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Atypical esophageal submucosal tumor lesion with aortoesophageal fistula after thoracic endovascular aortic repair

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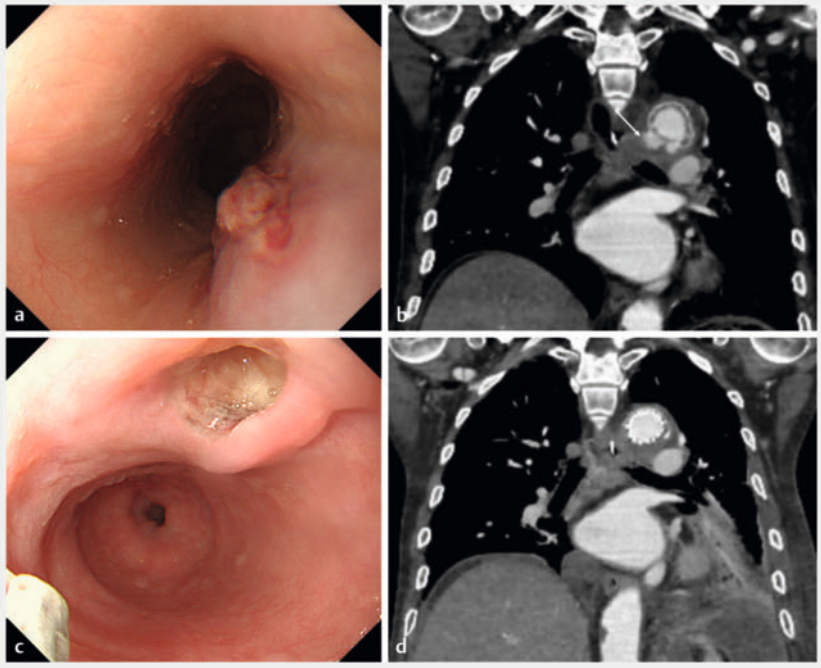
Aortoesophageal fistula (AEF) is a rare but lethal entity that is difficult to diagnose [1]. Despite the promising efficacy of thoracic endovascular aortic repair (TEVAR), which promotes the clinical use of this procedure, the incidence of AEF after TEVAR (post-TEVAR AEF) has increased, making it a major complication [2].

A 77-year-old man who had undergone TEVAR 2 years previously was hospitalized for an iliopsoas abscess. He also had intermittent tarry stools and progressive anemia. Upper gastrointestinal endoscopy (UGE) revealed a submucosal tumor (SMT)-like protrusion that included ulcerative lesions in the upper esophagus (▶ **Fig. 1 a**). Contrast-enhanced computed tomography (CT) imaging revealed extravasation of contrast outside the aortic lumen (▶ **Fig. 1 b**). The man's symptoms were due to the presence of a post-TEVAR AEF accompanied by a stent graft infection; subsequently, a second TEVAR procedure was performed. Seven days postoperatively, UGE revealed an ulcerative lesion without debris (▶ **Fig. 1 c**). Two months postoperatively, the contrast-enhanced CT image showed contrast agent in the aortic lumen with no evidence of leakage (▶ **Fig. 1 d**).

Also at 2 months postoperatively, UGE revealed a recess with an ulcer scar replacing the initial SMT-like lesion (SMTL) (▶ **Fig. 2 a**). Two centimeters from the initially detected AEF lesion on the anal side, another SMTL was identified, which had not been found at the first post-TEVAR AEF detection on UGE. The SMTL protruded into and withdrew out of the esophagus in synchronization with breathing (▶ **Fig. 2 b**). White-light and narrow-band endoscopic imaging showed that the normal mucosa was elongated with normal vessels near the SMTL (▶ **Video 1**).

We suspected that the secondary SMTL originated as a granular mass lesion due to mediastinal infection from the post-TEVAR AEF onto a fragile localized muscular defect [3–5]. Seven months later, a similar SMTL was identified at the AEF scar (▶ **Fig. 2 c, d**).

In this article we have described a rare endoscopic finding obtained during the long-term follow-up of a post-TEVAR AEF with SMTL showing anomalous movement.



▶ **Fig. 1** **a, b** Imaging studies in a patient with intermittent tarry stools and progressive anemia 2 years after thoracic endovascular aortic repair (TEVAR). **a** Upper gastrointestinal endoscopy (UGE) reveals a submucosal tumor-like protrusion, including an ulcerative lesion in the upper esophagus. **b** Contrast-enhanced computed tomography (CT) shows contrast extravasation outside the aortic lumen. **c** At 7 days after a second TEVAR, UGE shows an ulcerative lesion without debris. **d** At 2 months after the second TEVAR, contrast-enhanced CT shows the contrast agent entering the aortic lumen with no leakage.

Video 1 Atypical esophageal submucosal tumor lesion with aortoesophageal fistula after thoracic endovascular aortic repair.

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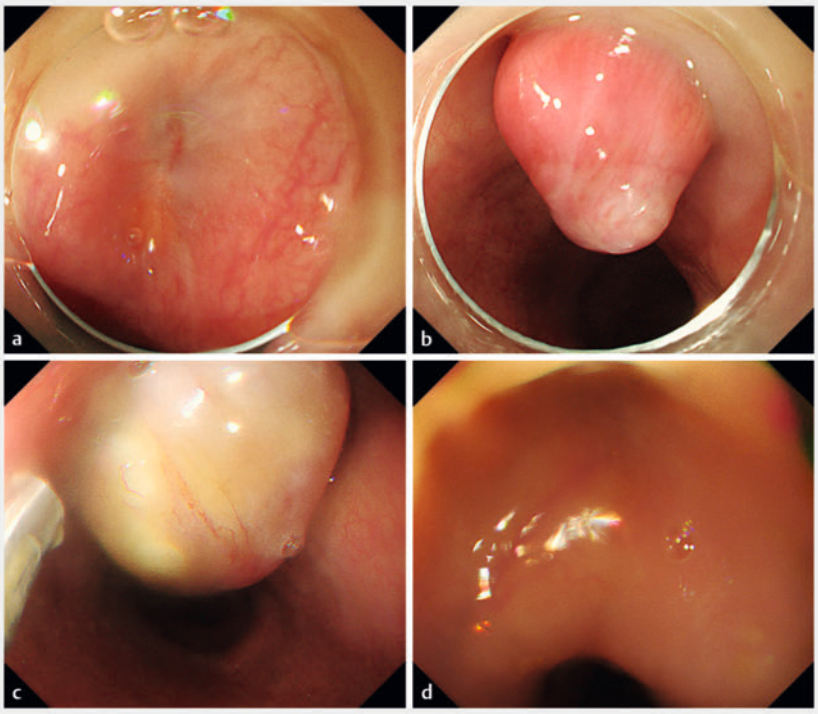
Conflict of Interest

The authors declare that they have no conflict of interest.



▶ **Fig. 2** UGE at 2 and 7 months after the second TEVAR. **a, b** After 2 months: **a** UGE shows a recess with an ulcer scar replacing the initial submucosal tumor-like lesion (SMTL) in the upper esophagus (seen in ▶ **Fig. 1 a**). **b** A second SMTL was located 2 cm from the anal side of the initial aortoesophageal fistula (AEF) lesion; this SMTL had not been detected during the initial identification of the AEF. The SMTL repeatedly protruded into and withdrew out of the esophagus in synchronization with breathing. **c, d** After 7 months: **c** UGE now shows an SMTL on the AEF scar, similar to that seen in **b**. **d** Recess with an ulcer scar replacing the second SMTL shown in **b**.

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