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TECHNICAL NOTE

Aortoesophageal fistula involving the central aortic arch salvaged with emergent percutaneous TEVAR, great vessel coverage and *in vivo* graft fenestration

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ABSTRACT
Immediate intervention is needed for aortoesophageal fistulas (AEF), a rare but highly lethal cause of massive gastrointestinal hemorrhage. Emergent thoracic endovascular aortic repair (TEVAR) is considered first-line treatment for massive bleeding from AEFs. We describe an unusual and challenging case of TEVAR coverage of an AEF involving the central aortic arch immediately followed by *in vivo* endograft fenestration to regain arch vessel perfusion. *In vivo* fenestration, currently a procedure for emergency or investigational purposes only, was shown to be life saving in our case. The main complications associated with the procedure included stroke and infection, requiring esophagectomy and cervical diversion as well as ongoing antibiotic treatment.

Fistulas between the thoracic aorta and the esophagus are a rare cause of massive gastrointestinal hemorrhage that is almost always fatal without immediate intervention. Aortoesophageal fistulas (AEFs) are associated with thoracic trauma, aortic aneurysms, ruptured penetrating aortic ulcers, esophageal malignancies, and complications of aortic surgery, occurring in up to 1.7% of cases (1, 2). While there are no definitive guidelines for AEF management, thoracic endovascular aortic repair (TEVAR) is often considered the first-line treatment to prevent immediate exsanguination from AEF (3). Many advocate definitive open surgical esophageal and aortic repair following TEVAR, as persistent aor-to-enteric communication may result in graft colonization and act as a persistent nidus for infection (4, 5).

Here, we describe a highly unusual case of AEF involving the central aortic arch treated emergently with TEVAR coverage of the fistula and the great vessels, in which *in vivo* fenestration of the endograft was performed to perfuse target vessels. TEVAR involving the central arch is challenging due to endograft coverage of the great vessels. *In vivo* fenestration, first described by McWilliams et al. (6), is a feasible method to emergently perfuse excluded vessels by modifying the endograft after deployment. The technique typically involves 1) perforation of the aortic endograft, 2) hole enlargement with an angioplasty balloon, and 3) fenestration stabilization with a covered stent.

Technique

A 38-year-old woman, with a history of lung cancer treated with radiation therapy complicated by an esophageal stricture, presented to the emergency department with gastrointestinal bleeding. Due to hemodynamic instability, massive transfusion protocol and anti-hypotensive agents were initiated. Following intubation, the patient was brought to the interventional radiology suite.

The aortogram revealed contrast extravasation from the aortic arch between the innominate and the left common carotid arteries into the esophagus, indicating an AEF (Fig. 1). At this time, the patient suffered a cardiac arrest, achieving spontaneous return of circulation after 27 minutes of resuscitation. During this period, tamponade of the esophagus was attempted with placement of an esophageal balloon transnasally and subsequently of a stent-graft (Viabahn 13 mm × 50 mm, Gore Medical) (Fig. 2), the largest emergently available stent in the hospital. Contrast leakage of the aorta persisted.

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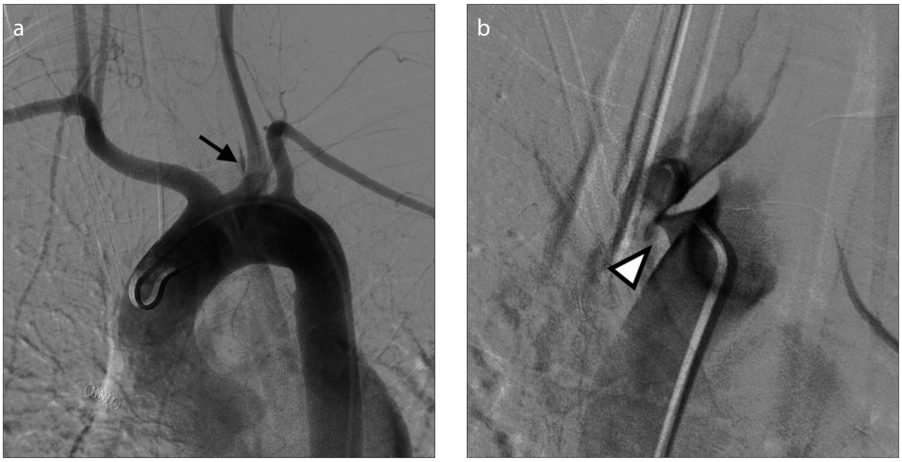

Figure 1. a, b. Initial aortogram **(a)** demonstrates contrast leakage into the esophagus *(arrow)*. Angiogram of the arch **(b)** reveals the large fistulous connection between the aorta (between left carotid artery and left subclavian artery) and the esophagus *(arrowhead)*.

Figure 2. Aortogram after esophageal stent placement shows persistent bleeding from the aorta into the esophagus.

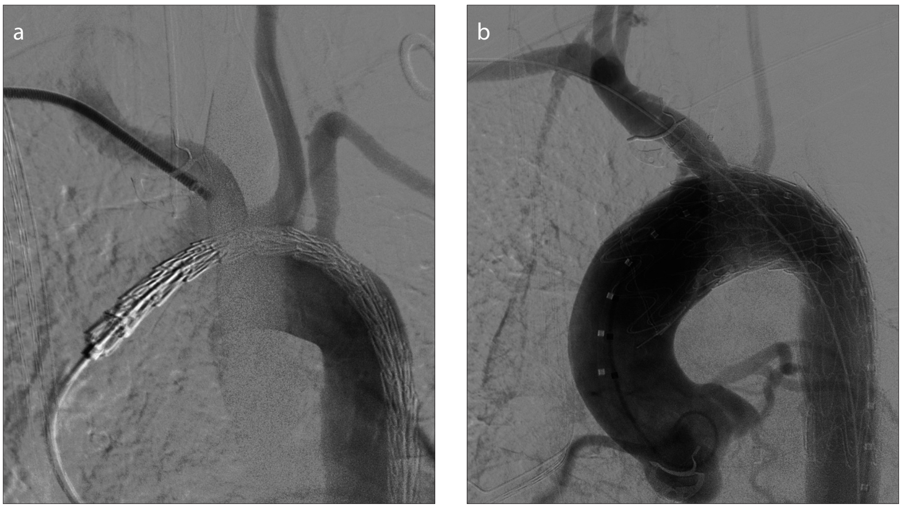

Figure 3. a, b. Contrast injection from the right brachial artery into the innominate artery prior to deployment of an aortic stent **(a)** shows opacification of the arch vessels. Esophageal balloon is seen in relief. Following innominate fenestration of the TEVAR **(b)**, flow into right innominate artery and retrograde filling of the left carotid artery is achieved.

Figure 4. Contrast extravasation from the stented right innominate artery is identified *(arrows)* with retrograde leak into the pleural space, resulting in large hemothorax. The leak was embolized followed by extension of the innominate stent graft.

After a multidisciplinary discussion with the patient's family and the thoracic, cardiac and vascular surgical and the intensive care teams, the decision was made to pursue an endovascular approach rather than open surgical repair of the AEF given the patient's critical status, history of radiation, and the location of the AEF in the arch. The plan was to deploy an endograft in the aortic arch immediately followed by fenestration from the right innominate and left carotid artery access with the understanding that the procedure conferred a high risk of stroke. It was uncertain whether the patient still had preserved neurologic function following prolonged cardiac arrest. Endovascular bypass was considered to preserve cerebral blood flow prior to TEVAR but was not an option given the urgent clinical situation. The presence of direct origin of the left vertebral artery from the arch precluded retrograde flow through a left subclavian artery chimney.

Following the deployment of a 34 mm × 150 mm conformable GORE® TAG® endograft (W.L Gore and Assoc.) in the aortic arch (Fig. 3a), fenestration was achieved with a Rosch-Uchida needle (Cook Medical) from a brachial artery sheath positioned in the innominate artery. Multiple attempts were made to puncture the endograft due to the acute angle of arch vessels relative to device contours and the medial shifting of the innominate with needle passes. The innominate artery fenestration was enlarged with an 8 mm balloon, followed by placement of an 8 mm Atrium iCAST endograft (Atrium Medical) across the fenestration (Fig. 3b). An

Main points

- Thoracic endovascular aortic repair with *in vivo* fenestration can be a life-saving procedure for an arch aortoesophageal fistula.
- *In vivo* fenestration involves perforation of a deployed aortic endograft, hole enlargement by balloon angioplasty, followed by stabilization with a covered stent.
- Complications of aortoesophageal fistula post *in vivo* fenestration include stroke and mediastinitis.

Figure 5. CT angiogram 3D reconstruction of the aortic arch demonstrates the fenestrated aortic stent with antegrade flow into the right carotid and subclavian artery.

initial plan to further retrograde fenestrate from the left carotid artery was abandoned given the presence of a preserved circle of Willis and demonstrated perfusion from the right innominate and carotid arteries.

Contrast leakage distal to the innominate branch endograft was noted, as well as an increase in intrathoracic pressure (Fig. 4). Multiple needle passes to attempt fenestration likely led to puncturing of the innominate branch into the pleural space. Chest tube placement yielded >1 L of blood with immediate improvement in ventilation. Embolization of the leak with five 10 mm × 30 cm Interlock coils (Boston Scientific) followed by placement of a 10 mm × 10 cm Viabahn stent graft (W.L. Gore and Assoc.) to extend the existing innominate endograft completely stopped the contrast extravasation.

Patent fenestrated aortic stent with flow to the right subclavian and right carotid arteries was demonstrated on computed tomography (CT) angiogram (Fig. 5). Brain imaging showed a left middle cerebral infarct, consistent with right-sided hemiplegia on exam. Indicative of the challenges of emergent large-bore arterial access, complications were seen at multiple access sites. On post-procedure day 2, the brachial site required thrombectomy and removal of a sheath found traversing through the median nerve. The groin site, complicated by an iliac dissection and common femoral artery thrombosis, was bypassed with ipsilateral great saphenous vein graft. Four-compartment lower extremity fasciotomies were also performed. Her hospital course was later complicated by aspiration pneumonia and mediastinitis, requiring esophagectomy with cervical diversion on post-procedure day 52.

The patient's neurologic function improved over her three-month hospital stay, regaining the limited ability to speak and ambulate with assistance. She was discharged to a rehabilitation facility with ongoing intravenous antibiotics and a plan for possible future open surgical TEVAR explant and aortic repair.

Discussion

The major challenge of our case was the central location of the AEF in the aortic arch, forcing the endograft placement to span all ostia of the main arch vessels (aortic arch zone 0). From the moment the endograft was deployed until the endograft was successfully fenestrated, the total lack of perfusion to the brain made stroke a virtually inevitable complication. Moreover, only the innominate artery could be re-opened, leaving the left common carotid and subclavian arteries covered. No methods to perfuse the target vessels prior to endograft placement, such as an innominate and left common carotid artery bypass, could be pursued at the time due to the critical condition of the patient. Therefore, TEVAR, while saving the patient's life, resulted in neurological deficits.

In retrospect, there may have been ways to reduce the stroke risk. If tamponade of the esophagus with a balloon were successful following the initial aortogram, the patient could have been stabilized and may have allowed time for creating a bypass to perfuse the target vessels during fenestration. In a single-center retrospective analysis, the stroke risk for aortic arch zone 0 in 32 patients was found to be 9.4% in non-emergent TEVAR cases with bypass creation pre-TEVAR (7). If a bypass still could not be achieved, a quick angiogram of the circle of Willis might help to prioritize which arch vessels to salvage first. Alternatively, a novel temporary endovascular shunt technique has been described as a way to perfuse the brain by placing long sheaths with side ports in the great vessels (8).

Even with successful TEVAR treatment, serious infectious risks remain a concern in AEF patients. The risk of graft infection from the persistent communication with the esophagus, a non-sterile cavity, is permanently elevated. In our case, the patient was taken to open surgery for definitive esophageal repair 52 days post-TEVAR due to concern for mediastinitis. Most authors believe that TEVAR should serve as a bridge to open surgical treatment of AEF rather than a stand-alone treatment, which is considered a Class IIb, Level of Evidence C recommendation by the American Heart Association (AHA) (3). A systematic review of 41 patients with AEF by Antoniou et al. (5) showed that recurrent or new infection developed in 44% of AEF patients treated with TEVAR, and these patients had a significantly higher complication rate and shorter overall survival. Earlier surgical intervention within a few days post-TEVAR is favored to improve survival based on limited case reports (9). According to AHA, 6–8 weeks of initial antibiotic therapy post-TEVAR is recommended, and lifelong suppressive antibiotics may be considered (Class IIb, Level of Evidence C) (3), although Antoniou et al. (5) found that life-long antibiotics did not necessarily achieve better outcome in preventing sepsis.

In conclusion, TEVAR can be a life-saving procedure for massive bleeding from AEF but is not without serious complication risks, including stroke and mediastinitis. As demonstrated in our case report, arch deployment of the endograft is a particularly challenging procedure where perfusion of arch vessels may be achieved emergently via *in situ* fenestration.

Conflict of interest disclosure

The authors declared no conflicts of interest.

References

1. Siegel RL, Miller KD, Jemal A. Cancer statistics, 2017. CA Cancer J Clin 2017; 67:7–30. [Crossref]
2. Low G, Panu A, Millo N, Leen E. Multimodality imaging of neoplastic and nonneoplastic solid lesions of the pancreas. Radiographics 2011; 31:993–1015. [Crossref]
3. Tempero MA, Malafa MP, Al-Hawary M, et al. Pancreatic adenocarcinoma, version 2.2017, nccn clinical practice guidelines in oncology. J Natl Compr Canc Netw 2017; 15:1028–1061. [Crossref]
4. Brennan DD, Zamboni GA, Raptopoulos VD, Kruskal JB. Comprehensive preoperative assessment of pancreatic adenocarcinoma with 64-section volumetric CT. Radiographics 2007; 27:1653–1666. [Crossref]
5. Sultana A, Smith CT, Cunningham D, Starling N, Neoptolemos JP, Ghaneh P. Meta-analyses of chemotherapy for locally advanced and metastatic pancreatic cancer. J Clin Oncol 2007; 25:2607–2615. [Crossref]
6. He C, Wang J, Sun S, et al. Irreversible electroporation versus radiotherapy after induction chemotherapy on survival in patients with locally advanced pancreatic cancer: A propensity score analysis. BMC Cancer 2019; 19:394. [Crossref]

7. Hu X, Qiu H, Zhang L, et al. Recurrent gliomas: Comparison of computed tomography (CT)-guided 125I seed implantation therapy and traditional radiochemotherapy. Cancer Biol Ther 2012; 13:840–847. [Crossref]
8. Stewart A, Parashar B, Patel M, et al. American brachytherapy society consensus guidelines for thoracic brachytherapy for lung cancer. Brachytherapy 2016; 15:1–11. [Crossref]
9. Demanes DJ, Ghilezan MI. High-dose-rate brachytherapy as monotherapy for prostate cancer. Brachytherapy 2014; 13:529–541. [Crossref]
10. Mo Z, Zhang T, Zhang Y, et al. Feasibility and clinical value of CT-guided (125)I brachytherapy for metastatic soft tissue sarcoma after first-line chemotherapy failure. Eur Radiol 2018; 28:1194–1203. [Crossref]
11. Yan H, Xiang Z, Zhong Z, et al. CT-guided (125) I brachytherapy in the treatment of distant metastases in the oral cavity and maxillofacial region. Transl Oncol 2017; 10:90–98. [Crossref]
12. Wang G, Zhang F, Yang B, et al. Feasibility and clinical value of CT-guided (125)I brachytherapy for bilateral lung recurrences from colorectal carcinoma. Radiology 2016; 278:897–905. [Crossref]
13. Zhang FJ, Li CX, Zhang L, Wu PH, Jiao DC, Duan GF. Short- to mid-term evaluation of CT-guided 125i brachytherapy on intra-hepatic recurrent tumors and/or extra-hepatic metastases after liver transplantation for hepatocellular carcinoma. Cancer Biol Ther 2009; 8:585–590. [Crossref]
14. Cron GO, Beghein N, Crokart N, et al. Changes in the tumor microenvironment during low-dose-rate permanent seed implantation iodine-125 brachytherapy. Int J Radiat Oncol Biol Phys 2005; 63:1245–1251. [Crossref]
15. Xiang Z, Li G, Liu Z, et al. 125I brachytherapy in locally advanced nonsmall cell lung cancer after progression of concurrent radiochemotherapy. Medicine (Baltimore) 2015; 94:e2249. [Crossref]
16. Yu YP, Yu Q, Guo JM, Jiang HT, Di XY, Zhu Y. (125) I particle implantation combined with chemoradiotherapy to treat advanced pancreatic cancer. Br J Radiol 2014; 87:20130641. [Crossref]
17. Wang J, Jiang Y, Li J, Tian S, Ran W, Xiu D. Intraoperative ultrasound-guided iodine-125 seed implantation for unresectable pancreatic carcinoma. J Exp Clin Cancer Res 2009; 28:88. [Crossref]
18. Stathis A, Moore MJ. Advanced pancreatic carcinoma: Current treatment and future challenges. Nat Rev Clin Oncol 2010; 7:163–172. [Crossref]
19. Zou YP, Li WM, Zheng F, et al. Intraoperative radiofrequency ablation combined with 125 iodine seed implantation for unresectable pancreatic cancer. World J Gastroenterol 2010; 16:5104–5110. [Crossref]
20. Zhongmin W, Yu L, Fenju L, Kemin C, Gang H. Clinical efficacy of CT-guided iodine-125 seed implantation therapy in patients with advanced pancreatic cancer. Eur Radiol 2010; 20:1786–1791. [Crossref]
21. Wang H, Wang J, Jiang Y, et al. The investigation of 125i seed implantation as a salvage modality for unresectable pancreatic carcinoma. J Exp Clin Cancer Res 2013; 32:106. [Crossref]
22. Liu K, Ji B, Zhang W, Liu S, Wang Y, Liu Y. Comparison of iodine-125 seed implantation and pancreaticoduodenectomy in the treatment of pancreatic cancer. Int J Med Sci 2014; 11:893–896. [Crossref]
23. Heinemann V, Quietzsch D, Gieseler F, et al. Randomized phase iii trial of gemcitabine plus cisplatin compared with gemcitabine alone in advanced pancreatic cancer. J Clin Oncol 2006; 24:3946–3952. [Crossref]
24. Louvet C, Labianca R, Hammel P, et al. Gemcitabine in combination with oxaliplatin compared with gemcitabine alone in locally advanced or metastatic pancreatic cancer: Results of a gercor and giscad phase III trial. J Clin Oncol 2005; 23:3509–3516. [Crossref]
25. Cunningham D, Chau I, Stocken DD, et al. Phase III randomized comparison of gemcitabine versus gemcitabine plus capecitabine in patients with advanced pancreatic cancer. J Clin Oncol 2009; 27:5513–5518. [Crossref]
26. Xu K, Niu L, Mu F, Hu Y. Cryosurgery in combination with brachytherapy of iodine-125 seeds for pancreatic cancer. Gland Surg 2013; 2:91–99. [Crossref]
27. Yu YP, Yu Q, Guo JM, Jiang HT, Di XY, Zhu Y. Effectiveness and security of CT-guided percutaneous implantation of (125)i seeds in pancreatic carcinoma. Br J Radiol 2014; 87:20130642. [Crossref]
28. Wang C, Chen Z, Sun W, et al. Palliative treatment of pelvic bone tumors using radioiodine ((125)I) brachytherapy. World J Surg Oncol 2016; 14:294. [Crossref]
29. Yao L, Cao Q, Wang J, et al. CT-guided (125) I seed interstitial brachytherapy as a salvage treatment for recurrent spinal metastases after external beam radiotherapy. Biomed Res Int 2016; 2016:8265907. [Crossref]
30. Yan H, Mo Z, Xiang Z, et al. CT-guided (125)I brachytherapy for locally recurrent nasopharyngeal carcinoma. J Cancer 2017; 8:2104–2113. [Crossref]
31. Jin Z, Guan L, Xiang GM, Gao BA. Radiation resistance of the lung adenocarcinoma is related to the akt-onzin-pou5f1 axis. Biochem Biophys Res Commun 2018; 499:538–543. [Crossref]