

Mycotic aneurysm of the distal thoracic aorta after botulinum toxin injection for esophageal dysmotility

Scott S. Berman, MD, MHA, FACS, DFSVS, and Joseph S. Sabat, MD, PhD, Tucson, Ariz

ABSTRACT

Endoscopic injection of botulinum toxin is a common method to treat esophageal dysmotility and achalasia. Patients undergoing this procedure who subsequently present with abdominal or back pain and constitutional symptoms should be evaluated for possible complications of the procedure, including occult esophageal perforation, mediastinitis, and mycotic aneurysm of the thoracic aorta. The case described herein illustrates the importance of serial imaging in a patient with persistent symptoms after botulinum toxin injection to identify and to treat occult aortic inoculation leading to mycotic aneurysm before sepsis and aortic rupture ensue with their attendant morbidity and mortality risks. (*J Vasc Surg Cases and Innovative Techniques* 2020;6:388-91.)

Keywords: Mycotic thoracic aneurysm; Botulinum toxin complications

Endoscopic injection of the esophagus with botulinum toxin (BTX) is an accepted treatment of achalasia or distal esophageal spasm. It is considered an effective and relatively low risk procedure with a low risk of major complications. We report the unusual case of mycotic aneurysm of the distal thoracic aorta after BTX injection in a patient with esophageal dysmotility. The patient has provided consent to publish the case details and images.

CASE REPORT

The patient is a 73-year-old Hispanic woman who was being treated for hiatal hernia and esophageal dysmotility. She had previously undergone esophagogastroduodenoscopy for dysphagia and was diagnosed with achalasia. She was advised to undergo esophageal manometry and possible myotomy but never followed through on that evaluation because of transportation issues. Six weeks before admission, she presented again to her gastroenterologist with recurrent dysphagia, underwent repeated esophagogastroduodenoscopy, and was noted to have a dilated esophagus with retained food material. At that procedure, she underwent endoscopic removal of the debris and 100 units of BTX injection. Two weeks later, she presented to her community hospital emergency department with abdominal pain and underwent computed tomography (CT) scan of her abdomen and pelvis with no significant

pathologic change noted to explain her symptoms. The only finding of significance was a dilated, patulous distal esophagus with small hiatal hernia and some layering fluid in the visualized distal esophagus.

One month later, she underwent a high-resolution esophageal motility study demonstrating ineffective esophageal motility. She presented again to the same rural community hospital emergency department 4 days after the motility study with complaints of abdominal pain and fever. She again had CT of the abdomen and pelvis, with the only finding noted by the radiologist "debris in the patulous distal esophagus with hiatal hernia. This places the patient at risk for aspiration. Correlate for achalasia." No mention was made of the new dilation of the midthoracic aorta seen in [Fig 1, A](#) that was not present on the scan from 1 month earlier ([Fig 1, B](#)). She was again discharged from the emergency department only to present again 2 days later to our emergency department in Tucson with ongoing complaints of fever and abdominal pain. Repeated CT scanning was obtained of the abdomen and pelvis, identifying the pathologic change in her thoracic aorta. She was subsequently admitted with the presumptive diagnosis of mycotic aneurysm and started on antibiotics, and vascular surgery consultation was requested. Her C-reactive protein level and erythrocyte sedimentation rate were elevated, although she had no leukocytosis. A meglumine diatrizoate (Gastrografin) upper gastrointestinal series was ordered, but this imaging failed to show evidence of esophageal perforation. The thoracic aorta was incompletely imaged on the initial CT scan performed in the emergency department, so 2 days after admission, CT angiography of the thoracic aorta was performed. This scan not only confirmed the aortic disease but documented an interval increase in size of the aneurysm by 1.6 cm compared with the scan 2 days earlier ([Figs 2 and 3](#)).

A cryopreserved aortic homograft was ordered for the patient, which arrived 21 hours later. Immediately on arrival of the graft, the patient underwent repair of the aneurysm through a sixth interspace left thoracotomy with the cryopreserved aortic homograft by a clamp-and-sew technique ([Fig 4](#)). Intraoperatively,

From the Vascular and Endovascular Surgery, Pima Heart and Vascular.

Author conflict of interest: none.

Correspondence: Scott S. Berman, MD, MHA, FACS, DFSVS, Vascular and Endovascular Surgery, Pima Heart and Vascular, 1815 W St Mary's Rd, Tucson, AZ 85745 (e-mail: bermanaz@gmail.com).

The editors and reviewers of this article have no relevant financial relationships to disclose per the Journal policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

2468-4287

© 2020 The Author(s). Published by Elsevier Inc. on behalf of Society for Vascular Surgery. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

<https://doi.org/10.1016/j.jvscit.2020.04.005>

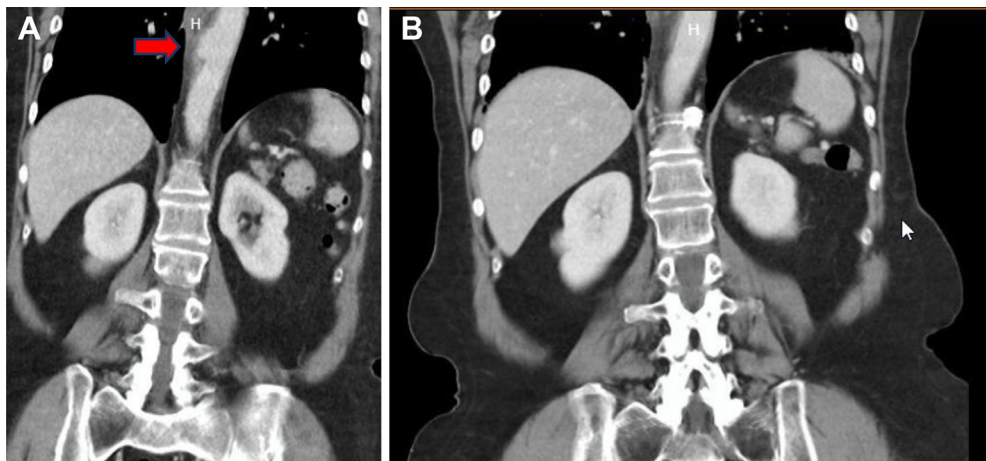


Fig 1. Computed tomography (CT) scans of abdomen and pelvis demonstrating new aortic dilation in the mid to distal thoracic aorta (**A**, arrow) not present on similar scan done 1 month previously (**B**).



Fig 2. Computed tomography (CT) scan obtained 7 weeks after botulinum toxin (BTX) injection into the esophagus showing increasing size of aortic aneurysm in the mid to distal thoracic aorta.

there was minimal inflammation of the mediastinal pleura and tissue overlying the thoracic aorta in the area of concern. The aorta immediately proximal and distal to the area of concern was normal in size and caliber (Fig 4). Culture specimens of the pleural fluid and the ulcerated segment inside the aneurysm were obtained. Segments of the aneurysm wall involving the ulcer were sent for culture and pathologic examination. No attempt was made to débride or to mobilize the medial wall of the aneurysm from the mediastinum, and no gross evidence of purulence or fistula track was revealed. Pathologic examination showed an adventitial abscess in the resected aorta,

although all cultures were negative. The patient's postoperative course was unremarkable, and she was discharged home on postoperative day 7. The infectious disease service recommended 6 weeks of intravenous administration of ertapenem to cover esophageal flora.

DISCUSSION

Endoscopic injection of BTX is widely accepted for treatment of achalasia and distal esophageal spasm, with a reported low risk of major complications. In a review of 386 patients undergoing 661 BTX injections, van Hoeij et al¹ reported 52 (7.9%) mild complications according to the Clavien-Dindo classification² occurring in 48 patients. The most common complications were heartburn, chest pain, and epigastric pain. One patient in their series died of mediastinitis. To date, there have been only two cases reported in the literature of thoracic aortic disease related to endoscopic BTX injection. In 2015, Chao et al³ reported a single case of mediastinitis and pseudoaneurysm formation after endoscopic BTX injection. In 2016, Tan et al⁴ published a case report of an 89-year-old woman who had undergone regular BTX injections for achalasia. Mediastinitis and a small pseudoaneurysm of the thoracic aorta developed. Because of her advanced age and comorbidities, the decision was made to treat her expectantly with antibiotics. She later suffered a cardiac arrest, and after being resuscitated, CT scanning demonstrated enlargement and rupture of the aortic pseudoaneurysm, of which she ultimately died.

Similar to the two cases reported in the literature, this case demonstrates a somewhat indolent presentation after BTX injection, although our case did not demonstrate significant mediastinitis. The presumed mechanism in the case of mycotic aneurysm after endoscopic BTX injection is by inoculation of upper gastrointestinal flora into the aortic wall by penetration of the injection needle through the attenuated, dilated esophageal

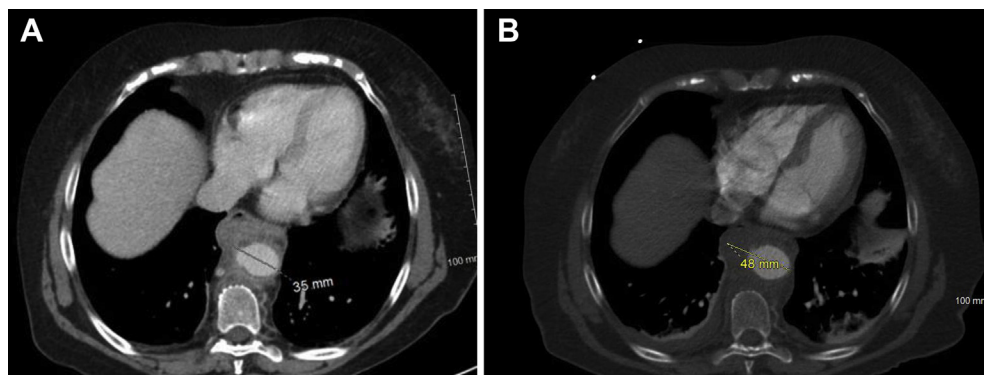


Fig 3. A, Axial computed tomography (CT) scan on admission demonstrating mycotic aneurysm of 3.5 cm. **B,** Repeated axial CT scan 48 hours later demonstrating enlargement of mycotic aneurysm to 4.8 cm.

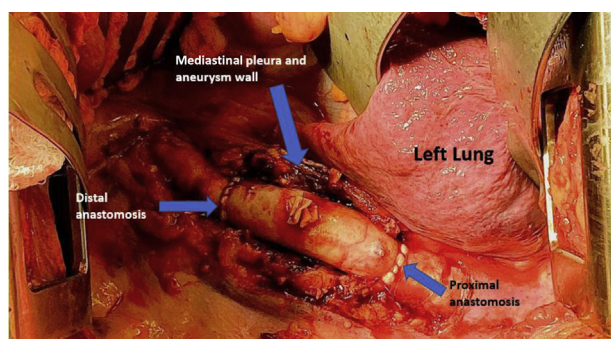


Fig 4. Intraoperative photograph of mycotic thoracic aneurysm repair showing cryopreserved human aortic homograft interposition through a sixth interspace left thoracotomy.

wall. This patient had persistent symptoms of pain within days of her treatment, although initial CT scanning at that time was devoid of significant findings. Her persistent pain and the development of fever and chills precipitated repeated evaluation, wherein the aortic disease was missed by the radiologist. Thankfully our patient pursued further evaluation within days of the missed diagnosis because of her persistent symptoms. The pathologic process was identified and was able to be managed before aortic rupture ensued in the setting of rapidly enlarging aortic aneurysm.

Surgical débridement of infected tissues and reconstruction combined with antibiotics are the mainstays of therapy. In this case, the patient was admitted and prescribed antibiotics while a homograft was acquired. Several alternative options exist for the surgical management of mycotic aneurysms, particularly when aortic homografts are unavailable or emergent intervention is needed. Rifampin-soaked or rifampin-bonded Dacron grafts may be used during open repair.⁵⁻⁷ Femoropopliteal vein autograft is another option for in situ reconstruction.⁸ An extra-anatomic “exclusion bypass” may be performed, followed by a staged approach through

a separate incision for débridement and exclusion of the infected thoracic segment of aorta.⁹

Endovascular stent grafts, especially those that can be pretreated with rifampin, offer a feasible alternative to open repair.¹⁰ The endovascular approach offers the advantages of being less invasive and represents an attractive option particularly in the setting of active hemorrhage or an unstable patient. Stent graft exclusion may be considered either definitive therapy or potentially a bridge to open repair.

CONCLUSIONS

Although an uncommon complication, persistent pain with other constitutional symptoms after BTX injection for esophageal dysmotility syndromes should warrant initial CT imaging to exclude mediastinitis and aortic disease. Serial investigations during a few weeks are suggested if symptoms fail to resolve predicated on this case report and similar publications.

REFERENCES

- van Hoeij FB, Tack JF, Pandolfino JE, Sternbach JM, Roman S, Smout AJ, et al. Complications of botulinum toxin injections for treatment of esophageal motility disorders. *Dis Esophagus* 2017;30:1-5.
- Clavien PA, Barkun J, De Oliveira ML, Vauthey JN, Dindo D, Schulick RD, et al. The Clavien-Dindo classification of surgical complications: five-year experience. *Ann Surg* 2009;250:187-96.
- Chao CY, Raj A, Saad N, Hourigan L, Holtmann G. Esophageal perforation, inflammatory mediastinitis and pseudoaneurysm of the thoracic aorta as potential complications of botulinum toxin injection for achalasia. *Dig Endosc* 2015;27:618-21.
- Tan MZ, Whitgift J, Warren H. Mediastinitis, pseudoaneurysm formation, aortic bleed, and death from endoscopic botulinum toxin injection. *Endoscopy* 2016;48(Suppl 1):E186-7.
- Oderich GS, Panneton JM, Bower TC, Cherry KJ Jr, Rowland CM, Noel AA, et al. Infected aortic aneurysms: aggressive presentation, complicated early outcome, but durable results. *J Vasc Surg* 2001;34:900-8.

6. Ewing MJ, Houck PD, Drake GB, Zehr KJ. Mycotic pseudoaneurysm of the aortic arch with purulent pericardial effusion. *Ann Thorac Surg* 2012;93:1301-3.
7. Uchida N, Katayama A, Tamura K, Miwa S, Masatsugu K, Sueda T. In situ replacement for mycotic aneurysms on the thoracic and abdominal aorta using rifampicin-bonded grafting and omental pedicle grafting. *Ann Thorac Surg* 2012;93:438-42.
8. Kieffer E, Petitjean C, Richard T, Godet G, Dhobb M, Ruotolo C. Exclusion-bypass for aneurysms of the descending thoracic and thoracoabdominal aorta. *Ann Vasc Surg* 1986;1:182-95.
9. Heneghan RE, Singh N, Starnes BW. Successful emergent endovascular repair of a ruptured mycotic thoracic aortic aneurysm. *Ann Vasc Surg* 2015;29:843.e1-6.
10. Clagett GP, Valentine RJ, Hagino RT. Autogenous aortoiliac/femoral reconstruction from superficial femoral-popliteal veins: feasibility and durability. *J Vasc Surg* 1997;25:255-70.

Submitted Mar 25, 2020; accepted Apr 17, 2020.