CASE REPORT

Successful management of tracheo-innominate artery fistula with endovascular stent graft repair

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Tracheo-innominate artery fistula is a highly lethal complication after tracheostomy. A 37-year-old man who had undergone a tracheostomy 14 years earlier because of dysphagia after brain surgery had a tracheo-innominate artery fistula with exsanguinating hemorrhage from his tracheostomy site. After temporary control of the bleeding, a stent graft was implanted in the innominate artery through the brachial artery. The patient recovered uneventfully and remained well 14 months after the procedure, with no sign of infection. Endovascular stent grafting may be the treatment of choice for patients with tracheo-innominate artery fistula. (J Vasc Surg 2001;33:1280-2.)

Tracheo-innominate artery fistula (TIF) is a rare but serious complication after tracheostomy.[1-3] This condition is inevitably fatal unless treated surgically.[3] However, the morbidity and mortality rates are high even after surgical treatment, because of the difficulty controlling hemorrhage in the perioperative period.[3,4]

Recently, endovascular stent grafting has been developed, and its effectiveness in many vascular diseases has been recognized.[5,6] Because of the serious condition of a patient with TIF, this minimally invasive technique may be the treatment of choice. We successfully treated a case of tracheo-innominate artery fistula by means of endovascular stent grafting. This is the first report of endovascular repair of TIF in the literature.

CASE REPORT

A 37-year-old man was transferred from another hospital on August 28, 1999, because of a sudden and massive (2 L) arterial hemorrhage from his tracheostomy site. He had undergone brain surgery and adjuvant radiation therapy for medullobastoma of the cerebellum in 1985 and subsequently had dysphagia that necessitated a tracheostomy. He had no other diseases and had managed his tracheostomy by himself, with routine exchange of tracheostomy tubes at the referring hospital. On admission, he was still severely hypotensive after resuscitation from respiratory arrest and underwent a transfusion of 5 units of red blood cells immediately. Temporary control of the bleeding was achieved by over-

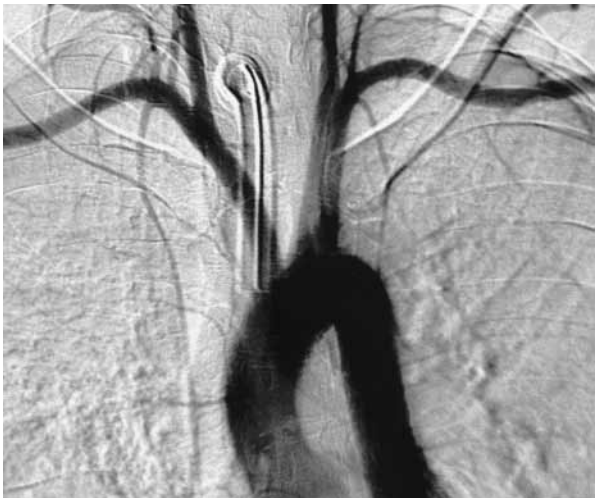

Fig 1. Angiogram on admission showing innominate artery crossing trachea at site of the cuff of the tube. No aneurysms or vascular anomalies were demonstrated.

inflating the cuff of the tracheostomy tube. The innominate artery was shown by means of angiography on admission to cross the trachea at the site of the cuff of the tube (Fig 1). The diseased trachea, associated with severe paratracheal inflammation and hematoma in the mediastinum, was demonstrated by means of computed tomography to be closely adjacent to the innominate artery (Fig 2, *A*).

Because a traditional surgical approach was considered to be difficult because of severe adhesions around the fistula and the patient's serious condition, endovascular treatment was performed with the patient under local anesthesia after obtaining consent from his family. After the right brachial artery was exposed, a 12F sheath was carefully placed into the artery. After

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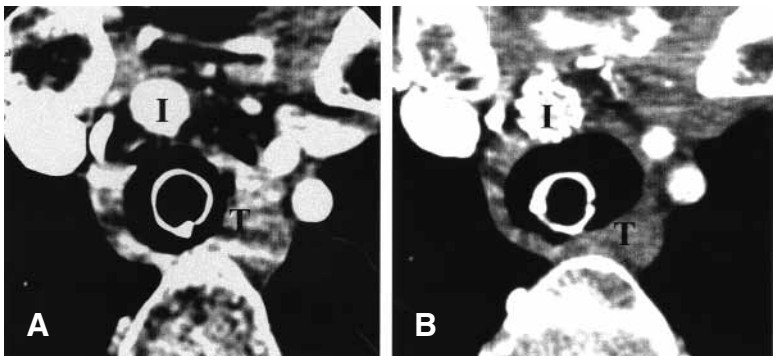

Fig 2. A, Computed tomogram on admission demonstrating markedly diseased trachea associated with hematoma in mediastinum. Note that trachea is closely adjacent to innominate artery, with severe paratracheal inflammation. **B,** Computed tomogram 10 months after procedure demonstrating improvement of paratracheal inflammatory changes, including disappearance of hematoma, without mediastinitis. *T,* Trachea; *I,* innominate artery.

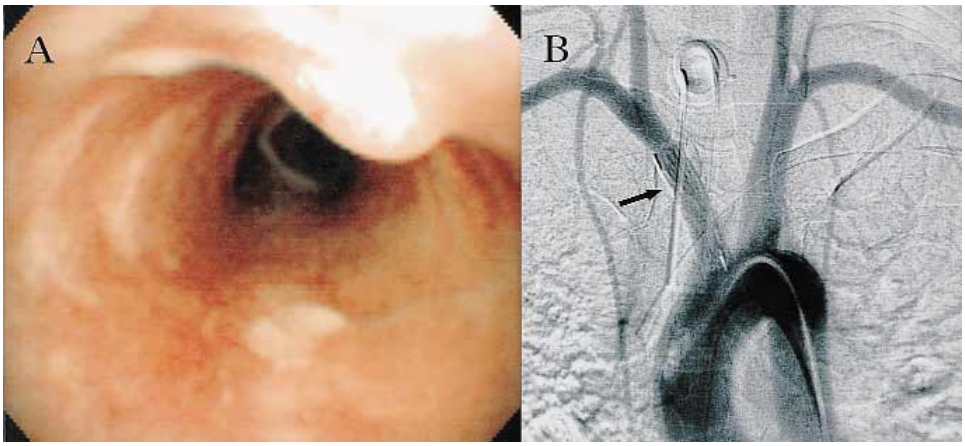

Fig 3. Bronchoscopy and angiography 8 weeks after the procedure. **A,** Bronchoscopic view showing mucous membrane covering inner surface of trachea. **B,** Angiogram demonstrating that stent was in satisfactory position. *Arrow,* Stent graft.

arteriographic evaluation, a self-expanding stent graft "Passager" (Meadox, Oakland, Calif), 10 mm in diameter and 60 mm in length, covered with a thin-wall of woven polyester fiber (Dacron) graft, was introduced through the sheath. The stent graft was implanted along the whole length of the innominate artery, maintaining sufficient blood flow to the right arm and the brain. Satisfactory alignment of the stent was confirmed, and hemostasis was obtained thereafter without inflating the cuff. Because *Staphylococcus aureus* and *Pseudomonas aeruginosa* were demonstrated in sputum cultures, cefazolin sodium and piperacillin sodium were administrated intravenously for 4 weeks to prevent graft infection. Sputum cultures subsequently were negative for *S aureus* and *P aeruginosa*. Blood cultures were all negative for the existence of microorganism during his hospital stay. The mucous membrane was shown by means of bronchoscopy to cover the inner surface of the trachea, and the stent was shown by means of angiography to be in a satisfactory position; no perigraft leakage was shown 8 weeks after the procedure (Fig 3). He recovered uneventfully and was discharged from the hospital. Thereafter, his tracheostomy was managed so that his trachea wall was not damaged by means of overinflation of the cuff. Improvement of the paratracheal inflammatory changes without mediastinitis was demonstrated by means of computed tomography (Fig 2, *B*). Fourteen months after the procedure, the patient has no symptoms related to the tracheostomy, and there are no signs of graft infection.

DISCUSSION

Since Korte[7] first reported a rupture of the innominate artery after tracheostomy in a 5-year-old with diphtheria in 1879, TIF has been recognized to be a potentially fatal complication of tracheostomy.[7] TIF has been reported to occur in 0.6% of patients after tracheostomy.[1,4] Although the pathophysiologic mechanism of injury in TIF is still unclear, pressure necrosis of the tracheal wall caused by the tracheostomy tube is considered to lead to erosion of the innominate artery.[2] Several factors are known to predispose patients to fistula formation, including low positioning of tracheostomy stoma, high riding of the innominate artery, overinflation of the tra-

cheostomy cuff, and infection.[2,3] Both the lower positioned tracheostomy and inappropriate management of tracheostomy may have contributed to erosion of the tracheal wall and the innominate artery in our patient.

The successful management of TIF depends on prompt diagnosis, control of bleeding, and prevention of secondary infection.[2] Although Jones et al[1] reported that bronchoscopy and angiography rarely gave the correct diagnosis, these methods may have practical roles in establishing a diagnosis. Bronchoscopy on admission was not performed in our patient because of the possibility of triggering a re-bleed. It has been reported that all patients with moderate bleeding from the tracheostomy site should be assumed to have TIF.[2] Approximately 35% of all patients with TIF have "herald" bleeding, and the other 65% of patients first have massive hemorrhage.[2] Our patient experienced herald bleeding approximately 2 hours before admission to the referring hospital, followed by 2 L of exsanguinating arterial hemorrhage and respiratory arrest. Control of the bleeding by overinflating the cuff of the tracheostomy tube in our patient confirmed the diagnosis of TIF. We failed to demonstrate the fistula, but we could make the diagnose of TIF because of (1) 2 L of massive arterial hemorrhage after typical herald bleeding; (2) control of bleeding by overinflating the cuff; (3) obtaining hemostasis after stent grafting; and (4) lack of history of injury and other hemorrhagic disorders. A computed tomography image was of some diagnostic help.

After temporary hemostasis is achieved, immediate surgical management has been advocated.[4] Exclusion of the innominate artery and extra-anatomic bypass grafting, such as carotid-carotid, axillo-axillo, or femoroaxillo bypass grafting, might be the treatment of choice because it is a less dangerous option than in situ reconstruction. However, these surgical approaches are not always satisfactory, because massive hemorrhage caused by clot removal frequently occurs during exposure of the origin of the innominate artery.[2,3] Survival with traditional open surgical treatment is reported to be approximately 25%.[4] Furthermore, Gelman[2] reported that only 40 of 71 patients who survived after operation survived for more than 2 months. Since the first report by Parodi, the effectiveness of endovascular stent grafting has been recognized in many vascular diseases.[5,6,8] Indeed, there is a possibility of graft contamination from the trachea through the fistula in endovascular stent grafting,[9] but this technique may be the treatment of choice, at least for massive bleeding from TIF in patients without septicemia. We obtained a satisfactory result without graft infection 14 months after the procedure with intensive administration of antibiotics, partially because we avoided surgical dissection of the fistula, which would have inevitably contaminated the surgical field.

In conclusion, we present a patient with TIF who underwent endovascular stent grafting with a favorable early result. This technique is suggested to be the treatment of choice for patients with TIF, although additional follow-up is mandatory.

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