopulmonary bypass	Alive	Unknown
		Died	Unknown
Unclear	Partial cardiopulmonary bypass	Alive with paraplegi	Unknown
Aneurysmorrhaphy; ligation left subclavian artery	Unclear	Alive	Unknown
Interposition graft; ligation left subclavian artery	Partial cardiopulmonary bypass	Alive	Unknown
Unclear	Unclear	Unclear	Unknown
		Unclear	
		Unclear	
		Died	
Extra-anatomical bypass	Unclear	Died	Unknown
Division of ligamentum arteriosum and aberrant left subclavian artery without repair of the aneurysm	No	Alive	Unknown
Interposition graft; ligation left subclavian artery	Deep hypothermic circulatory arrest	Alive	Unknown
Interposition graft and reimplantation of subclavian artery from the right chest	Partial cardiopulmonary bypass	Alive	Unknown
Interposition graft; ligation left subclavian artery	Unclear	Alive	Uknown
Excision of the aneurysm from the aorta and closure of the aortic defect, no reimplantation of left subclavian artery	No	Alive	Unknown
Interposition graft; ligation left subclavian artery	Deep hypothermic circulatory arrest	Alive	Unknown
Aortosubclavian bypass without repair of the aneurysm	No	Alive	Unknown
Interposition graft; ligation left subclavian artery	Partial cardiopulmonary bypass	Alive	Unknown
Interposition graft and subclavian carotid transposition	Deep hypothermic circulatory arrest	Alive	6
Endovascular repair	No	Alive	Unknown
Interposition and reimplantation of subclavian artery from the right chest	Deep hypothermic circulatory arrest	Alive	Unknown
Interposition graft and reimplantation of subclavian artery from the right chest	Deep hypothermic circulatory arrest	Alive	Unknown
Interposition graft and reimplantation of subclavian artery from the right chest	Partial cardiopulmonary bypass	Alive	Unknown
Aneurysmorrhaphy and repair of dissection	Deep hypothermic circulatory arrest	Alive	Unknown
Excision of the aneurysm from the aorta and closure of the aortic defect. Reimplantation of left subclavian artery	No	Alive	Unknown
Excision of the aneurysm from the aorta and closure of the aortic defect. Reimplantation of left subclavian artery	No	Uknown	Unknown

experience and the results of a review of the literature indicate that subclavian carotid transposition is an ideal technique for reconstruction of this artery, with excellent long-term patency.[8]

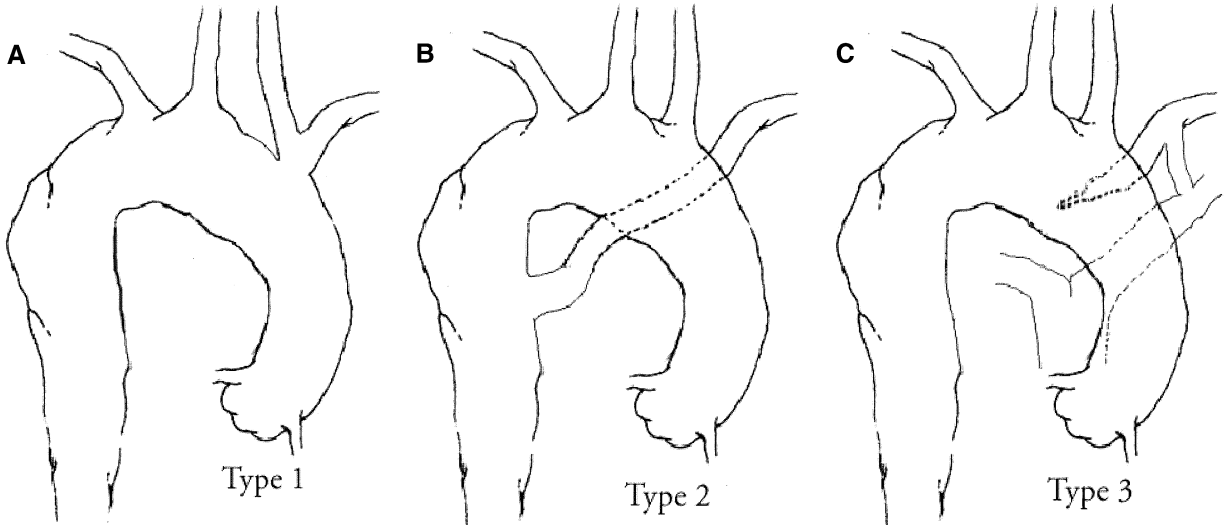

Fig 4. Classification of right-sided arch anomalies. **A**, Type I, with mirror image arch branches. **B**, Type II, with aberrant left subclavian artery. **C**, Type III, with isolated left subclavian artery.

A right thoracotomy provides good exposure of the ascending, transverse arch and descending thoracic aorta to allow reconstruction of these arteries and repair of a Kommerell's diverticulum, if present. In addition, it avoids the morbidity associated with bilateral thoracotomy or sternotomy and thoracotomy.

The management of the aneurysm will depend on the anatomy, size, and presence of a concomitant thoracic aortic aneurysm: Endoaneurysmorraphy is ideal for small Kommerell's aneurysms with a normal descending thoracic aorta, whereas an interposition graft is usually necessary for large Kommerell's aneurysms or Kommerell's aneurysms associated with an aneurysm of the descending thoracic aorta.

We advocate the use of distal circulatory support in all patients to reduce the incidence of paraplegia. The small series reported in the literature might underestimate the incidence of this complication, which was described in only 4% of patients. A left atriofemoral bypass without an oxygenator is a simple technique to provide distal circulatory support: It is adequate in most patients and has the advantage of not requiring full heparinization. In addition, the procedure may be easily converted to total cardiopulmonary bypass if the circuit has been appropriately predisposed for the conversion. Depending on the exact anatomy, circulatory arrest with deep hypothermia (as used in one of our patients) may be necessary, particularly for large aneurysms. However, when we anticipate that circulatory arrest may become necessary, as with large Kommerell's aneurysms, we prefer to insert a separate axillary graft in the left subclavian artery, using an infraclavicular incision. This graft is tunneled subcutaneously to the right hemithorax, retrieved after right thoracotomy into the surgical field, and may be used to perfuse the cerebral circulation during the period of arrest. This adjunct allows repair of the aortic arch with selective antegrade cerebral perfusion.

CONCLUSION

Kommerell's aneurysms associated with a right-sided aortic arch may be repaired through a right thoracotomy and atriofemoral bypass. A left subclavian-to-carotid transposition before the thoracic repair allows a practical approach to reconstruction of the subclavian artery. Particularly for large aneurysms, careful preoperative imaging and consideration of the individual anatomy in surgical planning are essential to anticipate the need for circulatory arrest and to achieve a successful outcome.

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