Thoracic Endovascular Aortic Repair for Aortobronchial Fistula

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The objective was to provide a systematic review of outcomes of thoracic endovascular aortic repair for aortobronchial fistula. A literature search identified 134 patients. The technical success rate was 93.2%. The overall 30-day mortality was 5.9%. After a mean follow-up of 17.4 months, the aortic-related mortality was 14.3%. Recurrence of the aortobronchial fistula was observed in 11.1% of the patients. Thoracic endovascular aortic repair of aortobronchial fistulas appears to be a viable alternative with excellent short-term results. Strict follow-up and aggressive adjunctive measures are needed to treat ongoing infection to prevent late related mortality.

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Primary and secondary aortobronchial fistulas are uniformly fatal if untreated and remain a formidable surgical problem in older, high-risk patients with hemorrhagic shock or sepsis. Despite advances in surgical technique, open repair still has a high operative mortality, which may reach 41% [1]. The high mortality is related to the need for thoracotomy, thoracic aortic cross clamping, and the surgical replacement or repair of the thoracic aorta with concomitant resection of involved pulmonary segments. Moreover, redo operations, which are required in secondary fistulas, worsen bleeding complication and prolonged operative time, leading to an increased surgical mortality and morbidity.

In the last decade, a number of case reports of successful initial treatment of aortobronchial fistulas using thoracic stent-grafts have been published, suggesting wider use of this approach. These reports often feature favorable short-term outcomes. Although endovascular stent-grafting is simpler, faster, and safer than surgery in unstable patients, the major concern is the durability of this approach. The stent-graft is in direct contact with a contaminated environment and therefore incurs a risk of fistula recurrence or stent-graft infection.

The aim of this article is to define the outcomes for thoracic endovascular aortic repair (TEVAR) of aortobronchial fistulas with a particular focus on midterm results.

Material and Methods

Search Strategy

A literature search was undertaken to identify all published studies in the past 10 years reporting thoracic endovascular aortic repair for aortobronchial fistula. Review methods were according to the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines. Candidate studies in English were sought through a computerized search of Embase, Medline, and Cochrane databases for the period of 1990 to September 2012. Key words entered in this search were “thoracic aorta” or “aortobronchial” or “aorto-bronchial” or “aortopulmonary” or “aorta-pulmonary” or “fistula” “aortotracheal” and “aortobronchopulmonary.” Articles were limited to those published in the English language. Additionally, manual evaluation of the reference lists of the retrieved articles and reviews on this area subject was undertaken.

Study Selection

Studies were considered for inclusion on the basis of these criteria: (1) reporting on thoracic endovascular aortic repair for management of aortobronchial fistula; and (2) reporting on clinical outcome. Studies containing duplicate data were excluded, and the manuscripts with the most recent or the best-documented material from the same authors were used for analysis. Articles were selected for further review and inclusion in the final analysis if they described individual outcomes for patients treated for aortobronchial fistulas.

Data Extraction

Data were extracted regarding age and sex; cause of fistula; presence or history of thoracic aortic surgery; time between previous aortic intervention and presentation of fistula; comorbidity; symptoms of the fistula; time between diagnosis and TEVAR; proximal landing zone; technical success of TEVAR defined by successful exclusion of the fistula during the initial endovascular procedure; treatment with antibiotic drugs and duration of this
treatment; in-hospital and long-term follow-up outcomes, including graft-related complications (endovascular leak, stent-graft migration); nongraft-related complications (sepsis, pneumonia, spinal cord ischemia, renal failure); early and late open and endovascular reinterventions; fistula recurrence; fistula-related mortality; and length of follow-up.

Results

Search Results

Forty-six articles [2-47] were integrated after a literature search identified 134 patients treated by TEVAR for aortobronchial fistula (Table 1).

Case Selection

Patient demographics, presenting features, and comorbidities are shown in Table 1. The mean age was 64.4 years and 76.3% of patients were male. Most patients presented with hemoptysis (93.7%). Most reported cases (55%) occurred in patients who had previously undergone thoracic aortic surgery.

Table 1. Case Selection, Clinical Presentation, Causes of Aortobronchial Fistulae, Surgical Management, and Outcomes

<table>
<thead>
<tr>
<th>Variables</th>
<th>(n = 134)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean age, years</td>
<td>64.4 ± 12.1</td>
</tr>
<tr>
<td>Male, %</td>
<td>76.3</td>
</tr>
<tr>
<td>Initial presentation</td>
<td></td>
</tr>
<tr>
<td>Hemoptysis</td>
<td>93.7% (104/111)</td>
</tr>
<tr>
<td>Hypovolemic shock</td>
<td>10.8% (12/111)</td>
</tr>
<tr>
<td>Systemic infection</td>
<td>12.3% (14/113)</td>
</tr>
<tr>
<td>Dyspnea</td>
<td>13% (13/100)</td>
</tr>
<tr>
<td>Thoracic pain</td>
<td>10.9% (12/110)</td>
</tr>
<tr>
<td>Cause of aortobronchial fistula</td>
<td></td>
</tr>
<tr>
<td>Previous thoracic aortic surgery</td>
<td>55% (71/129)</td>
</tr>
<tr>
<td>Thoracic aortic aneurysm</td>
<td>15.1% (20/132)</td>
</tr>
<tr>
<td>Previous TEVAR</td>
<td>2.3% (3/131)</td>
</tr>
<tr>
<td>Dissection</td>
<td>2.3% (3/131)</td>
</tr>
<tr>
<td>Penetrating ulcer</td>
<td>12.2% (16/131)</td>
</tr>
<tr>
<td>Mycotic</td>
<td>4.5% (6/131)</td>
</tr>
<tr>
<td>False aneurysm</td>
<td>34% (44/129)</td>
</tr>
<tr>
<td>TEVAR within 24 hours of diagnosis</td>
<td>84.8% (101/119)</td>
</tr>
<tr>
<td>Zone</td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>5.1% (5/97)</td>
</tr>
<tr>
<td>2</td>
<td>27.8% (27/97)</td>
</tr>
<tr>
<td>3</td>
<td>40.2% (36/97)</td>
</tr>
<tr>
<td>4</td>
<td>15.4% (15/97)</td>
</tr>
<tr>
<td>Antibiotic therapy &gt;4 weeks</td>
<td>54.4% (55/101)</td>
</tr>
<tr>
<td>Technical success</td>
<td>93.2% (125/134)</td>
</tr>
<tr>
<td>Additional procedure</td>
<td></td>
</tr>
<tr>
<td>Intercostal muscle flap</td>
<td>0.7% (1/134)</td>
</tr>
<tr>
<td>Pulmonary resection</td>
<td>2.1% (3/134)</td>
</tr>
</tbody>
</table>

(Continued)
**Midterm Outcomes**

Midterm outcomes were defined as those occurring after 30 days. After a mean follow-up of 17.4 months (range, 1 to 45), the all-cause mortality rate was 21.4% (27 of 126) and the aortic related mortality was 14.3% (18 of 126). Death was due to the recurrence of the aortobronchial fistula in 7 patients and to stent-graft infection in 3 patients. Recurrence of the aortobronchial fistula was observed in 14 patients (11.1%). Surgical conversion during the postoperative course with thoracic stent-graft explantation was required in 5 patients. Additional stent-graft placement was required in 7 patients (5.2%).

**Comment**

Aortobronchial fistula is a rarely encountered clinical entity. A literature review published in 2002 revealed only 76 patients reported through 2002 [1]. If left untreated, aortobronchial fistulas are uniformly fatal [48]. Until 1960, mycotic aneurysms were the leading cause of aortobronchial fistula. Currently, the most common causes are postsurgical and degenerative aneurysms. In 73% of cases, a false aneurysm was found at the fistula [1].

Hemoptysis is the first and often the only symptom of the fistula. Although hemoptysis is not a specific symptom of aortobronchial fistula, the condition must be strongly suspected when hemoptysis occurs in a patient with a history of thoracic aortic surgery or thoracic aortic aneurysm. Computed tomography often reveals a false aneurysm, aortic aneurysm, lung parenchyma consolidation, and compression of the bronchial tree. However, it is quite unlikely to yield a scan that directly visualizes the passage of contrast from the aorta into the airways. Normal radiographic studies do not reliably exclude a diagnosis of aortobronchial fistula. Bronchoscopy may demonstrate blood in the affected lobe as it may visualize the bronchial tear. However, dislodgment of occluding clots is a potential serious complication of bronchoscopy. Some investigators recommend that bronchoscopy should be performed only if immediate repair is available.

Traditional, open repair has been the gold standard for more than 40 years. This conventional approach combined replacement or bypass of the thoracic aorta with concomitant resection or repair of involved pulmonary segments. Despite significant improvement in surgical techniques, the operative mortality of open aortobronchial fistula repair ranges from 15% to 41% [1]. The mortality and morbidity of open repair is multifactorial: the emergent nature of repair, the access to the aorta, which is particularly demanding because of dense adhesions, and mediastinitis with a high risk of adjacent organ lesions and significant blood loss and the need for thoracic aortic cross-clamping. As a consequence, a less invasive approach would be even more favorable to patients.

In 1996, Chuter and coworkers [2] reported the successful treatment of aortobronchial fistula with a thoracic stent-graft. In 2009 Jonker and associates [49] carried out a systematic review looking at the outcomes of TEVAR for aortobronchial and aortoesophageal fistulas, reporting 70 cases. Since then, a further 63 cases have been reported in the literature. The significant increase in publications reporting aortobronchial fistula repair with TEVAR highlights the paradigm shift in the emergent management of this disease.

The perioperative outcomes of the endovascular approach have demonstrated favorable outcomes: a low 30-day mortality rate (5.9%) and no cases of paraplegia. The incidence of severe complications (pulmonary complication, sepsis, myocardial ischemia) was only 10.4%. Surgical conversion during the postoperative course with thoracic stent-graft explantation was required in 2 patients. Thoracic endovascular aortic repair of aortobronchial fistulas seems to be a viable alternative to conventional open repair, with excellent short-term results in comparison with the results of open surgery reported in the literature. Concerns about the risk of recurrence of the aortobronchial fistula or of infection of the stent-graft after endovascular repair have been raised [36, 40]. As TEVAR does nothing to address the issue of the defect in the respiratory tract, it leaves the patients at risk of aortobronchial fistula recurrence or stent-graft infection. In this review, the recurrence of the aortobronchial fistula occurred in 11.1% of the patients and surgical conversion was required in 3.5% of the patients. TEVAR can be considered as an effective midterm treatment for 88.9% of the patients. Several options have been proposed to decrease the rate of aortobronchial recurrence. De Rango and associates [36] suggest that TEVAR should be considered as a bridge therapy followed by a delayed durable open repair when the patient has achieved stability. Chiesa and associates [50] reported 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 or pulmonary resection with coverage of the stent-graft using muscle or pleural flaps. In their experience, the mortality rate was lower in cases that involved a combined approach, as compared with those that involved TEVAR alone.

The necessity for long-term antibiotic therapies has to be considered. Postoperative antibiotic regimen for more than 4 weeks was administered to only 54.4% of the patients. Postoperative antibiotic treatment differs completely among the various investigators, both with regard to the antibiotic and the duration of therapy. Antibiotic treatment should be applied after endovascular stent placement to potentially prevent thoracic stent-graft infection. The minimum time to allow for the beneficial effect of antibiotics is 3 to 7 days [51]. Some physicians insist that life-long treatment with oral suppressive antibiotics is required, whereas others claim that such treatment can be discontinued provided there is no clinical, bacteriologic, or radiologic evidence of ongoing sepsis [52, 53]. A conservative policy for antibiotic treatment might involve at least 4 weeks of perioperative intravenous antibiotics followed by case-specific administration of oral suppressive antibiotics according to clinical and laboratory parameters of infection. Clearly, any strategy in this
patient population needs to be highly individualized given the risks of major thoracic aortic surgery often in scarred operative field. There is a need for close observation in follow-up and probably long-term antibiotic therapy.

Analysis of these results enables us to draw some conclusions regarding the management of this uncommon but devastating complication. Firstly, the low perioperative mortality associated with TEVAR means it should be considered as a first-line bailout procedure for aortobronchial fistula. Furthermore, to avoid stent-graft infection, antibiotic therapy should consist of at least 4 weeks of postoperative intravenous antibiotics. Once the patient has fully recovered from the acute insult, further management strategies should be evaluated based on the clinical status and comorbidities of the patient.

After a careful preoperative assessment, for young and medically fit patients, delayed durable open surgical repair including stent-graft removal should be considered. Typically this will involve in situ revascularization using an allograft or a rifampicin or silver-bonded artificial graft with primary repair or resection and anastomosis of the bronchus or pulmonary resection with coverage of the graft. For patients with significant comorbidities, to prevent the risk of fistula recurrence and stent-graft infection, surgical conversion leaving the stent-graft in place and addressing the bronchial defect alone should be considered. For medically unfit patients, TEVAR alone is an effective midterm treatment.

Regardless of the surgical therapy employed, there is a need for close observational follow-up and long-term antibiotic therapy. Serial blood test analyses using biomarkers such as C-reactive protein and white blood cell count are effective at monitoring for signs of infection. Computed tomography (CT) is highly effective at looking for graft infection—the high spatial resolution offered by CT provides very sensitive evaluation of anatomic changes in perigraft areas. However, hematomas and seromas around grafts often appear similar to abscesses, thus making it difficult to distinguish between noninfected and infected prosthetic grafts on CT scans. If perigraft infection is being considered, 18-fluoro-deoxyglucose positron emission tomography (FDG-PET)/CT is useful for detecting high uptake around grafts and excluding other causes of inflammation. Repeated use of functional imaging of tissues can be a very helpful tool to direct the duration of the antibiotic therapy or the need for surgical conversion. We believe that a patient-tailored treatment is more efficient, and the surgical and antibiotic strategy should be reviewed according to the clinical status of the patients and to their individual postoperative clinical, biological, and functional FDG-PET/CT evaluation.

Although the present study reported perioperative and midterm outcomes of TEVAR for aortobronchial fistulas, it has several limitations. Data were obtained from case reports and case series with likely publication bias. A further bias was that favorable outcomes are more likely to be reported than unfavorable ones. That could partially explain the excellent outcomes reported.

In conclusion, thoracic endovascular aortic repair of aortobronchial fistulas seems to be a viable alternative to conventional open repair with excellent short-term results. Strict follow-up and aggressive adjunctive measures are needed to treat ongoing infection to prevent late related mortality. Further large cohort and long-term follow-up studies are required to define the outcome of this therapy.

References