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CASE REPORT

CLINICAL CASE

Innominate Artery Pseudoaneurysm Requiring Cartilage Tracheal Repair in a Child

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ABSTRACT

Mycotic aneurysms are rare but potentially catastrophic. We report a case of an innominate artery pseudoaneurysm in a 4-year-old patient that caused a tracheoinnominate fistula requiring tracheoplasty with a costal cartilage graft and a homograft iliac artery replacement of the diseased innominate artery, with a successful outcome. (J Am Coll Cardiol Case Rep 2024;29:102329) © 2024 The Authors. Published by Elsevier on behalf of the American College of Cardiology Foundation. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

HISTORY OF PRESENTATION

A 4-year-old child was presented to a regional hospital with a 7-day history of fever, lethargy, post-tussive vomiting, and anorexia. She received a diagnosis of suppurative otitis media and was treated with intravenous antibiotics. An ear swab showed growth of *Staphylococcus aureus*, scant *Streptococcus pyogenes,* and gram-negative rods. Blood culture results were positive for group A *Streptococcus*. After an episode of hypoxia and respiratory deterioration, she was intubated and received a diagnosis of bacterial tracheitis because purulent discharge was identified on the vocal cords. A suspicious chest radiograph with a widened mediastinum led to a computed tomography (CT) scan, which showed a pseudoaneurysm of the innominate artery causing tracheal compression. She was transferred to our tertiary hospital for specialist management.

LEARNING OBJECTIVES

- To recognize a rare but potentially fatal complication of streptococcal bacteremia.
- To demonstrate the advantage of a multidisciplinary team in the management of unexpected intraoperative findings to obtain a successful outcome.

PAST MEDICAL HISTORY

The patient had a history of mild asthma and was not taking any regular medications.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis included an infective, traumatic, or iatrogenic pseudoaneurysm and nonvascular mediastinal masses.

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ABBREVIATIONS AND ACRONYMS

CT = computed tomography

ENT = ear, nose, and throat

LBO = laryngobronchoesophagoscopy

MA = mycotic aneurysm

SCM = sternocleidomastoid

SVC = superior vena cava

TTE = transthoracic echocardiography

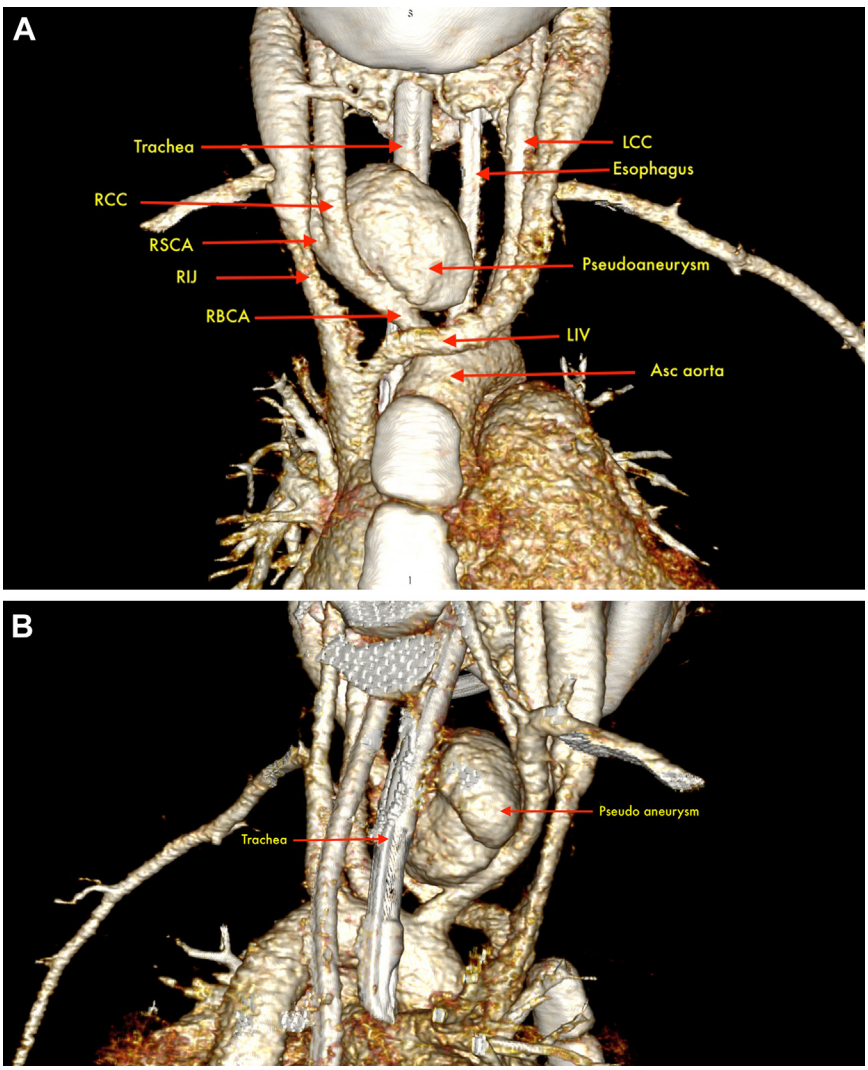
INVESTIGATIONS

A repeat CT scan with intravenous contrast enhancement revealed an innominate artery pseudoaneurysm with a surrounding hematoma measuring 32 mm × 19 mm × 30 mm and 10 mm at its neck. It extended from right to left in front of the trachea, was closely related to it (**Figures 1A and 1B and 2A and 2B** and Video 1), and was causing compression of the superior vena cava and both brachiocephalic veins. There was no extravasation of contrast medium and no evidence of fistulous communication at the time. Thoracic echocardiography (TTE) showed no evidence of infective endocarditis.

MANAGEMENT

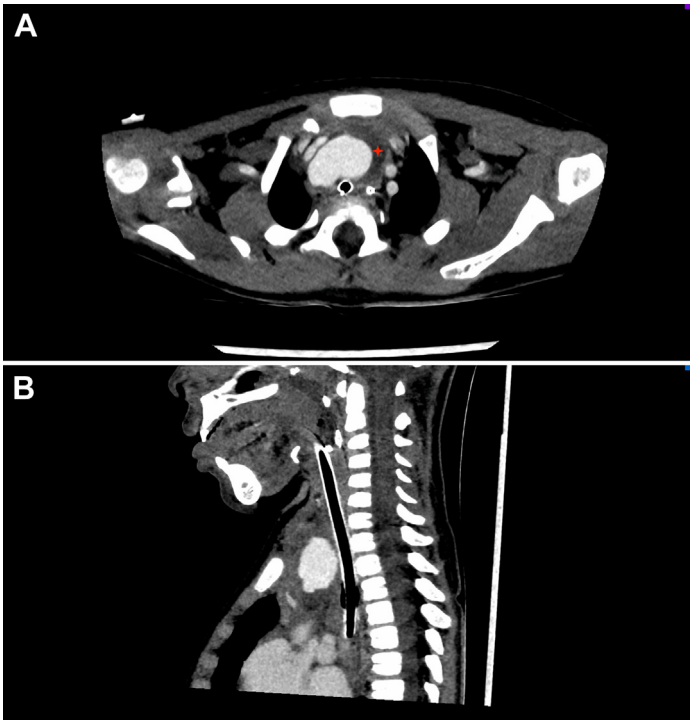
Given the high risk of rupture, the patient underwent surgical repair through a median sternotomy and with

FIGURE 1 Large Innominate Artery Pseudoaneurysm



(A) A 3-dimensional anterior view of a large innominate artery pseudoaneurysm in front of the trachea. (B) A 3-dimensional posterior view showing the close relationship of the pseudoaneurysm with the trachea. Asc = ascending; LCC = left common carotid artery; LIV = left innominate vein; RBCA = right brachiocephalic artery; RCC = right common carotid artery; RIJ = right internal jugular vein; RSCA = right subclavian artery.

FIGURE 2 **Mycotic Aneurysm of the Innominate Artery**

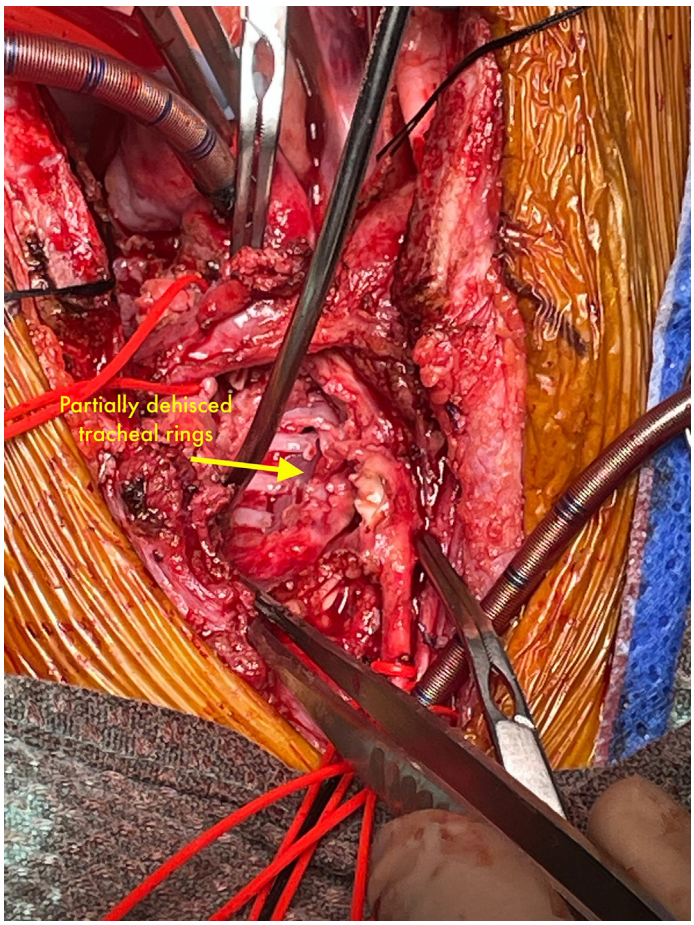


(A) Axial view of the innominate mycotic aneurysm anterior to the trachea. The surrounding hematoma is also noted (red star). (B) Sagittal view showing displacement of the trachea with the endotracheal tube posterior to the pseudoaneurysm.

cardiopulmonary bypass with a period of circulatory arrest at 20 °C. Intraoperatively, after entering the pseudoaneurysm, a large defect (4 cm in length) was identified on the posterior wall. After the infected tissue was debrided, the endotracheal tube became visible, with 3 tracheal rings partially dehisced anteriorly (**Figure 3**). After an intraoperative multidisciplinary discussion with anesthesia and ear, nose, and throat (ENT) specialists, the decision was made to harvest costal cartilage from the right chest wall and create a graft for repair of the anterior wall of the trachea. The composite perichondrial-cartilage graft was shaped into a keel, precisely secured to the anterior wall of trachea by using multiple interrupted polypropylene (Prolene, Ethicon) sutures and parachute down onto the trachea with confirmed no air leak (**Figures 4 and 5**). The medial one-half of the sternocleidomastoid (SCM) muscle was transected superiorly and used as a pedicle sutured over the tracheal graft to provide vascular supply to the tracheal repair site and tissue bulk between the trachea repair and the next step of arterial graft. The innominate artery stump was oversewn, and an iliac artery homograft was used to create a distal ascending aorta-to-right common carotid artery bypass (**Figure 6**).

She was extubated on postoperative day 9 and completed a 4-week course of antibiotics. Serial laryngobronchoesophagoscopies (LBOs) showed some granulation tissue at the graft site that was left untouched and was allowed to heal by secondary intention with a patent tracheal lumen and no significant tracheomalacia (**Figure 7**).

FIGURE 3 Tracheal Defect



The trachea had dehisced anteriorly, creating a large defect.

DISCUSSION

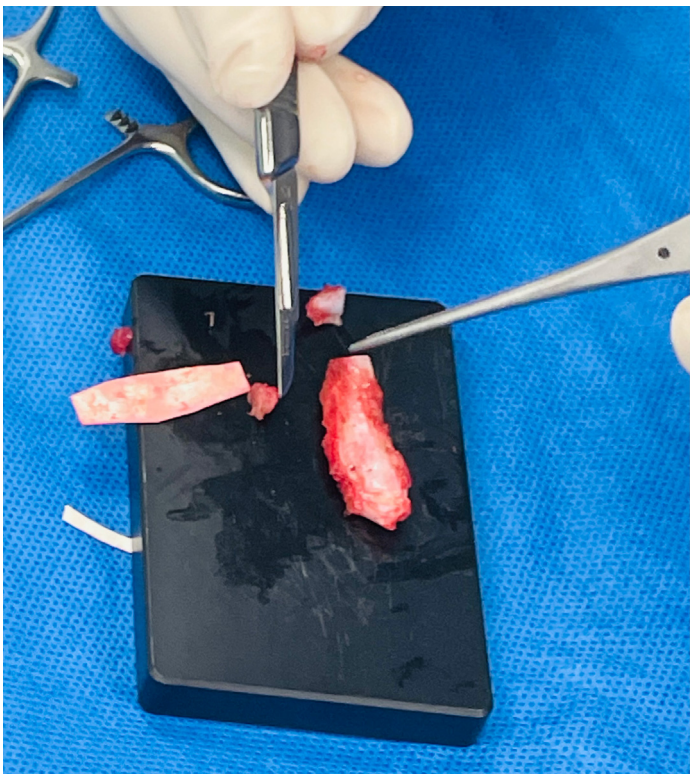
Mycotic aneurysms (MAs) are rare in childhood, especially in a previously undiseased aorta.[1] They can develop by direct bacterial invasion of the arterial wall or from distant hematogenous spread.

In the preantibiotic era, group A *Streptococcus* was the most common bacterial pathogen identified causing bacterial tracheitis. Following the development of effective antimicrobial agents, *Staphylococcus aureus* became the most common causative organism, and more recently, *Moraxella catarrhalis* has been identified with increased frequency.

The most frequent complication associated with the acute phase of illness is pneumonia. Other complications include acute respiratory distress syndrome, septic shock, toxic shock syndrome, pulmonary edema, pneumothorax, and rarely, cardiorespiratory arrest.[2]

Group A *Streptococcus* (*S pyogenes*), a virulent gram-positive organism, causes a wide spectrum of diseases, including pharyngitis, skin and soft tissue infections, bacteremia, and streptococcal toxic shock syndrome.[3,4] Mas caused by group A *Streptococcus* are extremely rare. Most of those MAs described in the literature are aortic, but they can also be peripheral,

FIGURE 4 Costal Cartilage Graft



The cartilage graft is shaped into a keel.

produce acute aneurysmal symptoms, and be associated with either concurrent bacteremia or pharyngitis.[5] The morbidity is low, but rupture is the most serious life-threatening complication and has been reported to occur in 50% to 85% of cases.[6,7]

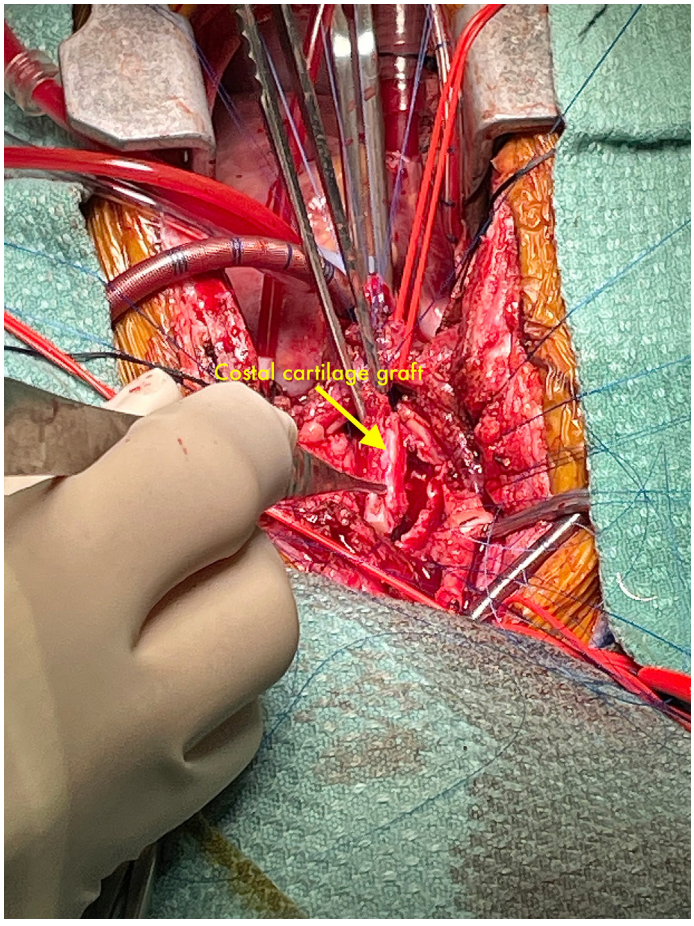
Additionally, aortobronchial fistula is a rare complication, with fistulous communication between the thoracic aorta (or branches) and the adjacent tracheobronchial system. If it is untreated, the mortality rate is virtually 100%. In this case, it was not possible to ascertain whether the pseudoaneurysm invaded the tracheal wall or the bacterial tracheitis perforated the anterior tracheal wall and subsequently created an inflammatory pseudoaneurysm. The preoperative imaging did not raise the suspicion of a communication between the pseudoaneurysm and the trachea. If there had been a high index of suspicion, preoperative bronchoscopy could have alerted us to the fistula.

The diagnosis of MA of the aorta is often difficult. In the acute stage, sepsis usually dominates the clinical picture. TTE is helpful depending on the location and to assess any associated abnormalities, but a CT angiogram is diagnostic. The incidence of negative blood culture results ranges from 25% to 37%, and that of negative tissue culture results ranges from 22% to 50%.[8] Even though we isolated group A *Streptococcus* from the initial blood cultures, we did not culture this organism on the pseudoaneurysm wall or other intraoperative specimens.

The choice and timing of an optimal procedure depend on several factors. These include impending rupture, the presence of a fistula, a lack of response to antibiotics, single or multiple MAs, pus surrounding the MA or adjacent vertebral infection, and patients' comorbidities.[7]

Extracorporeal circulatory support is of great help. Although we had wide exposure with a sternotomy

FIGURE 5 Parachute Technique



Graft is parachuted down to the trachea.

incision extending into the right side of the neck along the anterior line of the SCM muscle, the full extent of the damage to the innominate artery and the erosion of the anterior tracheal wall were not evident until we opened the pseudoaneurysm space under circulatory arrest conditions.

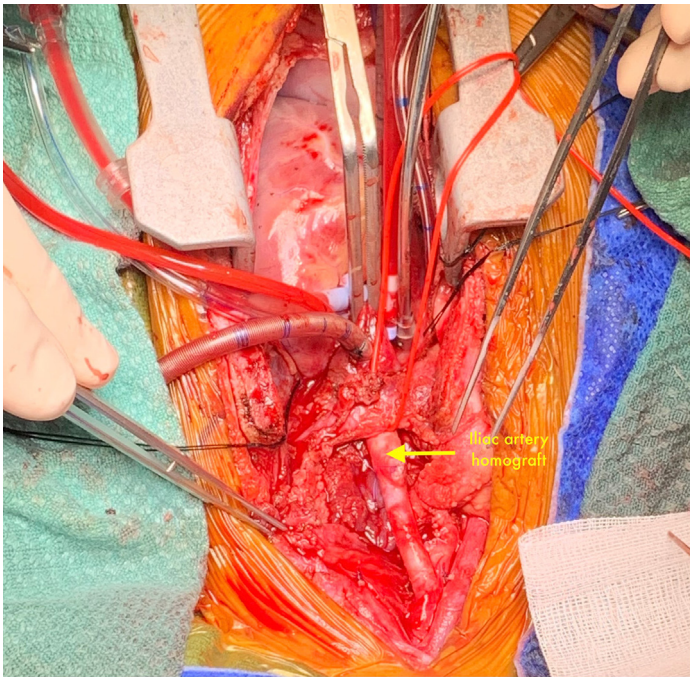
A ruptured pseudoaneurysm with hemodynamic instability represents a surgical emergency. A patient's condition may be too unstable for open surgery. In these extreme cases, endovascular treatment may be considered to stabilize the lesion and the hemodynamics, with definitive surgical repair at a later stage.[9]

The surgical management of pseudoaneurysms includes meticulous local debridement of all infected tissue and bypass of the infected arterial segment. Active infection precludes the use of prosthetic material to repair the aneurysm, and extra-anatomical bypass may be necessary as in this case, with the use of a homograft. Polyester grafts are often coated in rifampin or are silver impregnated, but the use of autologous venous grafts or cryopreserved arterial allografts reduces the risk of reinfection. The use of an SCM muscle flap over the costal cartilage graft brings in a reliable vascular supply, eliminates dead space, and protects the vessels from the infected graft site.

FOLLOW-UP

The patient is scheduled for follow-up with an ENT specialist, and final-look LBO is also scheduled.

FIGURE 6 Homograft Iliac Artery



A distal ascending aorta–to–right common carotid artery bypass is shown.

CONCLUSIONS

Upper respiratory infections associated with a vascular mediastinal mass should raise the suspicion of infectious pseudoaneurysm of the aorta and branches. Preoperative imaging and careful surgical planning are paramount to offer a good result. Multidisciplinary management bringing in reliable experience is vital when unexpected complex cases such as this arise.

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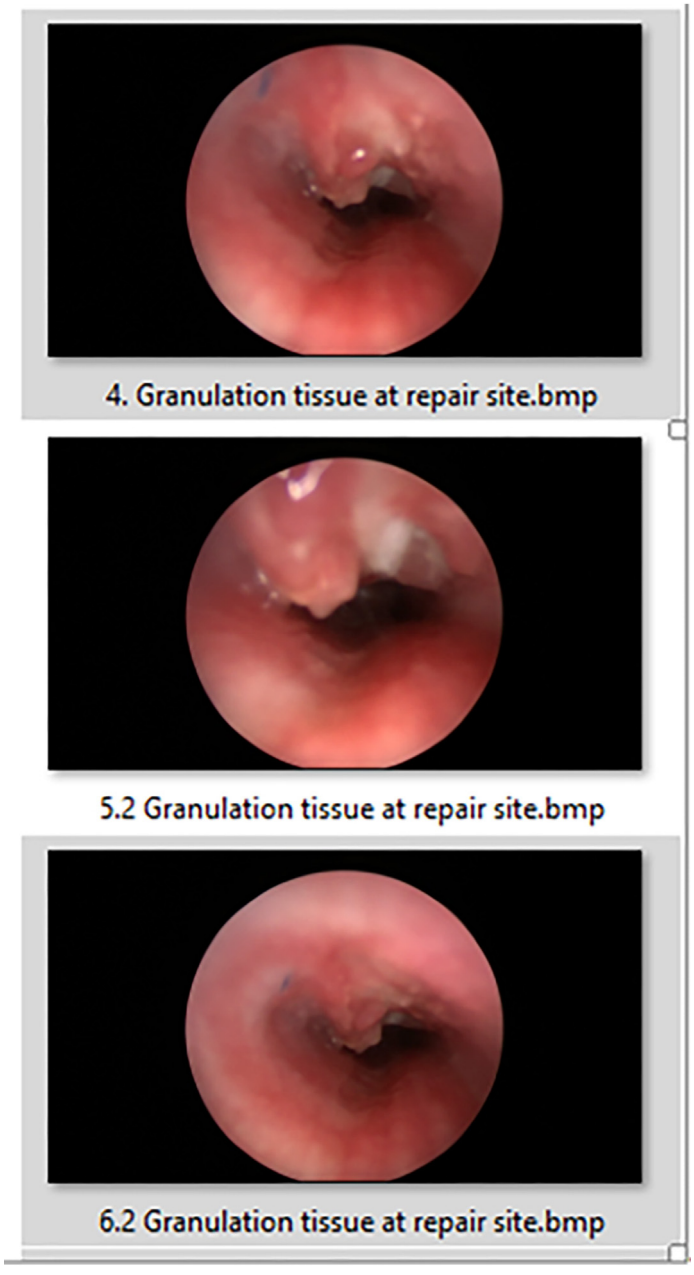
FIGURE 7 **Laryngobronchoesophagoscopy**



Granulation tissue at the repair site and a patent trachea are shown.

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KEY WORDS mycotic aneurysm, pseudoaneurysm, tracheal fistula, tracheoplasty

APPENDIX For a supplemental video, please see the online version of this paper.