VIDEO CASE REPORT

Postfundoplication submucosal prolapse syndrome

Shou-jiang Tang, MD,[1] Shengyu Zhang, MD,[2] Kenneth D. Vick, MD[3]

INTRODUCTION

Surgical fundoplication is an established treatment for refractory symptomatic esophageal hiatal hernia. Dysphagia after surgery is not uncommon.[1-5] Different etiologies include fundoplication hiatal stenosis, twisted and/or slipped fundoplication, and paraesophageal herniation. We report and describe a previously unrecognized pathology called postfundoplication submucosal prolapse syndrome (PFSPS); we coined this term based on the imaging and pathological findings.

A 79-year-old man underwent Nissen fundoplication 7 years earlier and developed progressive solid-to-liquid dysphagia over 10 months, associated with a 30-pound weight loss because of decreased oral intake. Upper endoscopy was performed at an outside facility, and the gastroscope could not be advanced through the stenotic gastroesophageal (GE) junction, presumably because of extrinsic compression. No ulceration or mucosal abnormality was observed. He was therefore transferred to our medical center for further management.

An abdominal and chest CT scan showed a patulous and fluid-filled esophagus with postoperative changes of the GE junction and abnormal wall thickening of the GE junction causing esophageal stricture (Fig. 1). Routine blood test results were all within normal limits, including white cell counts.

We performed an upper endoscopy and EUS using a linear echoendoscope. On endoscopy, the lumen around the GE junction and within the surgical wrap was moderately and circumferentially narrowed without internal ulceration or erosion (Fig. 2; Video 1, available online at www.giejournal.org). We were barely able to pass a diagnostic gastroscope (9.8 mm) through the stenosis into the stomach. On retroflexion, the surgical wrap appeared disrupted with significant redundant and indurated folds seen at the cardia and fundus (Figs. 3 and 4). The mucosa overlying these indurated folds appeared erythematous and blotchy.

The length of the stenosis was approximately 5 cm. After wire-guided balloon dilatation of the stenosis to 20 mm, the linear echoendoscope was advanced over the guidewire into the stomach. On EUS, the submucosal layer of the entire stenosis was circumferentially thickened to 8 to 11 mm (Fig. 5). There was no extrinsic mass or lymphadenopathy. Fine-needle biopsy of the thickened submucosa was performed, and cytology results showed benign epithelium with abundant inflammatory cells. We

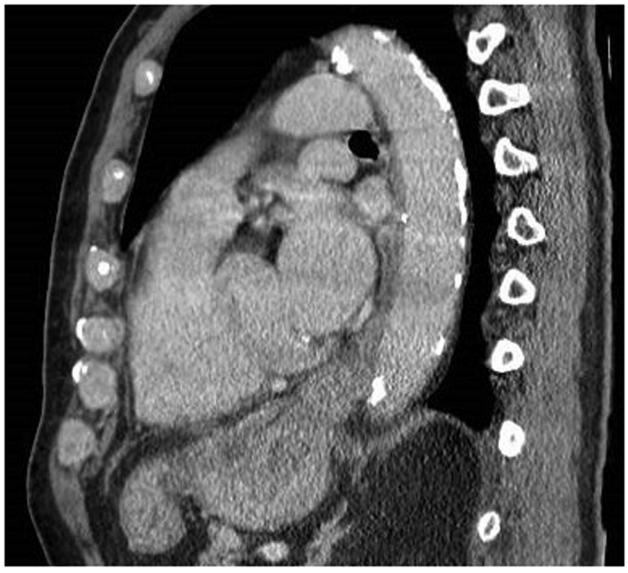

Figure 1. Chest CT scan shows a patulous and fluid-filled esophagus with postoperative changes of the gastroesophageal junction and abnormal wall thickening of the gastroesophageal junction, causing esophageal stricture.

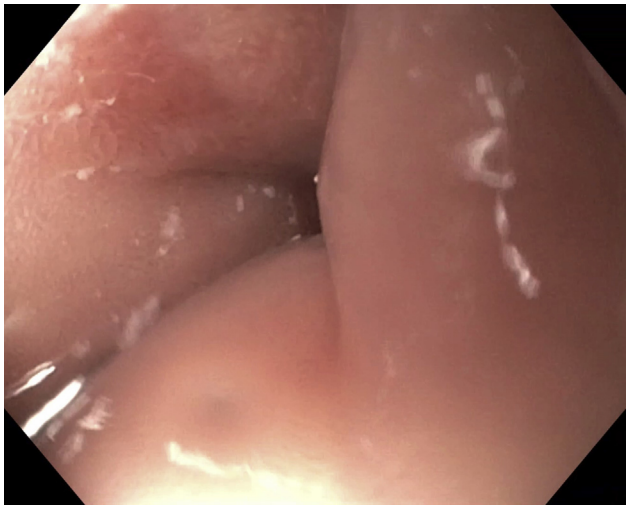

Figure 2. On endoscopy, the lumen around the gastroesophageal junction and within the surgical wrap is moderately and circumferentially narrowed without internal ulceration or erosion.

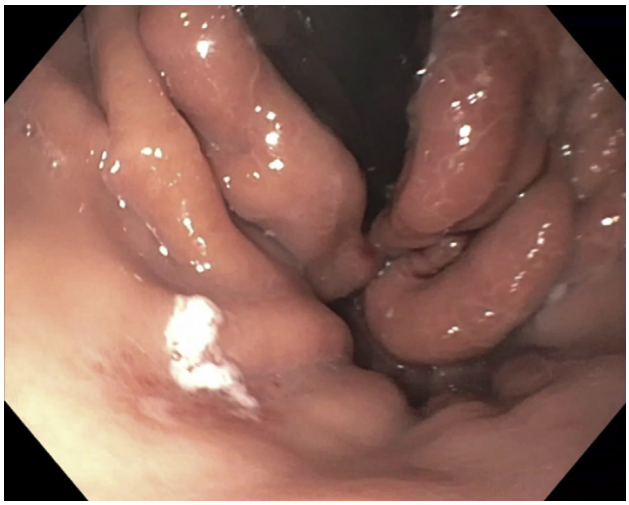

Figure 3. On retroflection, the surgical wraps are disrupted with significant redundant and indurated folds at the cardia.

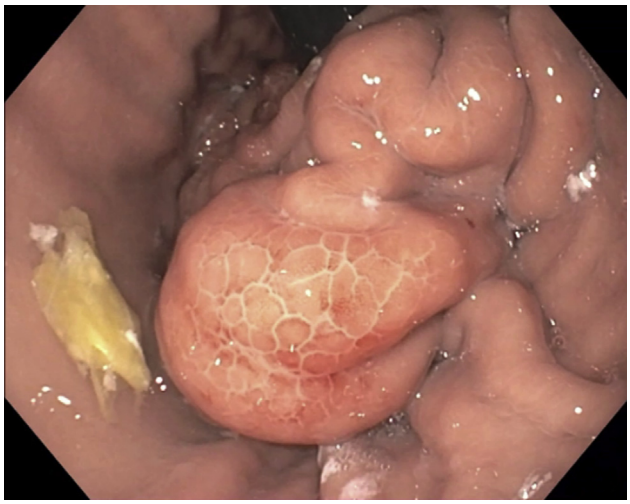

Figure 4. On retroflection, the surgical wraps are disrupted with significant redundant and indurated folds at the fundus. The mucosa overlying these indurated folds appears erythematous and blotchy. The stenosis is 5 cm long.

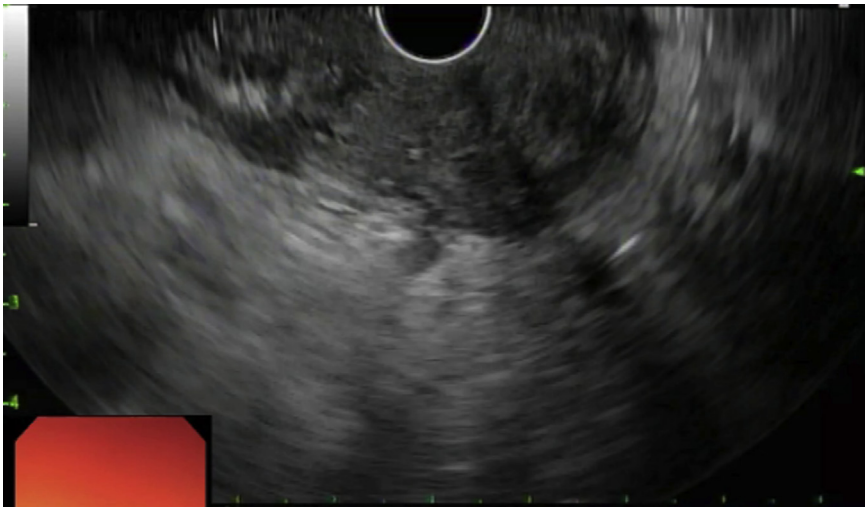

Figure 5. On EUS, the submucosal layer of the entire stenosis is circumferentially thickened to 8 to 11 mm.

also performed a snare biopsy of the redundant folds (Fig. 6).

A 14-mm × 12-mm × 8-mm tissue specimen (Fig. 7) was examined for pathology and revealed benign gastric mucosa with occasional submucosal lymphoid aggregates and fibroadipose tissue. No acute inflammation was seen in this snare biopsy specimen. The diagnosis of PFSPS was made, the term being coined based on the imaging and pathologic findings. We hypothesize that the mucosa and submucosal layers around the GE junction and within the surgical wraps developed chronic prolapse, thus moving distally and retracting back repeatedly. This process leads to submucosal prolapse and thickening or proliferation, with subsequent development of stenosis within the affected segment. Although this has not been previously described and reported, we believe that PFSPS is a previously unrecognized condition.[1-5] Sigmoid colon and rectal mucosal prolapse syndrome are similar but well recognized and can cause bleeding and obstructive symptoms.[6]

The patient's dysphagia was relieved for only 1 to 2 days after endoscopic balloon dilatation. As a result of symptom recurrence, he underwent laparoscopic enterolysis and relief of esophageal outlet obstruction, with revision of the Nissen fundoplication to Toupet fundoplication. Intraoperatively, there was an extensive amount of adhesion around the wrap itself. The fundoplication appeared to be very indurated and was adherent to the left lobe of the liver.

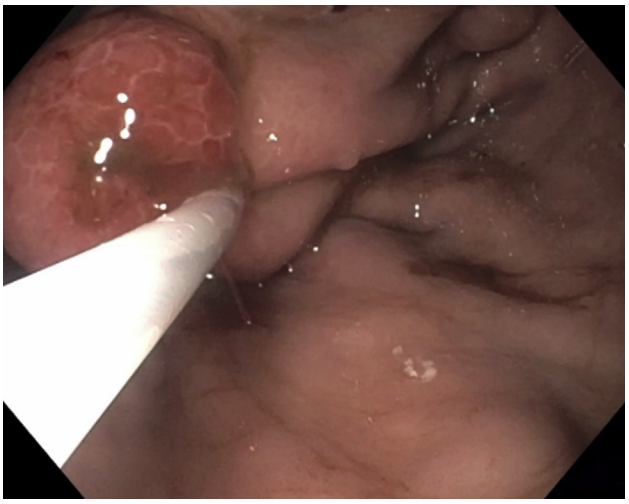

Figure 6. Snare biopsy of the redundant folds.

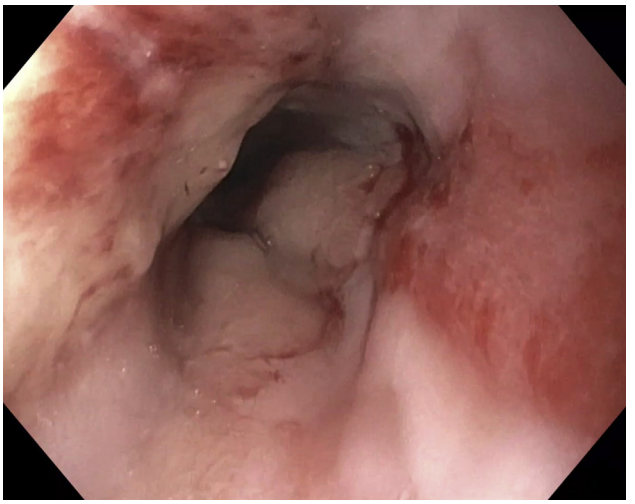

Figure 8. On upper endoscopy, the lumen from the distal esophagus to the intact surgical wrap is patent, allowing easy passage of a gastroscope.

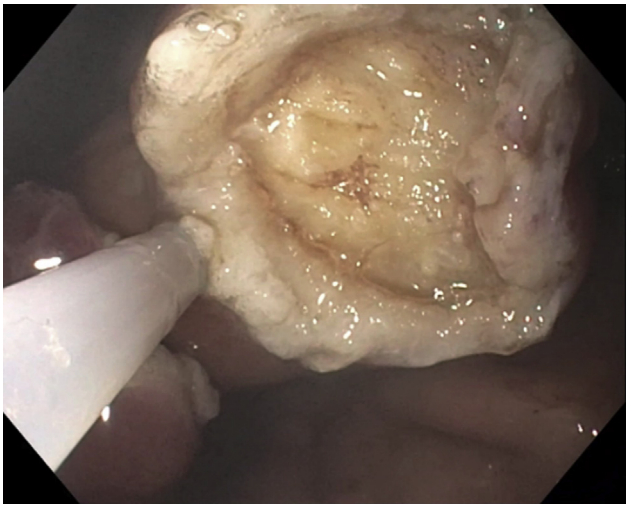

Figure 7. With snare biopsy, a 14-mm × 12-mm × 8-mm tissue specimen is reviewed. Pathologic examination revealed benign gastric mucosa with occasional submucosal lymphoid aggregates and fibroadipose tissue.

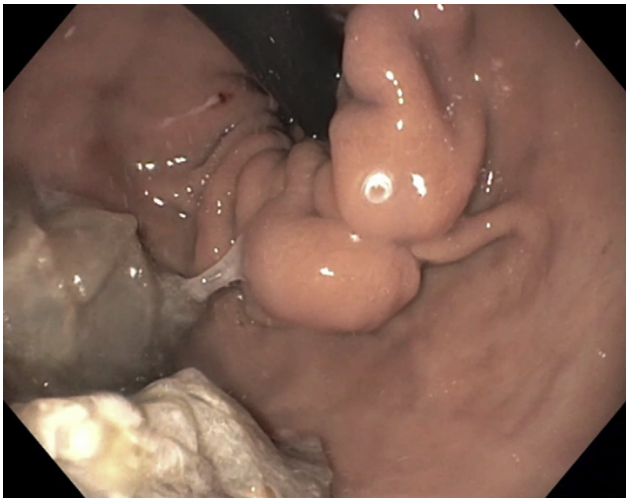

Figure 9. There are 2 linear reflux esophageal ulcerations in the distal esophagus.

The wrap was thought to be too tight, with excessive fundus within the wrap. Furthermore, the wrap had slipped cephalad, causing an hourglass deformity in the distal esophagus just above the GE junction. Within the adjacent gastric wall and plicated stomach, there was an extensive amount of edema and induration. No torsion of the wrap was seen. The wrap was taken down, and restrictive scar was lysed. At this point, the partial fundoplication was completed.

Dysphagia completely resolved after surgery initially; however, 2 months after surgery, some solid food dysphagia returned. On upper endoscopy, the lumen from the distal esophagus to the intact surgical wrap was patent, allowing easy passage of a gastroscope (Fig. 8). There were 2 linear reflux esophageal ulcerations in the distal esophagus (Fig. 9). Dysphagia was thought to be due to esophageal dysmotility from reflux esophagitis. One week after starting a proton pump inhibitor once a day, dysphagia completely resolved, and the patient is asymptomatic 10 months after surgery.

In the case of colorectal pathology, colon mucosal prolapse syndrome, also called solitary rectal ulcer syndrome, represents a common end-pathophysiologic process of focal rectal mucosal ischemia and ulcer formation that results from various purported etiologic factors: recurrent rectal intussusception, pelvic floor dyssynergia, and associated straining.[6] Surgery may sometimes be required, such as in patients with persistent rectal bleeding and/or significant rectal prolapse, recurrent digital manipulation, or instrumentation. We believe that PFSPS is an underrecognized condition by gastroenterologists and surgeons. The differential diagnosis includes fundoplication hiatal stenosis, twisted

and/or slipped fundoplication, paraesophageal herniation, and neoplastic process around the GE junction. Postfundoplication hiatal stenosis develops immediately or very early after fundoplication, and there is no associated submucosal thickening.[1-5] Patient symptoms and careful endoscopic examination of the surgical wrap with selective incorporation of EUS are key in the diagnosis and management of postfundoplication dysphagia.[7,8] Endoscopic dilation is unlikely to resolve dysphagia and maintain long-term luminal patency. We contend that these patients invariably will need surgical revision.

DISCLOSURE

All authors disclosed no financial relationships.

Abbreviations: GE, gastroesophageal; PFSPS, postfundoplication submucosal prolapse syndrome.

REFERENCES

1. Hunter JG, Swanstrom L, Waring JP. Dysphagia after laparoscopic antireflux surgery. The impact of operative technique. Ann Surg 1996;224:51-7.
2. Wills VL, Hunt DR. Dysphagia after antireflux surgery. Br J Surg 2001;88:486-99.
3. Watson DI, Jamieson GG, Mitchell PC, et al. Stenosis of the esophageal hiatus following laparoscopic fundoplication. Arch Surg 1995;130:1014-6.
4. Selima MA, Awad ZT, Filipi CJ. Hiatal stenosis after laparoscopic Nissen fundoplication: a report of 2 cases. JSLS 2002;6:397-9.
5. Orsoni P, Berdah S, Sebag F, et al. An unusual cause of dysphagia after laparoscopic fundoplication: a report on two cases. Surgery 1998;123:241-2.
6. Daram SR, Lahr C, Tang SJ. Anorectal bleeding: etiology, evaluation, and management (with videos). Gastrointest Endosc 2012;76:406-17.
7. Jailwala J, Massey B, Staff D, et al. Post-fundoplication symptoms: the role for endoscopic assessment of fundoplication integrity. Gastrointest Endosc 2001;54:351-6.
8. Jobe BA, Kahrilas PJ, Vernon AH, et al. Endoscopic appraisal of the gastroesophageal valve after antireflux surgery. Am J Gastroenterol 2004;99:233-43.

Division of Digestive Diseases, Department of Medicine, University of Mississippi Medical Center, Jackson, Mississippi (1), Department of Gastroenterology, Peking Union Medical College Hospital, Beijing, China (2), Department of Surgery, University of Mississippi Medical Center, Jackson, Mississippi (3).

If you would like to chat with an author of this article, you may contact Dr Tang at sjtang2000@yahoo.com.

https://doi.org/10.1016/j.vgie.2020.12.003