Dysphagia aortica secondary to thoracoabdominal aortic aneurysm resolved after endograft placement

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ABSTRACT

Dysphagia aortica is a rare entity defined as difficulty in swallowing due to external compression by the aorta. Aneurysmal dysphagia aortica successfully treated with thoracic endovascular aortic repair (TEVAR) is exceedingly rare. We report the case of a 74-year-old woman with known thoracoabdominal aneurysm who presented with acute shortness of breath and 3-month history of dysphagia. Computed tomography angiography revealed aneurysmal growth and massive esophageal dilation. She underwent TEVAR and visceral debranching, which led to complete symptom resolution correlated with sac regression. We also present a comprehensive review of the literature with a focus on cases of aneurysmal dysphagia aortica treated with TEVAR. (J Vasc Surg Cases and Innovative Techniques 2019;5:501-5.)

Keywords: Dysphagia; Dysphagia aortica; Aortic aneurysm; Thoracic endovascular aortic repair; Endovascular therapy

Dysphagia has a wide array of possible causes, including neuromuscular disease, obstructive lesions, and extrinsic compression. Dysphagia aortica is defined as difficulty in swallowing due to external compression by the aorta; it is a rare cause of dysphagia that should remain in the differential diagnosis. Esophageal compression can be caused by an ectatic, aneurysmal, or tortuous aorta, which can sometimes lead to serious complications, such as aortoesophageal fistulas. There are few reported cases of aneurysmal aortica dysphagia and even fewer cases of those successfully treated with thoracic endovascular aortic repair (TEVAR). Here we report a case of dysphagia aortica secondary to degenerative thoracoabdominal aortic aneurysm that was successfully treated with TEVAR with complete symptom resolution. Institutional Review Board approval was waived, and informed consent was obtained from the patient to publish the case details and images.

CASE REPORT

A 74-year-old woman with hypertension and a complex personal and family history of aortic diseases and repairs was referred to the vascular surgery service. She had previously undergone complete arch debranching and aortic replacement from the ascending aorta down to the bottom of T6 for treatment of aortic dissection and aneurysmal degenerations. This was accomplished through four aortic procedures in a span of 9 years. During the course of 7 months before this presentation, she had progressive enlargement of the thoracoabdominal aorta and was being evaluated for an elective repair.

She presented to the emergency department with acute-onset shortness of breath. She also had progressive dysphagia to both solids and liquids for the past 3 months with associated regurgitation and retrosternal chest pain. Computed tomography (CT) angiography revealed 1- to 2-cm growth of the thoracoabdominal aneurysm during 1 year with a diameter of 74 mm and new massive esophageal dilation (Fig 1). The patient was admitted, and esophagogastroduodenoscopy revealed a tight gastroesophageal junction secondary to compression from a pulsatile extrinsic mass. The esophagus was noted to be massively dilated with retained solids and liquid food particles, and the nasogastric tube was looped in the esophagus. She underwent a hybrid procedure involving visceral debranching (aortobi-iliac, aorta-superior mesenteric artery, aorta-celiac, aorta-birenal, and aorta-inferior mesenteric artery bypasses) as well as TEVAR extending from the previous endograft to the aortobi-iliac graft. Two perfusion branches were left open for delayed closure to decrease risk of spinal cord ischemia.

On postoperative day (POD) 1, the patient was taken back to the operating room secondary to bleeding requiring transfusions. A superior mesenteric bypass revision and left renal bypass thrombectomy were performed. On POD 4, the patient remained neurologically stable, and the two perfusion branches of the TEVAR were plugged. CT performed on POD 18 revealed left renal infarct and persisting megaesophagus proximal to the aneurysm. At this point, the patient was nutritionally sustained through a nasogastric feeding tube, and she tolerated the feeds well. Repeated esophagogastroduodenoscopy performed on POD 21 revealed significant extrinsic compression from the residual thoracic aortic aneurysm sac despite repair. The esophagus was dilated and filled with mucoid material. Ultimately, a gastrostomy tube was inserted, and the patient was discharged home on tube feeds. At 8-month follow-up, the patient was asymptomatic and eating well enough for the

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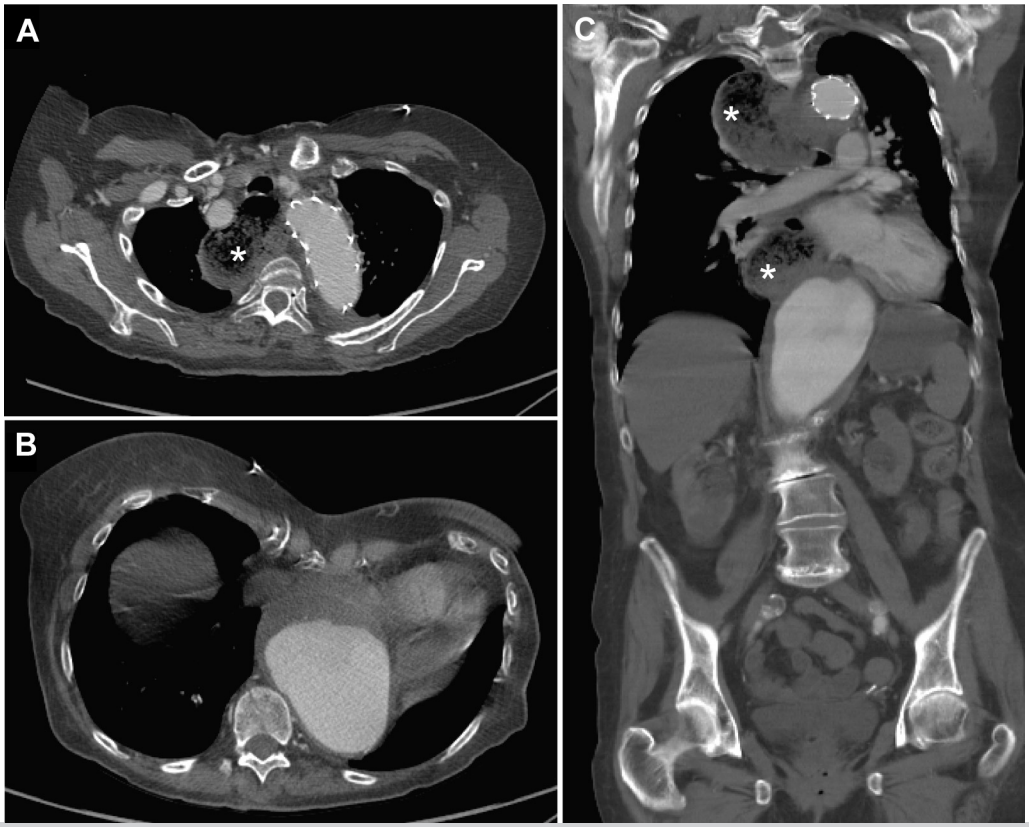

Fig 1. Computed tomography (CT) angiography demonstrating persistent thoracoabdominal aneurysm with associated esophageal dilation before thoracic endovascular aortic repair (TEVAR). Axial view in aortic arch **(A)** and diaphragm **(B)** and coronal view **(C)**. The esophagus is marked with an *asterisk*.

gastrostomy tube to be removed. At 2-year follow-up, the aneurysm sac regressed by 25 mm to a maximum diameter of 49 mm and the esophagus decreased in size by 30 mm to a maximum diameter of 20 mm (Fig 2). At 3-year follow-up, her dysphagia had completely resolved, and the patient had gained back >5% of her initial weight as well. Repeated CT angiography showed no evidence of endoleak and good perfusion of visceral vessels besides the known atrophic left kidney.

DISCUSSION

Dysphagia aortica describes difficulty in swallowing due to external compression by an aneurysmal, tortuous, or ectatic aorta. The term was first coined and used by Pape in 1932.[1] The incidence and prevalence are neither well reported nor studied, although a review of the literature revealed several dozen cases of dysphagia aortica. It is more commonly noted in short kyphotic elderly women and appears to have an association with conditions such as hypertension, left ventricular enlargement, and congestive heart failure.[2] In fact, there are several reports of improved dysphagia aortica after management of these medical comorbidities.[2,3]

Dysphagia aortica should be in the differential diagnosis of patients presenting with new-onset dysphagia with known thoracic aneurysm or when no alternative causes can be identified. A thorough diagnostic workup is warranted with a high index of suspicion because no single modality can definitively diagnose dysphagia aortica, and many of the modalities can produce false positives and negatives.[4] Some diagnostic modalities include barium swallow, which can reveal esophageal obstruction from a pulsatile mass, and esophageal manometry, which can reveal a focal high-pressure band with superimposed pulsations.[4] Dysphagia lusoria, difficulty in swallowing due to compression by an aberrant right subclavian artery, yields similar findings on manometry and can be distinguished from dysphagia aortica by contrast-enhanced CT.[4]

Given the rarity of this condition, no guidelines exist on management of dysphagia aortica. However, the consensus in the literature from case reports is that symptom severity should guide treatment. Mild cases warrant conservative treatment, which includes avoiding sticky solids and opting for a semisolid or liquid diet.[2,4] Treatment of associated medical comorbidities, such as hypertension and heart failure, can help manage symptoms as well.[2,4] In more severe cases, surgery may be warranted. Surgical procedures can target either the esophagus or the aorta and include anterolateral transposition of the distal esophagus, separation of the distal

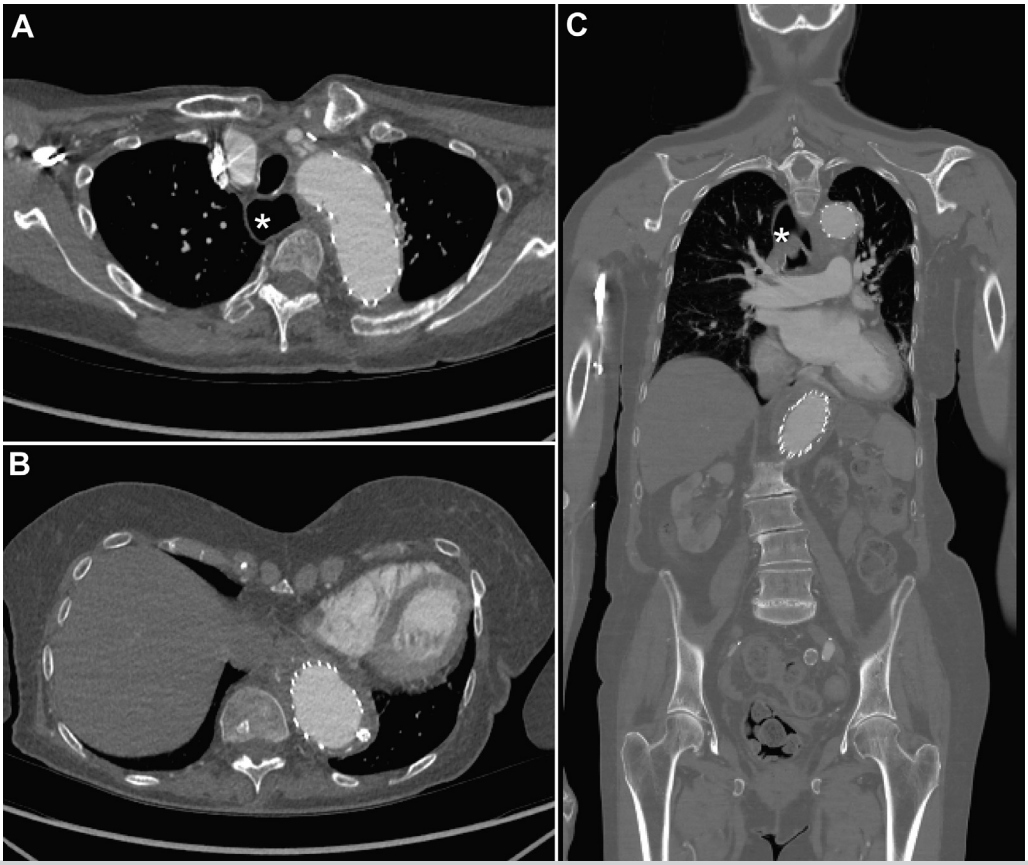

Fig 2. Postoperative computed tomography (CT) angiography after thoracic endovascular aortic repair (TEVAR) for thoracoabdominal aneurysm demonstrating decreased aneurysm size and resolution of esophageal dilation. Axial view in aortic arch **(A)** and diaphragm **(B)** and coronal view **(C)**. The esophagus is marked with an *asterisk*.

esophagus from the aorta, construction of a pleural sling to diminish angulation at the decussation site, esophagomyotomy, division of the right crus of the diaphragm, esophageal stenting, aortic resection, and aortic aneurysm repair.[2,4,5] Severely symptomatic patients who are poor surgical candidates can be symptomatically managed with feeding gastrostomy.[4,5]

To date, there have been fewer than 100 reported cases of dysphagia aortica; the majority of cases have been due to either aneurysms or tortuous anatomy. Other reported causes include pseudoaneurysms, dissections, ulcers, aortoesophageal fistulas, and infection. Thirty-five cases were due to thoracoabdominal aneurysms, and eight of these were treated endovascularly (Table). Of these eight cases, four patients were reported to have at least a partial improvement of dysphagia.[6-9] The most dramatic improvement was seen in a patient who underwent esophageal stenting in addition to TEVAR, which resulted in immediate tolerance of a solid diet.[6] Good symptomatic improvement, although more gradual, has been noted in patients treated only with TEVAR.[7-9] One patient had moderate symptomatic relief at 6 months, and her dysphagia to liquids, although not solids, had resolved.[7] Another was noted to have significantly improved dysphagia at 9 months, whereas one patient had only residual mild dysphagia at 2-month follow-up.[8,9] Improvement of symptoms correlated with sac regression and decrease in esophageal compression.[8,9] This association was seen with our patient as well. Two of the four patients who reported improvement in dysphagia after TEVAR were also treated for concurrent esophageal diseases, such as reflux or Barrett esophagus.[7,8] When the exact etiology of dysphagia is unclear and possibly multifactorial, addressing all possible underlying causes may lead to faster symptom resolution.

In the remaining four patients treated with TEVAR, postoperative dysphagic symptoms were not reported in one case,[10] and the other three patients did not survive long enough postoperatively for assessment of resolution of dysphagia. Causes of death included disease-specific causes, such as esophageal necrosis, and general causes, such as arrhythmia and hospital-acquired infection.[11-13] Despite these reports of poor outcomes, these patients were considerably more unstable or medically comorbid.

Table. Case reports of dysphagia aortica successfully treated with thoracic endovascular aortic repair (*TEVAR*)

Study	Demographic	Etiology	Presentation	Treatment	Outcome
Antón,[7] 2007	75 years, F	Aneurysm, ectasia	Dysphagia to solids and liquids; weight loss	TEVAR (arch to hiatus), diet modification	Moderate symptomatic relief, dysphagia to solids present at 6 months
De Praetere et al,[12] 2010	71 years, M	Aneurysm → rupture	Left thoracic pain, radiating to shoulder; nausea and vomiting, dysphagia	Emergent TEVAR, carotid-carotid bypass	Death POD 24: sepsis from esophageal necrosis
Georgiadis et al,[13] 2018	81 years, M	Aneurysm	Dysphagia to solids and liquids; weight loss; dyspnea; back pain	TEVAR	Death POD 40: hospital-acquired pneumonia
Godar et al,[9] 2013	35 years, F	Aneurysm (inflammatory)	Dysphagia; chest pain; dyspnea	TEVAR, surgical aortic arch repair	Mild dysphagia remaining at 2 months
Hua et al,[10] 2014	40 years, F	Aneurysm → rupture	Dysphagia to solids and liquids	Emergent TEVAR	Well at 5 months, no comments on dysphagia symptoms
Liao et al,[11] 2015	86 years, M	Aneurysm, tortuous anatomy	Dizziness; dysphagia; chest pain, radiating to back; nausea → dyspnea, acute respiratory failure	TEVAR	Death POD 2: respiratory failure
Okamura et al,[6] 2015	87 years, M	Aneurysm	Dysphagia; regurgitation; aspiration pneumonia	TEVAR, esophageal stenting	Immediate relief of dysphagia
Siddiqui et al,[8] 2011	55 years, M	Aneurysm	Dysphagia; heartburn; shortness of breath	TEVAR; PPI for Barrett esophagus at 6 months postoperatively	Dysphagia significantly improved at 9 months postoperatively

POD, Postoperative day; *PPI*, proton pump inhibitor.

CONCLUSIONS

We report one of the few cases of dysphagia aortica successfully treated with TEVAR. Our case reinforces that sac regression often correlates with dysphagia improvement and highlights the importance of medical management while waiting for symptom resolution. TEVAR can be an effective modality for treatment of aneurysmal dysphagia aortica, but this decision should be individualized on the basis of the patient's symptoms and comorbidities.

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