

Thoracic endovascular aortic repair of aortobronchial fistulas

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Background: Thoracic endovascular aortic repair of aortobronchial fistulas is an emerging treatment modality for this highly lethal condition. The feasibility and long-term durability of this form of intervention are largely unknown.

Methods: The records of five patients who received endografts to treat aortobronchial fistulas at our institution were reviewed. A literature review was also conducted using MEDLINE to identify reports detailing outcomes of patients undergoing thoracic endovascular aortic repair for this condition. Primary outcome end points included intraoperative mortality, 30-day mortality, and aortobronchial fistula recurrence.

Results: For the five patients treated at our institution, technical success was 100%. In follow-up, aortobronchial fistulas recurred in two patients, resulting in one patient death and one endograft explantation. We identified 32 reports that met inclusion for our final review. Inclusive of the five patients treated at our institution, 67 patients with reported outcomes comprised the overall analysis. Most patients (55%) had previously undergone thoracic aortic surgery. Commercially manufactured thoracic endografts were used in 75% of patients. No intraoperative mortality was reported, and the 30-day mortality was 1.5%. Aortobronchial fistula recurred after endovascular repair in six patients (9%) through a mean follow-up of 21.5 months. Three cases of recurrent aortobronchial fistula resulted in patient death.

Conclusions: Thoracic endovascular aortic repair of aortobronchial fistulas appears to be a viable alternative to conventional open repair with excellent short-term results. Recurrence of the aortobronchial fistula after endovascular repair is a potential complication necessitating long-term surveillance. Individual risk assessment is needed to determine if endovascular repair should be used as bridge therapy or as a definitive repair. (*J Vasc Surg* 2009;50:992-8.)

Aortobronchial fistula (ABF) is a rare, life-threatening condition that represents a communication between the thoracic aorta and the tracheobronchial airway. This condition often occurs in the setting of an atherosclerotic aneurysm or pseudoaneurysm after thoracic aortic surgery.¹ ABFs are highly lethal if they are not diagnosed and remain untreated.¹⁻³ A definitive diagnosis, however, can sometimes be difficult, but an ABF should be highly suspected in patients with a history of known or previously treated thoracic aortic pathology.

Conventional open surgical intervention for ABFs is associated with substantial morbidity and mortality.^{1,3} The high complication rate can be attributed to the need for an emergency intervention in patients with multiple pre-existing comorbidities through what will often be a reoperative surgical approach. Recent advances in endovascular technology have allowed for endoluminal repair of multiple aortic pathologies, with the potential for improved outcomes compared with traditional interventions. For ABFs, thoracic endovascular aortic repair (TEVAR) affords the ability to rapidly obtain hemorrhage control while avoiding the inherent morbidity associated with thoracic aortic exposure and aortic cross-clamping.

Experience with endovascular repair of ABFs is greatly limited by the rarity of this condition and the relatively recent evolution of TEVAR procedures. Reported outcomes after TEVAR for ABFs have been largely limited to individual case reports⁴⁻¹⁶ or small case series.¹⁷⁻²⁴ To evaluate the feasibility of TEVAR for ABFs, we reviewed our single-institution case series and initiated an extensive review of the published experience to date.

METHODS

Institutional Review Board approval was obtained for the review of all patient records relevant to this study. A search was conducted of our institutional TEVAR Database from March 1998 to September 2007. During this time period, 195 patients received endografts for multiple thoracic aortic pathologies, and five (2.6%) underwent emergency TEVAR for ABFs. A full review of radiographic, electronic, and paper records was performed. Anatomic requirements, sizing of endografts, and follow-up were as previously described.²⁵

A search of the MEDLINE database was conducted from January 1990 to January 2008 through PUBMED using the keywords *aortobronchial*, *aorto-bronchial*, *aortopulmonary*, *aorto-pulmonary*, *aortotracheal*, and *aortobronchopulmonary*. The search was limited to English-language literature and the treatment of adult patients (aged ≥ 18 years). Abstracts were screened for articles describing at least one patient undergoing TEVAR for an ABF. References in the identified articles were further screened for additional publications not identified in the initial database search. Articles were selected for further review and inclusion in the final analysis if they described individual outcomes for patients treated for ABFs. Patients reported as

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Competition of interest: Dr Farber is a consultant for Cook Inc, W. L. Gore & Associates Inc, Bolton Medical, and Medtronic Inc.

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0741-5214/\$36.00

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doi:10.1016/j.jvs.2009.03.001

Table I. Patient characteristics

Patient	Age	Sex	Diagnosis	Blood culture	Previous thoracic aortic surgery ^a
1	53	M	PA/mycotic	<i>S aureus</i>	DTA repair (0.7 y)
2	77	M	PA	...	Ao-L SCA bypass (35 y)
3	52	M	PA	...	
4	71	M	PA	...	DTA repair (7 y)
5	71	F	Mycotic	<i>S aureus</i>	

Ao-L SCA, Aorta to left subclavian artery; DTA, descending thoracic aneurysm; PA, pseudoaneurysm.

^aYears before endovascular intervention in parentheses.

part of an overall single-institution TEVAR series were typically excluded because these studies often only reported summarized outcome data of the endovascular treatment of multiple thoracic aortic pathologies. Multiple publications originating from a single institution describing the same patients were identified and the older publications were excluded.

Data points of interest included age, gender, associated thoracic aortic pathology, diagnostic procedures performed before TEVAR, endografts used, intraoperative mortality, 30-day mortality, and ABF recurrence. The number and percent reporting for each respective data point is presented when <100% of the data points were obtained from the total number of identified patients. Mean values that were reported for data points of interest in individual ABF case series^{20,21} were weighted against the number of reported patients in the respective report for the final calculations.

RESULTS

Characteristics of the five patients from our institutional series are compiled in Table I. Three patients presented with pseudoaneurysms associated with prior surgery of the thoracic aorta. An additional patient, who had undergone anterior spinal fixation for complications from multiple myeloma, presented with a small pseudoaneurysm at the level of his spinal hardware. Computed tomography (CT) angiography findings, laboratory studies, and physical examination findings were not suggestive of an infectious cause. The patient was assessed as most likely having sustained an iatrogenic thoracic aortic injury at the time of his spinal procedure. The final patient presented with a primary mycotic aneurysm.

CT was performed in four patients who presented to the hospital with a history of recurrent hemoptysis. Aortography was used to evaluate the fifth patient who presented with a primary mycotic thoracic aortic aneurysm after an acute episode of hemoptysis shortly after admission. Although all studies demonstrated associated thoracic aortic pathology, none directly demonstrated a fistula. Bronchoscopy was performed in two patients, without identification of a fistulous orifice.

Because the study period spanned the era before and after thoracic endograft approval in the United States,

the devices used included approved thoracic devices, compassionate-use thoracic devices, and modified abdominal endograft components (Table II). Technical success was 100% as assessed by intraoperative angiography and postoperative CT scans.

Patient outcomes are presented in Table II. Two patients experienced prolonged hospital stays (>7 days) after endovascular repair. In one of these patients, who presented with a pseudoaneurysm at the proximal anastomotic site of an aorta to left subclavian bypass, multiple pneumonias developed as well as a cerebrovascular accident 2 weeks after his intervention. After discharge to an acute care facility, he made a complete functional recovery. Life-long prophylactic antibiotics were recommended due to the potential for endograft contamination from the multiple infections the patient experienced during his hospital stay. CT scans at 6 months and 1 year demonstrated progressive resolution of his periaortic hematoma (Fig). In the second patient, who presented with a primary mycotic aneurysm, the hospital stay was prolonged due to multiple fungal and bacterial pneumonias. This patient was discharged to an acute care facility and died 5 months after TEVAR from complications of pre-existing end-stage renal disease.

Two patients experienced recurrent episodes of hemoptysis \leq 48 hours after TEVAR. In both patients, CT angiography evaluation demonstrated no evidence of endoleak and bronchoscopy demonstrated no evidence of active bleeding. These episodes resolved and were attributed to expectoration of residual clots in the airways because there was no significant change in the patients' hemoglobin or other signs of a persistent fistulous connection.

Two patients (40%) experienced an ABF recurrence during follow-up. In one patient, hemoptysis developed 11 months after TEVAR during hospitalization for acute pancreatitis. CT angiography revealed periaortic inflammation at the level of the endograft. The patient refused intervention, despite being amenable to further endovascular repair, and died after a massive hemoptysis.

The second patient, who had developed a pseudoaneurysm after spinal surgery, presented 33 months after TEVAR with recurrent hemoptysis, *Pseudomonas* bacteremia, and pneumonia. The patient had previously experienced multiple admissions at another hospital for various infections. CT angiography at the time of presentation demonstrated proximal aortic aneurysmal enlargement and periaortic inflammation. The patient's immune dysfunction from myeloma may have been a contributing factor in the development of an endograft infection. The patient underwent endograft explantation through a two-stage procedure. During the initial stage, an ascending to descending thoracic aortic bypass was performed through a median sternotomy. After partial recovery, the patient underwent thoracic aortic resection and wedge resection of the left upper lobe through a left thoracotomy. The patient made a full recovery without complications. Imaging at 6 months demonstrated no anastomotic concerns and resolution of the residual pulmonary inflammation.

Table II. Patient outcomes

Patient	Endograft	No. ^a	Length of stay, d	Antibiotics at discharge	Length of follow-up, mon	Recurrent ABF	Mortality (mon)
1	Zenith ^b	1	7	Yes	11	Yes	Yes (11 mon)
2	Zenith ^b	3	50	Yes	12	No	No
3	TAG ^c	1	1	No	35	Yes	No
4	TAG ^c	1	6	No	6	No	No
5	Talent ^d	2	64	Yes	5	No	Yes (5 mon)

ABF, Aortobronchial fistula.

^aNumber of endograft components deployed.

^bZenith AAA endovascular graft (Cook, Bloomington, Ind).

^cW. L. Gore & Associates, Flagstaff, Ariz.

^dMedtronic AVE, Santa Rose, Calif.

Literature review. We reviewed 40 case reports and case series, of which eight were excluded from the final analysis due to reports on the same patients in prior publications. Included were 11 publications that reported patients with ABF along with other thoracic aortic pathologies.²⁶⁻³⁶ The 32 reports used in the final analysis are presented in Table III. Inclusive of the five patients described from our institutional series, data from 67 patients were available for the overall analysis.

Of the 64 patients for whom age and gender were reported, the mean age was 64.3 years and 69% were men. Most reported cases (55%) occurred in patients who had previously undergone thoracic aortic surgery, and of these 36 patients, 14 (39%) had undergone the initial aortic operation for aneurysms or pseudoaneurysm of the descending thoracic aorta and 13 (36%) for congenital aortic coarctation. Accounting for reoperative aortic procedures, the mean time from the most recent aortic surgery to TEVAR for the 26 patients reported was 13.2 years.

Most patients (75%) received commercially manufactured thoracic endografts (Table IV). Hand-made devices comprised approximately 14%, although these predominantly occurred in earlier reports or from countries with limited endograft availability. A mean of 1.3 components were deployed per patient. Nine secondary endovascular procedures were performed in eight patients, and seven of these were performed \leq 30 days from the initial procedure. Seven procedures (78%) were performed because of a documented endoleak.

Early postoperative recurrent hemoptysis was reported in eight patients (12%), although in seven the hemoptysis resolved without intervention. Hemoptysis recurred 48 hours after TEVAR in one patient. This was found to be due to an endoleak with persistence of the ABF⁷ and was successfully treated with deployment of an additional component.

Length of stay averaged 11.5 days (range, 1-57 days), but this was only reported in 38 patients (57%). No intraoperative deaths were reported. The only death (1.5%) that occurred \leq 30 days was in a patient with pneumonia who refused intubation.⁶ The ABF recurred in six patients (9%) at a mean of 13.2 months after TEVAR (range, 2 days-33 months). Three cases of recurrence were successfully

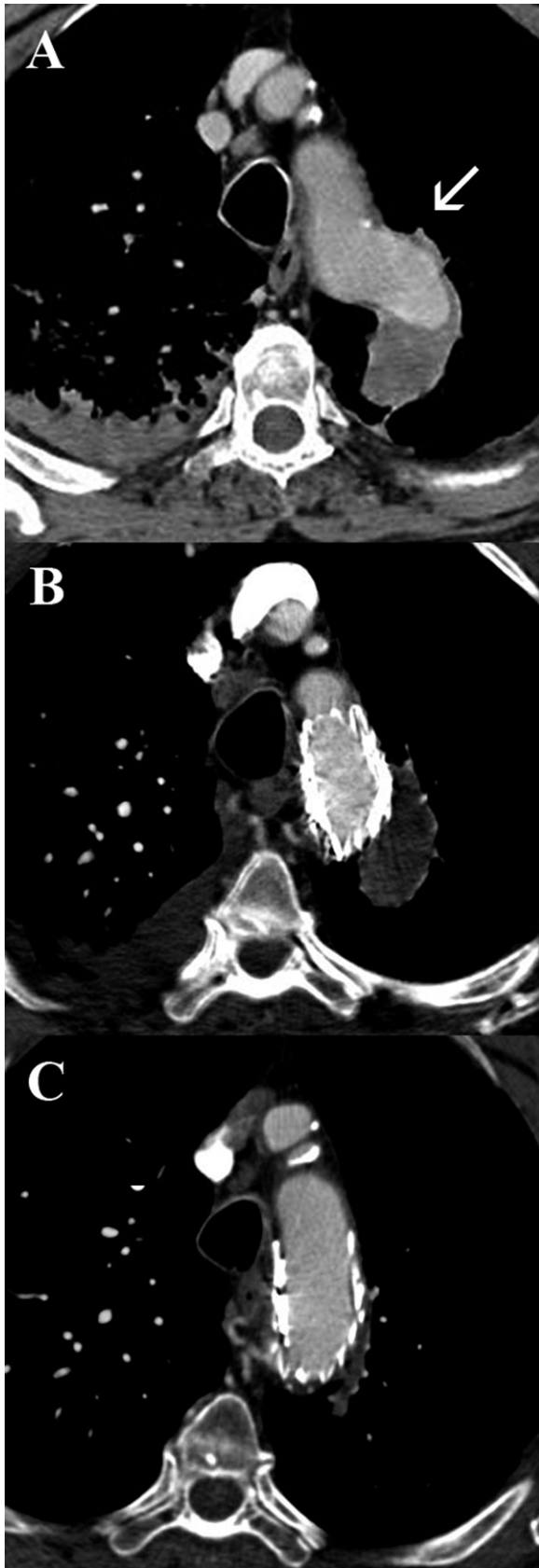
treated with additional operative or endovascular interventions, and three cases resulted in patient death.

DISCUSSION

ABF is a rarely encountered clinical entity. A literature review published in 1991 identified 63 reported cases.¹ An additional review of postoperative ABFs revealed only 76 patients reported through 2002.³ When left untreated, this condition is invariably fatal, with reported mortality rates of 100%.¹⁻³

Patients who develop ABFs often present with hemoptysis that is mild and intermittent in nature for which the etiologic differential is diverse. The ability to diagnose this condition is further confounded by the lack of a diagnostic test that can clearly demonstrate the presence of the fistula. Although helical CT imaging will rarely demonstrate a fistulous communication between the aorta and the lung, thoracic aortic pathology with adjacent lung parenchymal changes are highly suggestive of the diagnosis in this clinical setting.³⁷ Aortography and bronchoscopy are often part of the evaluation of hemoptysis; however, both of these modalities often fail to demonstrate the fistula² and may potentially induce massive hemorrhage.³

Traditional operative intervention consists of closure of the aortic defect with anatomic reconstruction, débridement and drainage, or possibly extra-anatomic bypass and resection. This treatment affords the ability to directly address both the aortic and bronchopulmonary defects as well as perform surgical débridement of infected tissues in cases of a mycotic aneurysm. Open surgical repair has been reported to carry an operative mortality rate of 15.3% to 24%.^{1,3,38} Much of this mortality may largely be due to the need to perform reoperative thoracic exposures in patients who have previously undergone thoracic aortic surgery.^{1,3} In a review of the literature, Piciche et al³ reported a 16% operative mortality for patients undergoing open repair of postoperative ABFs, with six of these eight deaths occurring intraoperatively.³ The results from this review of TEVAR of ABFs compare favorably with these historic outcomes, as we could find no reports of intraoperative death and only one hospital death (1.5%) at 30 days in a patient who refused intervention for pneumonia.⁶



Primary mycotic aneurysms have historically been the leading cause of ABFs, but this condition now most commonly presents in the setting of atherosclerotic aneurysms or pseudoaneurysms associated with prior thoracic aortic surgery.¹ Experience with using endografts in patients with mycotic aneurysms is limited, and unlike open surgery, TEVAR does not afford the opportunity to perform débridement of the infected aorta and surrounding tissues. Whether the local infection can be completely resolved when an endoprosthesis is placed within the infected field is unknown. A recent review of the endovascular treatment of mycotic aneurysms suggests that this condition may be successfully managed with endografts; however, patients with ruptured aneurysms, which included patients with ABFs, were more likely to have a persistent infection.³⁹

Our two cases of recurrent ABFs were both related to infection. One ABF recurred at 11 months, despite maintenance antibiotic therapy, in a patient who was infected at the time of TEVAR. The other ABF occurred secondary to a late endograft infection, potentially as a consequence of the patient's immune dysfunction. In situations of an associated graft infection or mycotic aneurysm, it may be more appropriate to use endografts to temporarily achieve hemorrhage control and serve as a bridge to an open surgical intervention. The plan of treatment should be individualized for each patient according to his or her risk factors at the time of presentation.

The appropriate duration of postoperative antibiotics in the presence or absence of documented infection in the setting of ABFs is undefined. Although ABFs may often develop in the absence of an infectious etiology, the lack of closure of the pulmonary defect after TEVAR may expose the endoprosthesis to a contaminated environment. Whether this may increase the patient's risk for the development of an endograft infection is unknown. Our current practice is to prescribe life-long antibiotic therapy for patients after TEVAR for infectious aortic pathology. In the absence of an infectious aortic etiology and an uncomplicated postoperative course, the patient will not be discharged on antibiotics.

Recurrent hemoptysis after TEVAR was commonly reported in the postoperative period, with an incidence of 12%. Many of these cases prompted a diagnostic evaluation owing to concern for persistence of the ABF, but in only one patient was the hemoptysis attributed to the presence of an endoleak with a persistent communication between the aneurysm sac and the bronchopulmonary system. These cases of early postoperative he-

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Fig. Computed tomography angiography of the chest (A) before (B) 6 months, and (C) 12 months after endovascular repair in a patient who presented with massive hemoptysis and was found to have a pseudoaneurysm (*arrow*) at the proximal aspect of an aorta to left subclavian artery bypass. Exclusion of the aneurysm and progressive resolution of the associated hematoma is demonstrated.

Table III. Published case reports and case series of thoracic endovascular aortic repair of aortobronchial fistulas

First author	Year	Patients, No.	Mortality, No. (%)		Recurrent ABF, No. (%)	Mean follow-up, mon
			Intra-op	30-day		
Chuter ⁴	1996	1	0 (0)	0 (0)	0 (0)	7
Campagna ⁵	1996	1	0 (0)	0 (0)	0 (0)	36
Semba ²⁶	1998	1	0 (0)	0 (0)	0 (0)	25
Miyata ¹⁷	1999	2	0 (0)	0 (0)	0 (0)	8 ^a
Karmy-Jones ⁶	1999	1	0 (0)	1 (100)	0 (0)	0.7
Caiati ⁷	2001	1	0 (0)	0 (0)	1 (100)	2
Kramer ²⁷	2001	1	0 (0)	0 (0)	0 (0)	12
Yoo ⁸	2001	1	0 (0)	0 (0)	0 (0)	0.5
Dorweiler ²⁸	2001	3	0 (0)	0 (0)	0 (0)	35.3
Leobon ²⁹	2002	3	0 (0)	0 (0)	0 (0)	19.3
Schoder ³⁰	2002	3	0 (0)	0 (0)	1 (33)	23 ^a
Alric ³¹	2002	1	0 (0)	0 (0)	0 (0)	NR
Bockler ¹⁸	2004	8	0 (0)	0 (0)	1 (13)	28.5
Kotzampassakis ⁹	2004	1	0 (0)	0 (0)	0 (0)	0.5
Numan ¹⁰	2004	1	0 (0)	0 (0)	0 (0)	6
Saratzis ¹¹	2005	1	0 (0)	0 (0)	0 (0)	7
Munneke ¹²	2005	1	0 (0)	0 (0)	0 (0)	12
Jones ³²	2005	1	0 (0)	0 (0)	0 (0)	28
Kawl ¹³	2005	1	0 (0)	0 (0)	0 (0)	2
Beland ¹⁴	2005	1	0 (0)	0 (0)	0 (0)	12
Sayed ²⁴	2005	3	0 (0)	0 (0)	1 (33)	16.7
Abdul-Ghani ¹⁵	2006	1	0 (0)	0 (0)	0 (0)	9
Kaya ³³	2006	2	0 (0)	0 (0)	0 (0)	NR
Matsagas ³⁴	2006	1	0 (0)	0 (0)	0 (0)	18
Kwok ³⁵	2006	1	0 (0)	0 (0)	0 (0)	NR
Takahashi ¹⁹	2006	2	0 (0)	0 (0)	0 (0)	18
Quintana ¹⁶	2006	1	0 (0)	0 (0)	0 (0)	12
Pirrelli ²⁰	2006	5	0 (0)	0 (0)	0 (0)	26.4
Wheatley ²¹	2007	7	0 (0)	0 (0)	0 (0)	42.6
Kokotsakis ²²	2007	2	0 (0)	0 (0)	0 (0)	31.5
Sachithanandan ²³	2007	2	0 (0)	0 (0)	0 (0)	9
Marcheix ³⁶	2007	1	0 (0)	0 (0)	0 (0)	12.9
Present series	2008	5	0 (0)	0 (0)	2 (40)	13.8
Overall		67	0 (0)	1 (1.5)	6 (9.0)	21.5 ^b

ABF, Aortobronchial fistula; NR, not reported.

^aFollow-up reported on one patient in the case series.

^bFollow-up reported for 60 of 67 patients.

moptysis resolution without intervention are most likely the result of resorption and expectoration of the residual mediastinal hematoma after exclusion of the fistula, as suggested by Dorweiler et al.²⁸ Resolution of postintervention hemoptysis secondary to hematoma expectoration may be protracted and has been reported to persist for as long as 2 weeks.²⁸ Given the potential for recurrent ABF from inadequate exclusion from the aortic circulation or retrograde perfusion of the aneurysm sac from intercostal or bronchial vessels, diagnostic evaluation should be conducted if clinically warranted.

The durability of TEVAR for the treatment of ABF is partially assessed by the recurrence of this condition. Our literature review found a reported recurrence rate of 9.0%, presenting at a mean of 13.2 months from the time of intervention. Other than the one case that occurred at 2 days after TEVAR, the remaining cases all presented at ≥ 6 months. These late failures may be the result of primary or secondary aortic infection, as illustrated by our two patients with ABF recurrence, or as a result of progression of the

associated thoracic aortic pathology present at the time of TEVAR. This potential for recurrence, and other late endograft complications, necessitates life-long imaging surveillance as performed for other thoracic aortic pathologies after endovascular repair.

One limitation of this review is that the cases summarized may represent a bias toward good outcomes because clinicians may be unlikely to publish unfavorable results after endovascular management of this condition. In addition, follow-up was limited in several of the case reports and case series. Therefore, the true procedurally related mortality may not be reflected within the first 30 days, although mortality beyond this time frame is probably more a function of the inherent insult of this condition and associated comorbidities in this patient population.

Although early outcomes from this review appear to be favorable compared with reviews of the literature reporting outcomes of open surgical intervention, the results from these studies mainly reflect historical open surgical outcomes from the pre-endovascular era.^{1,3} Outcomes re-

Table IV. Summary of thoracic aortic pathology and endografts used^a

Variable	No.	%
Associated thoracic aortic pathology	66	99
Previous thoracic aortic surgery	36	54.5
Penetrating ulcer	10	15.2
Descending thoracic aortic aneurysm	7	10.6
Primary mycotic aneurysm	6	9.1
Aortic dissection	3	4.5
Other	5	7.6
Endografts	63	94
Talent Thoracic ^b	27	42.9
TAG/Excluder Thoracic ^c	17	27.0
Hand-made	9	14.3
AAA endograft components	7	11.1
Zenith Thoracic ^d	2	3.2
Endofit Thoracic Stent Graft ^e	1	1.6

AAA, Abdominal aortic aneurysm.

^aNumber and percentage reported for the respective value.

^bMedtronic AVE, Santa Rosa, Calif.

^cW. L. Gore and Associates Inc., Flagstaff, Ariz.

^dCook Inc, Bloomington, Ind.

^eEndomed, Phoenix, Ariz.

ported in these studies may not be representative of the results that could presently be obtained due to modern advancements in the field of cardiovascular surgery.

Conversely though, many of the patients who underwent TEVAR in this overall series might have been prohibitive candidates for open surgery and might not have been offered an intervention in the absence of an endovascular option. Direct comparisons between open surgical and endovascular interventions are impractical because of the emergency nature of this condition and the rarity with which it is encountered. Continued evaluation of the feasibility of TEVAR for the treatment of ABFs will largely continue to come from the clinical experience obtained from the endovascular treatment of other thoracic aortic conditions.

CONCLUSIONS

TEVAR is a promising intervention for the treatment of ABFs, with minimal procedurally related mortality. Whether this intervention represents a definitive form of repair or a temporizing measure to obtain hemorrhage control for some associated thoracic aortic conditions remains to be defined. Although early postintervention hemoptysis is common and most likely due to expectoration of hematoma, evaluation for failure of exclusion should be performed if clinically warranted. In addition, the ABF may recur after an initially successful TEVAR procedure, necessitating continued clinical and radiographic surveillance.

AUTHOR CONTRIBUTIONS

Conception and design: PR, JB, MF

Analysis and interpretation: PR, JB, MF

Data collection: PR, JB

Writing the article: PR, JB, MF

Critical revision of the article: PR, MF

Final approval of the article: PR, JB, MF

Statistical analysis: PR, JB

Obtained funding: Not applicable

Overall responsibility: MF

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Submitted Dec 11, 2008; accepted Mar 6, 2009.