Combined endovascular and surgical approach for aortobronchial fistula

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Objective: The perioperative outcomes of the endovascular approach to aortobronchial fistula have been favorable. However, it is uncertain whether thoracic endovascular aneurysm repair (TEVAR) alone provides a complete and durable cure for an aortobronchial fistula. TEVAR does nothing to address the issue of the defect in the respiratory tract, leaving the patient at risk of aortobronchial fistula recurrence and/or stent graft infection. The authors believe that the bronchial defect should be addressed.

Methods: Over the last 10 years, 5 patients were treated for an aortobronchial fistula using a combined endovascular and surgical approach (primary treatment in 3 patients and secondary after TEVAR in 2 patients). All the patients underwent emergency stent graft placement and concomitant ($n = 1$) or staged ($n = 4$) open repair including pulmonary resection with coverage of the stent graft using muscle or pleural flaps. All patients received a 6-week course of broad-spectrum intravenous antibiotics followed by lifelong oral antibiotics.

Results: All patients survived the surgical procedure. After a mean follow-up of 23.2 months, 4 patients are asymptomatic and postprocedure computed tomography scans were unremarkable. One patient treated for an aortobronchial fistula after TEVAR was readmitted 4 months after surgical conversion. Stent graft explantation and silver-coated tube graft replacement of the descending thoracic aorta were performed for severe mediastinitis with associated thoracic stent graft infection. The postoperative course of this patient was uneventful.

Conclusions: Emergency TEVAR for an aortobronchial fistula is an appealing strategy for this devastating complication. However, to achieve a lasting result, direct contact between the stent graft and the pulmonary tissue should be avoided to prevent further erosive damage. Concomitant or staged repair should entail primary repair or resection and anastomosis of the bronchus and/or pulmonary resection with coverage of the stent graft using muscle or pleural flaps combined with broad-spectrum intravenous antibiotic therapy. Long-term surveillance and continued investigation are warranted. (*J Thorac Cardiovasc Surg* 2014;148:2108-11)

Aortobronchial fistula is a rare clinical entity. A literature review published in 2013 identified only 124 patients.¹ Untreated primary and secondary aortobronchial fistulae are fatal and remain a formidable surgical problem in older high-risk patients with hemorrhagic shock or sepsis. Despite significant improvement in surgical techniques, the operative mortality of open aortobronchial fistula repair ranges from 15% to 41%.² The high mortality is related to the need for thoracotomy, thoracic aortic crossclamping, and the surgical replacement or repair of the thoracic aorta with concomitant resection of the pulmonary segments involved. Moreover, redo operations, performed for secondary fistulas, are associated with increased bleeding

and operative time, with subsequent increased surgical morbidity and mortality.

Within the last decade, several case reports have described initial success with treatment of aortobronchial fistulas using thoracic stent grafts, suggesting more widespread use of this approach. The perioperative outcomes of the endovascular approach have been favorable with low 30-day mortality (6.4%) and no cases of paraplegia.¹ Concerns remain, however, about the risk of aortobronchial fistula recurrence and/or infection of the stent graft with the endovascular approach.² We have recently performed a systematic review of the literature¹ and reported that recurrence of the aortobronchial fistula and stent graft infection were observed in 11.2% and 1.7% of patients, respectively. Surgical conversion was required in 4.3% of patients. To decrease the risk of recurrence, we believe that the bronchial defect should be addressed. Direct contact between the stent graft and pulmonary tissues should be avoided to prevent further erosive damage. The concomitant repair may entail primary repair or resection and anastomosis of the bronchus and/or pulmonary resection with coverage of the stent graft using muscle or pleural flaps.

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Abbreviations and Acronyms

- CT = computed tomography
- TEVAR = thoracic endovascular aneurysm repair

The aim of this article was to present our experience of the combined endovascular and surgical approach for aortobronchial fistula.

METHODS

Patients treated for an aortobronchial fistula in our department between 2003 and 2013 were reviewed. Five patients were included. The clinical features, treatment modalities, and outcomes are summarized in Tables 1 and 2.

Primary Aortobronchial Fistula

Two women (aged 75 and 84 years) and 1 man (89 year old) underwent emergency thoracic endovascular aneurysm repair (TEVAR) for primary aortobronchial fistula. The 84-year-old woman had miliary tuberculosis. Two different stent grafts were used: Conformable-TAG (n = 2) (WL Gore & Associates, Inc, Flagstaff, Ariz) and Valiant stent graft (n = 1) (Medtronic Inc, Minneapolis, Minn). A concomitant left upper lobectomy was performed in 1 patient, and a left lower lobectomy or a pulmonary segmentectomy was performed as a staged procedure in the other 2 patients. For all patients, aortic debridement and coverage of the stent graft was achieved using pleural or pericardial flaps (Figure 1). Radical aortic debridement included resection of the necrotic tissues until the stent graft was visible. Two patients received a 6-week course of broad-spectrum intravenous antibiotics followed by lifelong oral antibiotic suppression. The patient who had miliary tuberculosis received prolonged (18 months) antituberculous drug therapy (isoniazid, rifampin, and ethambutol).

Aortobronchial Fistula After TEVAR

Two men (58 and 70 year old) underwent surgical conversion for secondary aortobronchial fistula after TEVAR. The initial indication for TEVAR was degenerative descending thoracic aortic aneurysm (n = 1), and dissecting descending thoracic aortic aneurysm (n = 1). The proximal landing zone was zone 3 in both patients. The patients had been treated using the Valiant and the Talent stent graft. These 2 patients had a chronic type I (distal) and type II endoleak.

Pulmonary segmentectomy was performed in both patients via a left thoracotomy and coverage of the stent graft was achieved using a pleural flap in 1 patient and the serratus muscle in the other (Figure 2).

Both patients received broad-spectrum intravenous antibiotics for 6 weeks followed by lifelong oral antibiotic suppression.

RESULTS

All patients survived the surgical procedure. Four patients had an uneventful postoperative course. After a mean follow-up of 23.2 months, these 4 patients are asymptomatic. Postprocedure computed tomography (CT) scans, white cell counts, and C-reactive protein levels were unremarkable. In the 2 individuals who underwent complementary follow-up with positron emission tomography scans, normalization of the metabolism around the stent graft was demonstrated (Figure 2).

One patient treated for an aortobronchial fistula after TEVAR was readmitted 4 months after surgical conversion for severe mediastinitis with thoracic stent graft infection. Via a redo left thoracotomy, on femoro-femoral cardiopulmonary bypass, the stent graft was explanted and the descending thoracic aneurysm was repaired using a silver-coated tube graft. The postoperative course was uneventful and after a follow-up of 6 months, the patient remains asymptomatic. The postprocedure CT scan at 18 months did not identify any abnormalities.

DISCUSSION

Aortobronchial fistula is a rare diagnosis. Conservative nonsurgical therapy results invariably in a fatal outcome as a result of massive hemoptysis or chronic mediastinitis. Causes of aortobronchial fistula formation include thoracic aortic aneurysms, advanced lung cancer, pulmonary infections, and open or endovascular repair of the thoracic aorta. With the increasing use of thoracic stent grafts, the number of patients treated for aortobronchial fistula after TEVAR has increased. The incidence of aortobronchial fistula after TEVAR is not negligible (1.7%), and is comparable to that after open repair.³ This warrants an ad hoc long-term follow-up after TEVAR, particularly in patients who have undergone emergency or complicated procedures. The postulated cause of aortobronchial fistula after TEVAR includes stent graft coverage of the bronchial arteries leading to ischemic necrosis of the bronchial wall, chronic endoleaks leading to erosion into the adjacent lung, and penetration of the stent graft through the aortic wall into the lung.⁴ In our 2 patients who experienced an aortobronchial fistula after TEVAR, chronic

TABLE 1. Clinical presentation at diagnosis

Patient	Gender	Age (years)	Hemoptysis	Fever >38°C or <36°C	Hyperleukocytosis	Respiratory rate >20/min	Heart rate >90/min	SIRS	ASA score	Bacteriology results
1	Female	75	Yes	Yes	Yes	Yes	Yes	Yes	4	0
2	Female	84	Yes	Yes	No	No	No	No	4	<i>Mycobacterium tuberculosis</i>
2	Male	89	Yes	Yes	Yes	Yes	Yes	Yes	4	0
4	Male	58	Yes	Yes	No	Yes	No	No	3	0
5	Male	70	Yes	Yes	Yes	Yes	Yes	Yes	3	0

SIRS, Systemic inflammatory response syndrome (2 or more of the following variables are present: temperature >38°C (100.4°F) or <36°C (96.8°F); heart rate >90 beats/min; respiratory rate >20 breaths/min or arterial carbon dioxide tension (Paco₂) <32 mm Hg; abnormal white blood cell count [>12,000/μL or <4000/μL]); ASA, American Society of Anesthesiologists.

TABLE 2. Demographic data and survey of patients treated by a combined endovascular and surgical approach for an aortobronchial fistula

Patient	Gender	Age (years)	Aortic pathology	Stent graft	Time to conversion	Conversion	Outcome	Follow-up (months)
1	Female	75	Primary aortobronchial fistula	C-Tag	0 d	Left upper lobectomy Aortic debridement Pleural flap	Discharged	43
2	Female	84	Primary aortobronchial fistula	Valiant	7 d	Left lower lobectomy Aortic debridement Pleural flap	Discharged	23
2	Male	89	Primary aortobronchial fistula	C-TAG	7 d	Left lower segmentectomy Aortic debridement Pericardial flap	Discharged	3
4	Male	58	Secondary aortobronchial fistula after TEVAR	Valiant	28 mo	Pulmonary segmentectomy Aortic debridement Pleural flap	Discharged	24
5	Male	70	Secondary aortobronchial fistula after TEVAR	Valiant	13 mo	Pulmonary segmentectomy Aortic debridement Serratus muscle flap	Stent graft infection Descending aortic replacement	14

TEVAR, Thoracic endovascular aneurysm repair.

endoleak was the most likely mechanism for the formation of aortobronchial fistula (these 2 patients had a chronic type I and type II endoleak). Characteristics such as the number of stent grafts used and the length of the aorta covered have not been found to be significant.³

Open repair involves combined replacement or bypass of the thoracic aorta with concomitant resection or repair of

the lung involved. Despite significant refinement in surgical techniques, the operative mortality of open aorto-esophageal fistula repair has been reported to be up to 41%.² Several alternative strategies have recently been reported in the literature. Since 1996, 124 patients treated by TEVAR for an aortobronchial fistula have been reported. The perioperative outcomes of the endovascular approach were

FIGURE 1. A patient treated for a primary aortobronchial fistula in a staged fashion. A, Intraoperative view. B, The stent graft (1) is exposed after lung mobilization. C, The lower left lung fistula was treated by pulmonary segmentectomy. Aortic debridement was performed and coverage of the stent graft was achieved using a pericardial flap (2). The pericardial defect was addressed with bovine pericardium (3).

FIGURE 2. Positron emission tomography scan of a patient treated for a primary aortobronchial fistula caused by pulmonary tuberculosis. A, 3 months after discharge: hypermetabolism of the thoracic stent graft, maximum standardized uptake value 10.1. B, 25 months after discharge: no hypermetabolism of the thoracic stent graft, maximum standardized uptake value 2.4.

favorable¹: low 30-day mortality (6.4%) and no cases of paraplegia. The incidence of severe complications (pulmonary, sepsis, myocardial ischemia) was only 11.3%. In this review, aortobronchial fistula recurred in 11.2% of the patients and surgical conversion was required in 4.3% of the patients. The evidence emerging from these results prompted us to change our clinical practice. However, it is uncertain whether TEVAR alone provides a complete and durable cure for aortobronchial fistula. TEVAR does nothing to address the issue of the defect in the respiratory tract, leaving the patients at risk of aortobronchial fistula recurrence and/or stent graft infection. We believe that the bronchial defect should be addressed. Direct contact between the stent graft and the pulmonary tissue should be avoided to prevent further erosive damage. The concomitant repair may entail primary repair or resection and anastomosis of the bronchus and/or pulmonary resection with coverage of the stent graft using muscle or pleural flaps. These patients need to be followed closely to evaluate the opportunity and the appropriate timing for a secondary surgical procedure. The treatment strategy and interval to additional surgical procedures are individualized according to the patient's physical condition and severity of sepsis. This aggressive surgical approach was efficient in 4 of the 5 patients. The patient who underwent surgical conversion was treated for a secondary aortobronchial fistula (after TEVAR). We can speculate that the risk of stent graft infection is probably higher when the patient has had a long-standing communication between the stent graft and the lung without broad-spectrum antibiotic therapy before the surgical approach to isolate the graft. Another surgical strategy is to consider TEVAR as a bridge therapy; in young and fit patients, TEVAR is followed by a delayed, durable, open repair when the patient has regained relative physiologic stability.⁵

Long-term antibiotic therapies must be considered. Postoperative antibiotics were administered in all our patients for 6 weeks. A conservative policy for antibiotic treatment might involve at least 4 weeks of periprocedural intravenous antibiotics followed by case-specific administration of oral suppressive antibiotics according to the clinical and laboratory parameters of infection.

The main limitation of the present study is that it is retrospective and the sample size is relatively small.

CONCLUSIONS

The perioperative outcomes of the endovascular approach for aortobronchial fistula have been favorable. TEVAR does nothing to address the issue of the defect in the respiratory tract, leaving the patients at risk of aortobronchial fistula recurrence and/or stent graft infection. We believe that the bronchial defect should be actively managed. The concomitant repair may entail primary repair or resection and anastomosis of the bronchus and/or pulmonary resection with coverage of the stent graft using muscle or pleural flaps. Long-term surveillance and continued investigation are warranted.

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