◆ CASE REPORT ◆

An Unusual Case of Dysphagia After Endovascular Exclusion of Thoracoabdominal Aortic Aneurysm

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Purpose: To report an unusual case of dysphagia that developed immediately after stent-grafting of a thoracoabdominal aortic aneurysm.
Case Report: A 79-year-old woman was submitted to a staged hybrid repair of a thoracoabdominal aortic aneurysm and developed new onset dysphagia and regurgitation early after stent-grafting of the thoracic aorta. Esophageal imaging showed a marked endoluminal stenosis, suggesting the development of secondary achalasia. The patient was submitted to endoscopic injections of botulinum toxin at the lower esophageal sphincter, which completely resolved the symptoms.
Conclusion: Acute dysphagic syndrome after thoracic aorta endografting has been anecdotically reported, and its etiology remains undefined. In this report, we illustrate the clinical features of this rare condition, discuss etiological hypotheses, and suggest a noninvasive therapeutic approach.

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Key words: thoracic aortic aneurysm, thoracic endovascular aortic repair, complication, dysphagia, esophageal achalasia

Large thoracic or thoracoabdominal aortic aneurysms (TAAA) may occasionally cause an extrinsic compression of the esophagus that in time may produce a mechanical dysphagia (''dysphagia aortica''). This condition is rare and usually associated with old age, female gender, short stature, hypertension, and kyphosis.[1] The diagnosis of dysphagia aortica is based on specific radiological and manometric findings.[2] Conventional surgical TAA repair relieves esophageal compression, whereas the effects of thoracic endovascular aortic exclusion (TEVAR) are yet to be completely understood.

We report a case of dysphagia that developed immediately after TEVAR, with imaging suggesting esophageal achalasia. To the best of our knowledge, only one other instance of this complication has appeared in the English literature. Chuter et al.[3] reported a case of dysphagia that occurred after branched endovascular TAAA repair, but no details of the case were given.

CASE REPORT

A 79-year-old woman was admitted to our hospital for a 6.8-cm type III TAAA. She had a history of hypertension, chronic obstructive pulmonary disease, hypertrophic cardiomyopathy with severe left atrial enlargement, and smoking. Also, she had previously

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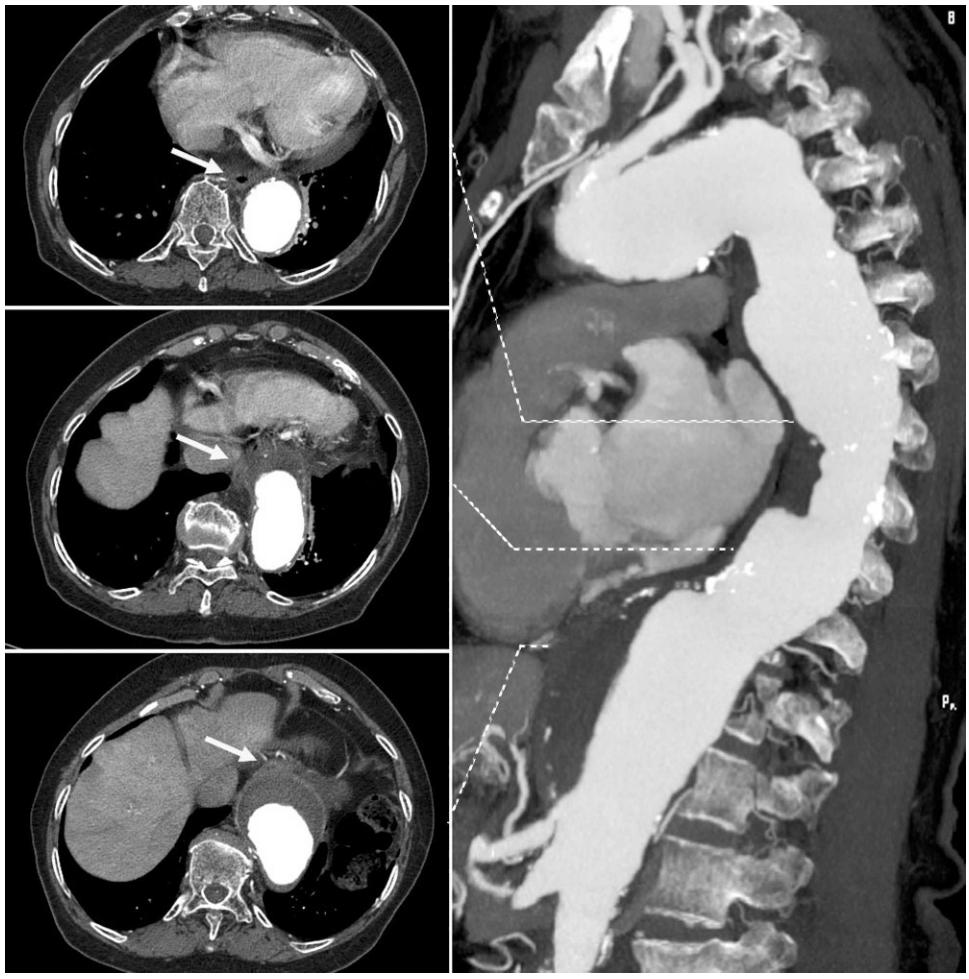

Figure 1 ◆ Preoperative CT scan showing a type III thoracoabdominal aortic aneurysm that anteriorly displaces the esophagus at its distal third (arrows). Also of note is the marked thoracic kyphosis.

undergone surgical infrarenal aortic repair of a ruptured aneurysm. She was short in stature (154 cm) and had evident thoracic kyphosis.

Upon admission, the patient was asymptomatic, with no complaints regarding food intake. At preoperative computed tomography (CT), the distal third of the esophagus was anteriorly displaced by the aortic aneurysm (Fig. 1), without any evidence of upper esophageal dilatation or food retention. Due to severe comorbidities, the patient underwent a hybrid approach. The first surgical stage consisted of a retrograde 3-vessel visceral revascularization via a median relaparotomy, with a customized trifurcated graft sutured to the left limb of the aortobifemoral graft and directed to the celiac trunk, superior mesenteric artery, and right renal artery, as previously described in detail.[4,5] Left renal artery revascularization was aborted for technical reasons.

The postoperative course was uneventful, and the patient was discharged home, where she remained asymptomatic. Three weeks later, she was successfully submitted to the endovascular procedure under general anesthesia. Remote access was gained through the right limb of the existing aortobifemoral graft, and 2 36-mm Zenith TX2 stent-grafts (202 and 127 mm long; William Cook Europe, Bjaeverskov, Denmark) were advanced and deployed in the thoracoabdominal aorta, achieving complete exclusion of the aneu-

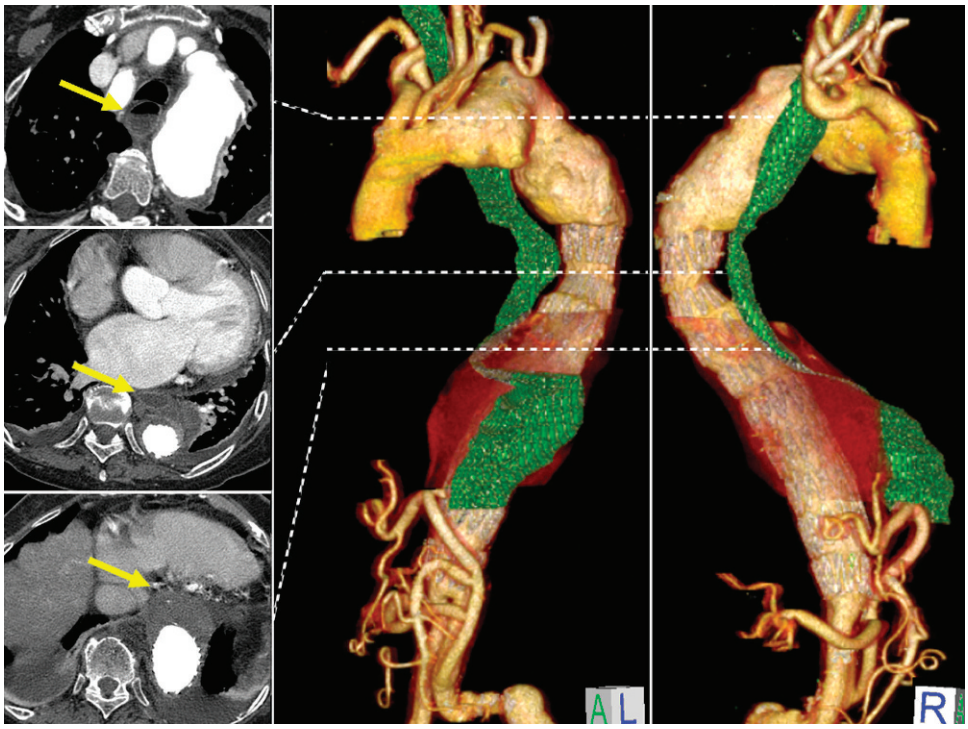

Figure 2 ◆ Postoperative CT after visceral debranching and thoracoabdominal aneurysm endovascular exclusion. Esophageal compression and upper esophageal dilatation are shown on the axial images (arrows) and 3-dimensional reconstructions (yellow is aortic lumen, red is thrombus, and green is the esophagus).

rysm. No perioperative neurological complications were observed.

On resumption of oral food intake (postoperative day 1), the patient immediately complained of dysphagia both for liquids and solids, with constant regurgitation, evolving to complete aphagia within 24 hours. Postoperative CT revealed complete thrombosis of the aneurysm, no endoleak, patency of the visceral bypass grafts, and a marked narrowing of the distal esophagus, with upper dilatation and food retention (Fig. 2). The diameter and shape of the aneurysm remained unchanged. Endoscopy showed distal esophageal stenosis with a normal mucosa. The results of biopsies showed nonspecific mucosal inflammation. Esophageal manometry was not performed due to the lack of patient compliance. A barium esophagogram demonstrated a typical ''bird's beak'' appearance, with dilatation of the upper and middle esophagus and lack of primary peristalsis noticed during fluoroscopy, mimicking secondary achalasia (Fig. 3). The patient was treated with endoscopic injections of 100 IU of botulinum toxin A at the lower esophageal sphincter, obtaining complete relief of symptoms. The remainder of the postoperative course was uneventful. At 6-month follow-up, she is well and asymptomatic.

DISCUSSION

Late aortic remodeling following TEVAR includes aneurysm thrombosis and sac shrinkage. These positive prognostic factors, however, along with a variety of changes in the aortic contour, may occasionally add mechanical stress to the esophageal wall, leading to severe and possibly lethal adverse events, such as esophageal wall erosion and fistulization.[6]

Early esophageal complications after stent-graft deployment are rare, and the etiology remains uncertain. Chuter et al.[3] described a patient who developed similar clinical features, with endoscopy positive for achalasia. In our past experience, we encountered

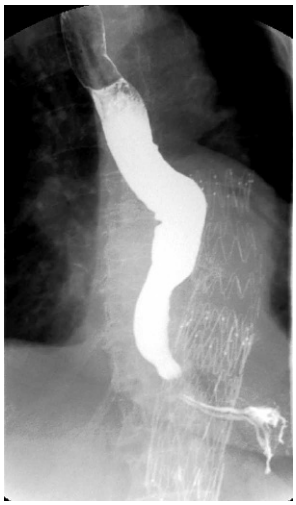

Figure 3 ◆ Postoperative barium esophagogram demonstrating a typical ''bird's beak'' appearance.

another early esophageal complication after TEVAR, but in that case, the dysphagic symptoms were clearly due to compression from a periaortic collection related to a fractured longitudinal strut of a TAG endograft. That patient was successfully managed with open surgical repair.

Preoperatively, the patient in our current report had asymptomatic displacement of the esophagus caused by the aneurysm sac and developed an esophageal functional disorder immediately after the endovascular procedure. The mechanisms underlying this acute dysphagia are unclear. We can speculate, however, that the endograft deployment and thrombosis of the aneurysm may have increased aortic stiffness and impaired esophageal peristalsis. Also, occlusion of the arterial branches feeding the esophagus may have occurred as a result of aneurysm thrombosis, leading to acute hypoperfusion of the lower esophagus and dysfunction of the Auerbach plexus. Finally, a local inflammatory response to the endograft may also have contributed to the development of the syndrome.

Achalasia is an esophageal disorder that presents with symptoms of dysphagia both for liquid and solid food with regurgitation. Typical findings required for diagnosis include esophageal aperistalsis and a poorly relaxing lower esophageal sphincter at esophageal manometry; a smoothly tapering distal esophagus (''bird beak''), with upper esophageal dilatation, at barium esophagogram; and a distended esophagus, with retained food and absence of peristalsis, at endoscopy.[7]

In this case, clinical features, as well as endoscopic and esophagographic findings, resembled that of achalasia, consistent with the findings of Chuter et al.[3] The timing of symptom onset immediately after stent-grafting suggested a causal relationship between the procedure and the esophageal disorder. Unfortunately, the absence of a manometric study limited the definitive diagnosis.

Intrasphincteric injection of botulinum toxin into the lower esophageal sphincter has been reported as a useful and safe method to definitively treat atypical manifestations of achalasia and complex clinical situations in which different factors may contribute to the patient's symptoms.[8] In our case, this procedure resulted in relief from symptoms, thus demonstrating the presence of an esophageal motility disorder, with poor relaxation of the lower esophageal sphincter.

Conclusion

This case shows that TEVAR may acutely alter esophageal physiology, resulting in early onset of esophageal functional disorders.

REFERENCES

1. Wilkinson JM, Euinton HA, Smith LF, et al. Diagnostic dilemmas in dysphagia aortica. *Eur J Cardiothorac Surg*. 1997;11:222–227.
2. Sundaram U, Traube M. Radiologic and manometric study of the gastroesophageal junction in dysphagia aortica. *J Clin Gastroenterol*. 1995;21:275–278.
3. Chuter TA, Rapp JH, Hiramoto JS, et al. Endovascular treatment of thoracoabdominal aortic aneurysms. *J Vasc Surg*. 2008;47:6–16.
4. Chiesa R, Tshomba Y, Melissano G, et al. Hybrid approach to thoracoabdominal aortic aneurysms in patients with prior aortic surgery. *J Vasc Surg*. 2007;45:1128–1135.

5. Chiesa R, Melissano G, Civilini E, et al. Two-stage combined endovascular and surgical approach for recurrent thoracoabdominal aortic aneurysm. *J Endovasc Ther*. 2004;11:330–333.
6. Chocron S, Stoica L, Koch S, et al. Is endovascular stent grafting for descending thoracic aortic disease recommendable in patients with dysphagia? *J Thorac Cardiovasc Surg*. 2002;124:1239–1241.
7. Pohl D, Tutuian R. Achalasia: an overview of diagnosis and treatment. *J Gastrointestin Liver Dis*. 2007;16:297–303.
8. Katzka DA, Castell DO. Use of botulinum toxin as a diagnostic/therapeutic trial to help clarify an indication for definitive therapy in patients with achalasia. *Am J Gastroenterol*. 1999;94: 637–642.