Case Reports

Anomalous Right Subclavian Artery Aneurysms

Report of 3 Cases, with a Review of the Literature

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During the past 2 years, 3 anomalous right subclavian artery aneurysms have been encountered at the St. Louis Heart Institute. The 1st patient, a 72-year-old woman, was found to have an asymptomatic 5-cm-diameter anomalous right subclavian artery aneurysm after surgery for suspected rupture of an abdominal aortic aneurysm. Resection was not attempted because of her poor cardiopulmonary and renal condition. One year later, the patient remains alive with marked cardiopulmonary limitations. The 2nd patient, a 77-year-old man, experienced dysphagia and severe weight loss because of a 14-cm-diameter aneurysm. Three days after undergoing surgical repair, he required reoperation for graft occlusion with right upper-extremity ischemia. Six months after hospital discharge, he died of pulmonary insufficiency and metastatic colon cancer. The 3rd patient, a 73-year-old woman, required emergency surgical intervention because of acute rupture and hypovolemic shock. Thirteen days later, she died of aspiration, asphyxia, and cardiac arrest.

On the basis of our experience and a review of the literature, we conclude that symptomatic anomalous right subclavian artery aneurysms are rare, and that surgical intervention entails a relatively high morbidity and mortality rate. If long-term survival is anticipated, associated medical illnesses should be considered before surgery is undertaken. ***(Texas Heart Institute Journal 1991;18:209-18)***

Anomalous right subclavian artery is the most common congenital anomaly of the aortic arch, occurring in 0.5% of the population (approximately 1 in every 200 persons).[1] Aneurysms of the anomalous right subclavian artery have been reported since the 17th 00s. Although most such aneurysms are asymptomatic, some cause dysphagia. This symptom was referred to as "dysphagia lusoria" by Hunauld[2] in 1735 and by Bayford[3] in 1794. It was 1st treated surgically by Gross[4] in 1946. Surprisingly, reviews by Austin[5] and Jauch[6] revealed a total of only 37 aneurysms of an anomalous right subclavian artery to have been reported in the world literature by 1988. Our most recent review revealed 49 aneurysms of the anomalous right subclavian artery, of which 33 were treated surgically.

An aneurysm can arise de novo from the anomalous right subclavian artery, or it can represent a developmental remnant of the right ascending aortic arch, from which the anomalous right subclavian artery arises (Fig. 1). This remnant, known as a "Kommerell diverticulum," looks aneurysmal but has the gross and histologic characteristics of the aorta.[7] Although an intact Kommerell diverticulum seldom ruptures, with time it may become aneurysmal, or it may thrombose or perforate, necessitating surgical intervention.[8-11]

We report our experience with 3 patients who had anomalous right subclavian artery aneurysms, which we encountered over a 2-year period.

Key words: *Aneurysm, anomalous right subclavian artery; diverticulum, Kommerell; carotid-subclavian artery bypass; congenital anomalies*

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Case Reports

Case 1

A 72-year-old woman was admitted to our institution for uncontrollable hypertension (blood pressure, 210/110 mmHg) and diffuse abdominal and lower-back pain. For 5 months, her physician had been observing her closely because she had an abdominal aortic aneurysm. Physical examination revealed a thin, frail

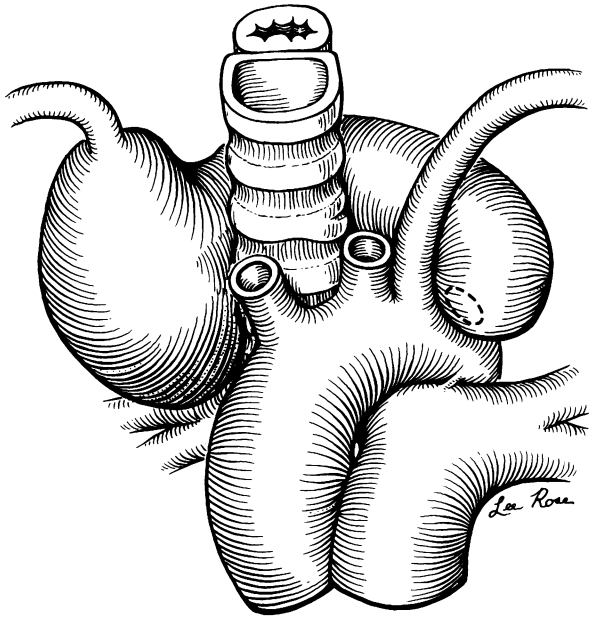

Fig. 1 *Anatomy of an anomalous right subclavian artery passing behind the esophagus, and an aneurysm of the subclavian artery beginning at the Kommerell diverticulum.*

woman who experienced moderate pain over an easily palpable 8- x 11-cm aneurysm. An ultrasound study suggested the aneurysm's infrarenal location. A quantitative radionuclide renal scan using sodium pertechnetate (^{99m}Tc) showed a lack of blood flow to the left kidney, which was atrophic on ultrasound examination. Arteriography and contrast abdominal computed tomographic scanning were deferred because the patient had experienced a severe anaphylactic reaction to intravenous contrast dye during a previous hospital admission. In addition, she had a serum creatinine of 2.7 mg/dL, indicating severe renal insufficiency. The results of liver, pancreatic, and gallbladder studies were normal. Her resting partial pressure oxygen tension (PO_2) was 57 mmHg, and her partial pressure carbon dioxide tension (PCO_2) was 42 mmHg. Chest radiographic findings were consistent with severe chronic obstructive pulmonary disease and a 150-pack-year smoking history. Chest radiography also showed ectasia of the thoracic aorta, and an asymmetric superior mediastinal mass that lay mainly to the right of the midline (Fig. 2A). To further delineate the thoracoabdominal aorta, the patient was scheduled for magnetic resonance imaging (MRI). Before this technique could be performed, however, she experienced sudden increased abdominal pain that was centered over her pulsating aorta, radiating to her back. She was taken to the operating room for an exploratory laparotomy.

The laparotomy disclosed an 8-cm-diameter aneurysm involving the mesenteric and infrarenal aorta. Another 5-cm-diameter aneurysm began at the origin of the superior mesenteric artery and projected into the small bowel mesentery. The left kidney was atrophic, and the left renal vein was thrombosed. In light of these findings and with no evidence of rupture, we decided not to proceed with resection.

Postoperatively, the patient had a moderately difficult recovery, characterized by bradycardia, heart block, respiratory insufficiency, and confusion. Before being discharged from the hospital, she finally underwent MRI, which showed an anomalous right subclavian artery with a 5-cm-diameter aneurysm at its origin (Fig. 2B). The extent of the abdominal aortic aneurysm was also documented (Figs. 2C and 2D).

The final diagnosis was an unresectable mesenteric and infrarenal abdominal aneurysm with an associated superior mesenteric artery aneurysm, an anomalous right subclavian artery, and severe renal and pulmonary insufficiency. The patient's pain was most likely related to progressive expansion of the abdominal aortic aneurysm or superior mesenteric artery aneurysm rather than to renal infarction. One year postoperatively, the patient is alive but suffers from severe chronic obstructive pulmonary disease and progressive renal insufficiency.

Case 2

A 77-year-old man was admitted because of dysphagia and a 20-pound weight-loss over a 2-month period. He had chronic obstructive pulmonary disease, with a resting PO_2 of 60 mmHg and a PCO_2 of 40 mmHg. Recent chest radiography had shown an enlarged mass in the mediastinum and upper right hemithorax. Computed tomography (CT) confirmed the presence of an anomalous right subclavian artery aneurysm, 14 cm in diameter (Fig. 3). The patient's symptoms were compatible with those of dysphagia lusoria, as described by Holzapfel[12] in 1899. Because of progressive dysphagia and inanition, nutritional support was instituted. Aortic arch arteriography and cardiac catheterization disclosed a large aneurysm, which emanated from a Kommerell diverticulum in the proximal descending aortic area and extended to the right thoracic outlet.

The patient underwent a left thoracotomy for ligation and patch closure of the Kommerell diverticulum at its origin in the descending thoracic aorta. The chest incision was then closed, and the patient was repositioned for a right thoracotomy. The subclavian artery was transected at the thoracic outlet, and the proximal portion of the artery was ligated to obliterate the aneurysm, as described by Baillot and associates.[13] An 8-mm GORE-TEX® graft was interposed from the ascending aorta to the transected end of the subclavian artery (Fig. 4). Once subclavian flow had been reestablished, the chest was closed, and the patient was transferred to the recovery room in satisfactory condition.

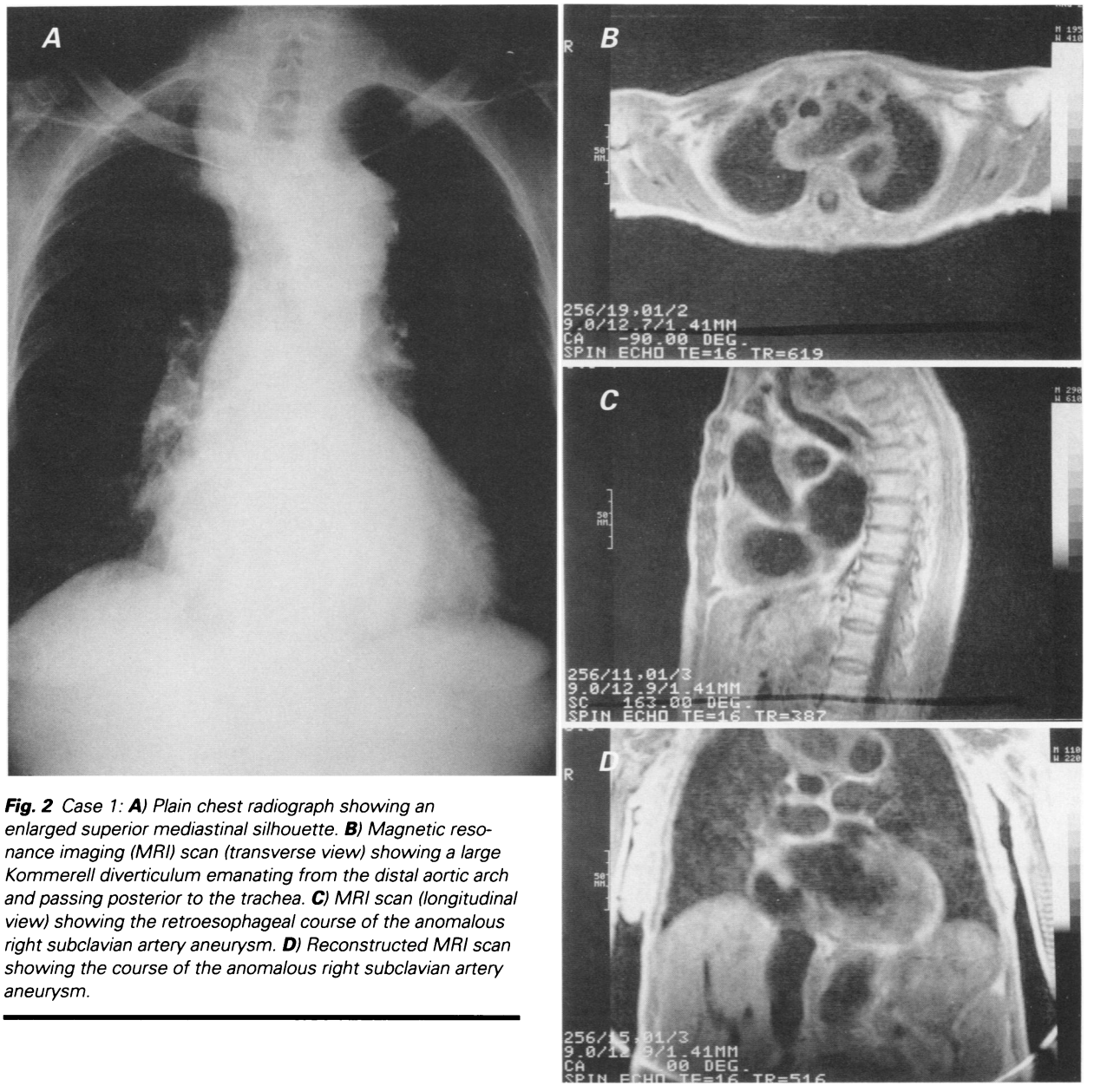

Fig. 2 *Case 1: **A**) Plain chest radiograph showing an enlarged superior mediastinal silhouette. **B**) Magnetic resonance imaging (MRI) scan (transverse view) showing a large Kommerell diverticulum emanating from the distal aortic arch and passing posterior to the trachea. **C**) MRI scan (longitudinal view) showing the retroesophageal course of the anomalous right subclavian artery aneurysm. **D**) Reconstructed MRI scan showing the course of the anomalous right subclavian artery aneurysm.*

Postoperatively, he remained on mechanical ventilation support for 3 days because he had pain and splinting, with respiratory insufficiency. Doppler studies revealed a low-amplitude monophasic pulse wave form in his right upper extremity, indicating occlusion of the GORE-TEX® graft. He was returned to the operating room for re-exploration, thrombectomy, and revision of the graft at its juncture with the ascending aorta. These procedures allowed normal graft function to be restored, and the patient was discharged from the hospital 16 days after surgery, in stable condition. Six months later, he died of complications secondary to respiratory insufficiency and metastatic colon cancer.

Case 3

During an airline flight, a 73-year-old woman suddenly collapsed, necessitating an emergency landing at St. Louis International Airport. Paramedics inserted an endotracheal tube and began intravenous fluid and dopamine support to maintain a systolic blood pressure of 90 mmHg. Upon arrival at our emergency room, the patient was hypotensive (systolic blood pressure, 60 to 70 mmHg) and responded appropriately to verbal commands. Her pupils were equal, and no focal neurologic deficits were noted. Her past medical and surgical history, as reported by her husband, was unremarkable. Chest radiography showed a mass in the right hemithorax, and needle aspiration yielded blood. Computed tomography revealed a ruptured aneurysm of the aortic arch posteriorly but could not indicate whether the aneurysm was proximal or just distal to the aortic isthmus (Figs. 5A, 5B, and 5C). With blood transfusions and dopa-

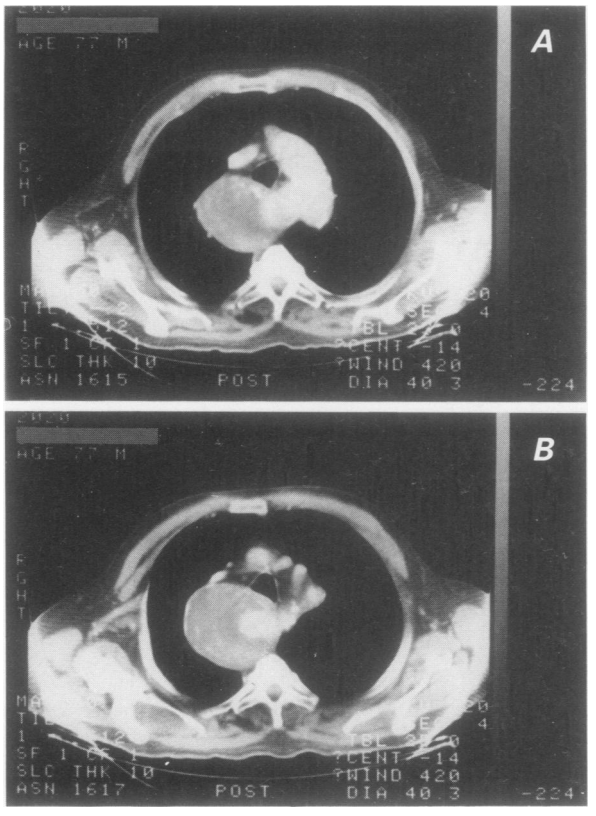

Fig. 3 *Computed tomographic (CT) scans in Case 2.* ***A****) Transverse view of the aortic arch showing a Kommerell diverticulum that turns into a large anomalous right subclavian artery aneurysm.* ***B****) Further enlargement of the anomalous right subclavian artery aneurysm (14 cm).*

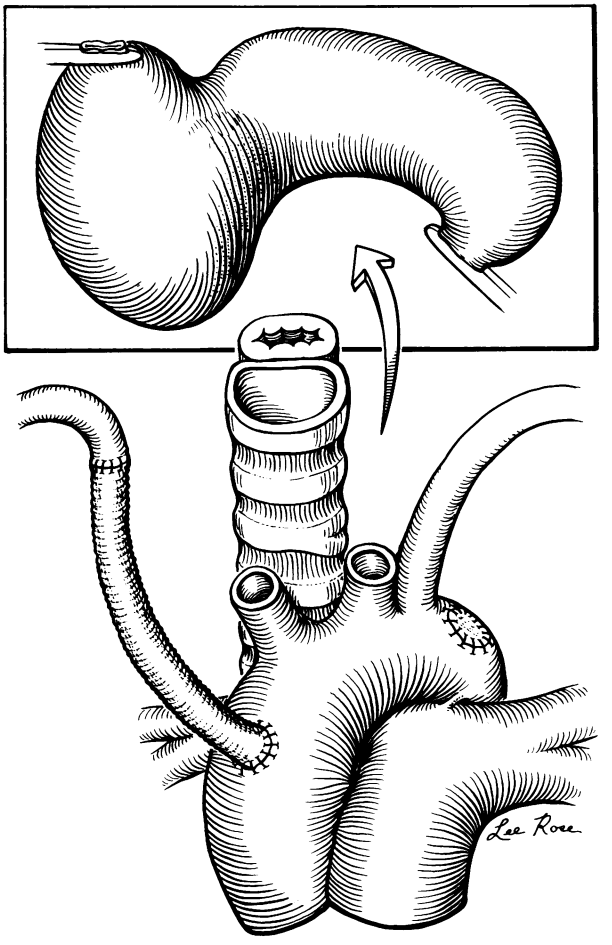

Fig. 4 *Schematic drawing of final operative procedure in Case 2, illustrating resection of the anomalous right subclavian artery aneurysm and grafting of the ascending aorta to the subclavian artery.*

mine support, the patient's blood pressure temporarily remained stable. Her sensorium waxed and waned, depending on her blood pressure. Emergency arteriography of the aortic arch disclosed a large, out-pouching Kommerell diverticulum that emanated from the aortic arch (Fig. 5D).

The patient was taken immediately to the operating room, where cardiopulmonary bypass was instituted via femoro-femoral and right atrial cannulation. Circulatory arrest was initiated at 16 °C, as described by Cooley,[14] during which time the heart was arrested with hyperkalemic autologous blood cardioplegia and topical hypothermia. The patient was then placed in a 20° Trendelenburg position, and pump flow was decreased to 50 mL/min. Once the supracoronary ascending aorta had been opened longitudinally, the interior of the aortic arch revealed frank rupture of a saccular aneurysm (Kommerell diverticulum) located posteriorly between the left common carotid and left subclavian arteries. Primary patch closure was impossible because the thin, friable aortic wall was totally disrupted. The Kommerell diverticulum was ligated distal to the site of rupture and was resected at its origin from the aortic arch. The aortic arch was then resected, preserving the outer curvature and greater arch vessels, which were then sutured onto a beveled woven Dacron tube graft as a unit, with no individual anastomoses.[14,15] Rewarming was initiated, and normal cardiac activity was resumed. A tube was inserted into the right side of the chest to evacuate old blood.

The patient was weaned from cardiopulmonary bypass without incident and was taken to the recovery room in critical, but stable, condition. Her postoperative course was complicated by vascular insufficiency of the right upper extremity, as evidenced by a monophasic, systolic, right-radial-artery pressure of 60 mmHg, compared to a left-radial-artery pressure of 125 mmHg. She exhibited no focal neurologic deficits and was weaned from the ventilator and extubated on the 6th postoperative day. Subsequently, she suffered periodic apnea (Ondine's curse)[16] and bouts of aspiration, but both she and her family refused permission for reintubation or a tracheos-

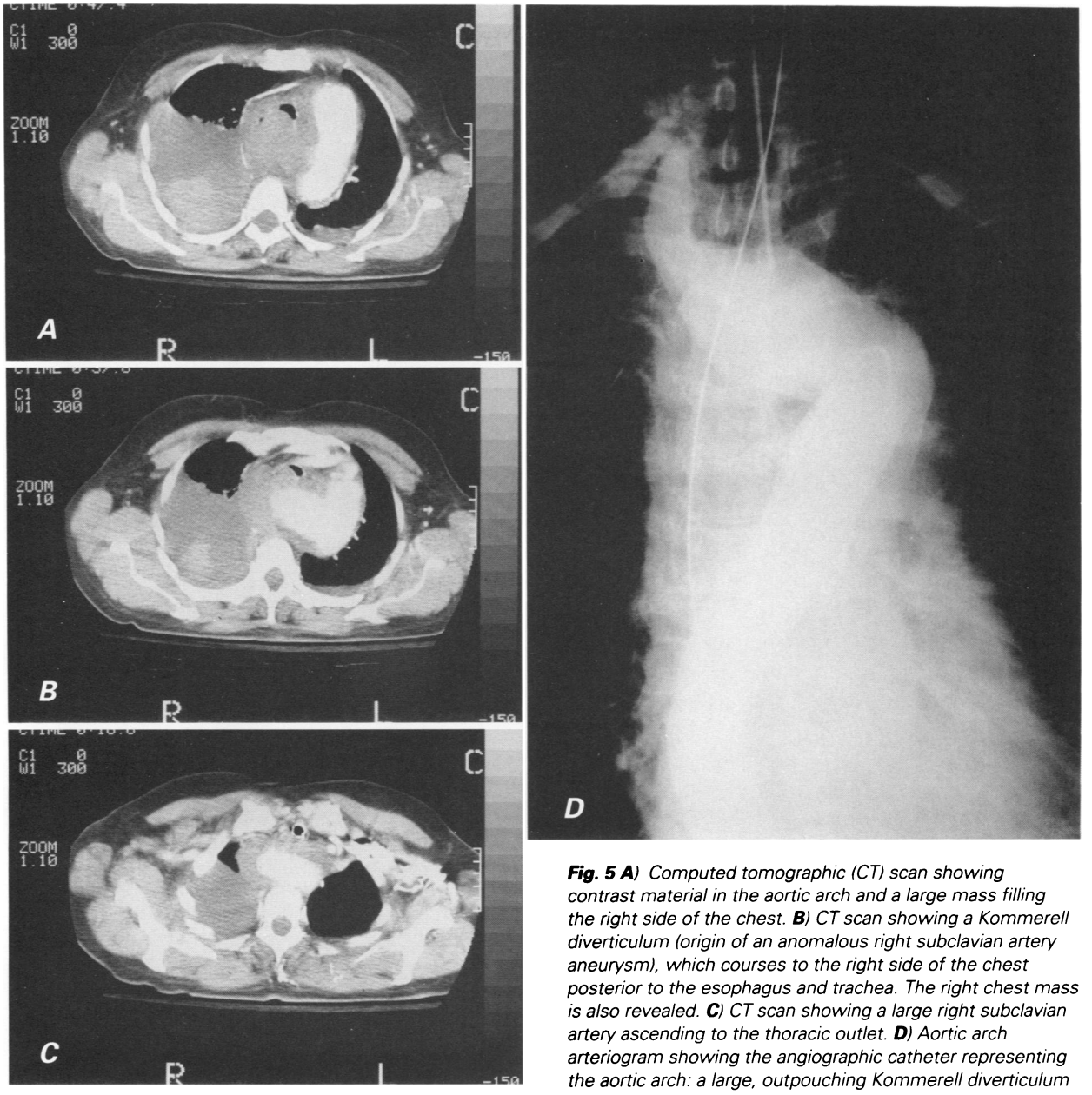

***Fig. 5 A**) Computed tomographic (CT) scan showing contrast material in the aortic arch and a large mass filling the right side of the chest. **B**) CT scan showing a Kommerell diverticulum (origin of an anomalous right subclavian artery aneurysm), which courses to the right side of the chest posterior to the esophagus and trachea. The right chest mass is also revealed. **C**) CT scan showing a large right subclavian artery ascending to the thoracic outlet. **D**) Aortic arch arteriogram showing the angiographic catheter representing the aortic arch: a large, outpouching Kommerell diverticulum emanates from the aortic arch; the left subclavian and the right and left common carotid arteries are faintly silhouetted.*

tomy. Thirteen days after surgery, she died of aspiration, asphyxia, and cardiac arrest. Autopsy revealed an anomalous right subclavian artery aneurysm, with rupture of a large Kommerell diverticulum in an unusually proximal location on the posterior wall of the aortic arch. Only then did family members reveal that the patient had been told several years earlier that she had an "unresectable arch aneurysm."

Discussion

Literature reviews of aberrant right subclavian artery aneurysms confirm their being a rare surgical entity with high morbidity and mortality. The most recent reviews, by Austin[5] and Jauch,[6] revealed a total of 37 anomalous right subclavian artery aneurysms up to 1988. This study and additional isolated reports[17-22] raise the number of anomalous right subclavian artery aneurysms to 49 (Table I).

Most of the patients reported were men between 50 and 75 years old, and two-thirds of them had symptoms—most commonly dysphagia, dyspnea, and chest pain. One of 3 entities usually prevailed: an uncomplicated aortic Kommerell diverticulum; a Kommerell diverticulum that had been transformed into an aneurysm; or an aneurysm of a simple (nondiverticular) anomalous right subclavian artery. The 1st entity was by far the most common finding.

Table I. Review of the Literature Describing Anomalous Right Subclavian Artery Aneurysms.

Case	Author	Year	Age	Sex	Diameter/ Size (cm)	Symptoms	Method of Diagnosis	Surgical Therapy	Subclavian Artery Reconstruction	Survival
1	McCallen and Schaff[10]	1956	50	M	4	None	Operation	Thoracotomy	ND	Yes
2	Richards and Elliott[23]	1957	70	M	4	Dysphagia, neck swelling, rupture	Autopsy	No	No	No
3	Shannon[24]	1961	62	F	ND	Chest pain	Angiography	Left thoracotomy	No	No
4	Candardjis[25]	1961	42	F	ND	Dyspnea	Angiography	Left thoracotomy	No	Yes
5	Ryan[26]	1962	19	M	ND	Dysphagia, dyspnea, rupture	Autopsy	No	No	No
6	Gomes[11]	1968	60	M	7	None	Angiography	Left thoracotomy	No	Yes
7	Lynn[27]	1969	61	M	"Grapefruit"	Hematemesis, rupture	Operation	Median sternotomy	No	No
8	Hunter[9]	1970	71	M	10	Cough	Operation	Median sternotomy	No	No
9	Hunter[9]	1970	55	M	10	Dysphagia	Angiography	Left thoracotomy	No	Yes
10	Hunter[9]	1970	71	M	15	Dysphagia, dyspnea, chest pain, cough	Angiography	Left thoracotomy	No	Yes
11	Campbell[28]	1971	69	M	8	Dysphagia	Angiography	Right thoracotomy	Yes	Yes
12	Engelman and Madayag[29]	1972	74	M	"Large"	None	Angiography	Median sternotomy	No	No
13	Engelman and Madayag[29]	1972	71	M	ND	None	Angiography	No	No	Yes
14	Sarot[30]	1973	65	M	ND	Dysphagia, pain, neck swelling	Angiography	Right thoracotomy	No	Yes
15	Sakurai[31]	1973	40	F	3	Dyspnea	Angiography	Left thoracotomy	No	Yes
16	Lui[32]	1973	77	M	ND	None	Angiography	No	No	Yes
17	Dikman[33]	1974	77	F	9	Hoarseness, vertigo, rupture	Autopsy	No	No	No
18	Stoney[34]	1975	63	M	ND	Dysphagia, dyspnea	Angiography	Left thoracotomy, right supraclavicular incision	Yes	Yes
19	Reynes[35]	1976	72	F	5	Vertigo, hematemesis, rupture	Angiography	No	No	No

CT = computed tomography; MRI = magnetic resonance imaging; ND = not described

Case	Author	Year	Age	Sex	Diameter/ Size (cm)	Symptoms	Method of Diagnosis	Surgical Therapy	Subclavian Artery Reconstruction	Survival
20	Wagner and Thiry[36]	1976	75	F	11	Dyspnea, pain	Autopsy	No	No	No
21	Wagner and Thiry[36]	1976	62	M	ND	Dysphagia, dyspnea, rupture	Angiography	ND	ND	No
22	Wagner and Thiry[36]	1976	79	F	"Spoon"	Dysphagia, torticollis	Autopsy	No	No	No
23	Wagner and Thiry[36]	1976	78	F	3	Dyspnea, pain, rupture	Autopsy	No	No	No
24	Cunningham[37]	1977	56	M	ND	None	Angiography	ND	No	No
25	Rodgers[38]	1978	11	F	15	Dyspnea, Horner's syndrome (right eye)	Angiography	Right thoracotomy	No	Yes
26	Dartevelle[39]	1978	73	F	2.5	Dysphagia	Angiography	Left thoracotomy	No	Yes
27	Jourdan[40]	1979	53	M	"Tennis ball"	Dysphagia, dyspnea, chest pain	Operation	Right thoracotomy	Yes	Yes
28	Schmidt[41]	1980	67	M	5	Splinter hemorrhages	Angiography	Left thoracotomy	No	Yes
29	McIntyre and Lynn[42]	1980	64	M	ND	Chest pain	CT scan, operation	Right thoracotomy	No	Yes
30	Goebel and Turina[43]	1981	49	F	ND	ND	Angioplasty	Right and left thoracotomies	No	Unknown
31	Frija[44]	1982	66	M	ND	Dysphagia	CT scan	ND	ND	ND
32	Esquivel and Miller[45]	1984	74	M	ND	Chest pain, hoarseness	CT scan	Left thoracotomy, right chest supraclavicular incision	Yes	Yes
33	Glock[46]	1984	75	M	10	Hematemesis, rupture	Angiography	Sternotomy, left thoracotomy	ND	No
34	Lupetin[47]	1984	77	M	ND	None	CT scan	ND	ND	ND
35	Baillot and Cosgrove[13]	1984	66	F	7	None	CT scan, angiography	Sternotomy with cardiopulmonary bypass, hypothermic arrest, and coronary bypass	Yes	Yes
36	Austin and Wolfe[5]	1985	67	F	5	None	CT scan	Right thoracotomy	Yes	Yes
37	Poon and Stewart[18]	1986	72	M	4	None	CT scan, angiography	No	No	Yes

CT = computed tomography; MRI = magnetic resonance imaging; ND = not described

Case	Author	Year	Age	Sex	Diameter/ Size (cm)	Symptoms	Method of Diagnosis	Surgical Therapy	Subclavian Artery Reconstruction	Survival
38	Vega[17]	1987	74	M	6	Chest pain	CT scan, angiography	Right thoracotomy, ligation	Yes	No
39	Jauch[6]	1988	74	F	8	Chest pain	CT scan, angiography	Left thoracotomy	No	Yes
40	Jebara[19]	1988	67	F	5	Dysphagia	CT scan, angiography	Sternotomy	Yes	Yes
41	Hardy[16]	1989	64	F	7	Tracheal compression, stridor	Angiography	Sternotomy, ligation	No	No
42	Mulligan[21]	1989	40	M	ND	Aortic dissectional rupture	CT scan, angiography	Thoracotomy	No	Yes
43	Mulligan[21]	1989	79	F	ND	Ruptured anomalous right subclavian artery aneurysm and aortic arch	Angiography	Thoracotomy	ND	No
44	Kullnig[20]	1989	66	M	"Large"	Hematemesis	CT scan	No	No	No
45	Stone[22]	1990	72	M	10	Dysphagia	CT scan, angiography	Left thoracotomy, right supraclavicular incision	Yes	No
46	Gordini[48]	1991	53	M	4.7	Hoarseness, dyspnea	CT scan, MRI, angiography	Right supraclavicular incision, left thoracotomy	Yes	Yes
47	Knight and Codd	1991	72	F	5	None	MRI	No	No	Yes
48	Knight and Codd	1991	77	M	14	Dysphagia	CT scan, angiography	Left and right thoracotomies	Yes	Yes
49	Knight and Codd	1991	73	F	"Large"	Ruptured anomalous right subclavian artery aneurysm and aortic arch	CT scan, angiography	Median sternotomy	No	No

CT = computed tomography; MRI = magnetic resonance imaging; ND = not described

Symptomatic anomalous right subclavian artery aneurysms are true surgical challenges. Even when operative repair is successful, long-term survival may be limited by significant associated medical problems. Our 3 cases included a diverse array of presentations and complications. In case 3, rupture of the anomalous right subclavian artery aneurysm was difficult to distinguish from a ruptured atherosclerotic saccular aneurysm of the mid-aortic arch. Failure to differentiate between these 2 entities can result in right-arm ischemia.

As suggested by Baillot and associates,[13] 1 proven surgical option involves obliteration of the aneurysm, intraluminal patch closure of the Kommerell diverticulum, and transection of the distal subclavian artery, followed by carotid-subclavian artery bypass. Another option consists of a bilateral thoracotomy technique in which the aneurysm is excised or oblit-

erated, and the right subclavian artery is regrafted from the ascending aorta, as in our 2nd case (Fig. 4). The most accepted technique reported in recent literature involved ligation of the subclavian artery distal to the aneurysm, excision of the aneurysm, and oversewing of the aortic end. Of the 49 cases reported, only 33 came to surgery, of which 11 had right subclavian artery bypass. In 3 of the 22 patients who did not undergo bypass, limb ischemia developed, resulting in fingertip necrosis. Most authors agree that revascularization decreases the risk of ischemia and prevents a subclavian steal syndrome.[49] Technically, revascularization is most easily performed through a right thoracotomy incision, but this approach limits control of the treacherous connection between the anomalous subclavian artery aneurysm and the aorta. In most cases, preliminary supraclavicular extra-anatomic restoration of flow to the right subclavian artery, followed by a left thoracotomy, is preferred. This approach might have eliminated our problem in case 2, in which postoperative graft occlusion necessitated reopening the right thorax for thrombectomy and graft revision.

The 1st surgeon to reestablish flow to the right subclavian artery was Campbell,[28] who, in 1971, opened an aberrant right subclavian artery aneurysm and placed a fabric graft within it. He then sewed the aneurysmal wall over the graft, leaving the graft in a retroesophageal position. In 1975, Stoney and colleagues[34] performed a successful resection and bypass procedure using a left thoracotomy and right supraclavicular approach. Four years later, Jourdan and associates[40] completely excised an aneurysm through a right thoracotomy and interposed a 12-mm Dacron graft, leaving it in the retroesophageal position. In 1984, Esquivel and Miller[45] performed an axillo-axillary bypass with an 8-mm polytetrafluoroethylene (PTFE) graft and then excised the aneurysm through a left thoracotomy. That same year, Baillot[13] performed a combined aortocoronary bypass procedure and repair of an anomalous right subclavian artery aneurysm with the help of hypothermic circulatory arrest, then completed the revascularization with a carotid-subclavian bypass through a right supraclavicular incision.

Of the 49 patients in this current review, 33 were treated surgically; of the 16 remaining patients, 2 are not described. The diameters of the aneurysms ranged from 2.5 to 15 cm. Of the 33 patients who underwent surgery, 10 died, for an operative mortality of 30%. Of the 14 patients who were not treated surgically, 8 died, for a nonoperative mortality of 57%.

Rupture of the aberrant right subclavian artery aneurysm occurred in 11 patients, and 5 of these patients underwent surgery. The 6 nonoperative patients died of a ruptured anomalous subclavian artery aneurysm, resulting in a nonoperative mortality rate of 100%. One of the 5 surgery patients survived; hence, the operative mortality rate was 80%.

Our series illustrates the ongoing difficulties surgeons experience in treating these patients. As case 3 shows, an anomalous right subclavian artery aneurysm can be extremely difficult to diagnose. On emergency preoperative angiograms, such an aneurysm may be interpreted as a ruptured innominate artery aneurysm or a saccular aneurysm of the aortic arch. It is not uncommon for the patient to also have other aneurysms; as in case 1, these can be more life-threatening than the subclavian lesion itself. These patients, in addition to being elderly, often have associated medical conditions such as coronary artery disease, chronic obstructive pulmonary disease, cerebrovascular disorders, and renal impairment. These conditions will adversely affect the patient's long-term survival after surgical repair of an anomalous right subclavian artery aneurysm.

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