



## Rumination syndrome after esophagectomy

Daniele Bernardi · Emanuele Asti · Lavinia Barbieri · Luigi Bonavina 

Received: 2 December 2018 / Accepted: 19 December 2018 / Published online: 15 January 2019  
 © Springer-Verlag GmbH Austria, part of Springer Nature 2019

### Summary

**Background** Gastric conduit dysfunction after esophagectomy is a disabling condition that may require revisional surgery. Rumination syndrome is an uncommon and poorly recognized functional foregut disorder of unknown etiology characterized by effortless oral regurgitation of recently ingested food. Rumination is associated with increased intragastric pressure generated by an unconscious mechanism eliciting contraction of the abdominal wall and relaxation of the diaphragm. Rumination syndrome after esophagectomy has not been previously reported in the literature.

**Methods** Two female patients were referred for inability to eat and weight loss due to severe food regurgitation following esophagectomy and gastric conduit replacement. Their previous medical and psychiatric history was apparently unremarkable. Symptoms occurred within the first postoperative year and progressively worsened. High-dose proton pump inhibitors, erythromycin, metoclopramide, antidepressant medications, pneumatic pyloric dilatation, and laparoscopic pyloromyotomy failed to relieve symptoms, and both patients eventually required permanent tube jejunostomy for nutritional support. Subsequently, thoracoscopic implant of a neurostimulator and a laparoscopic Roux-en-Y gastrojejunostomy were performed and failed in both patients.

**Results** All medical and surgical attempts to relieve symptoms were clinically unsuccessful in these pa-

tients in whom radiological and endoscopic investigations did not demonstrate trans-diaphragmatic hernia or mechanical obstruction of the gastric conduit. Eventually, the diagnosis of rumination syndrome was made based on the Rome IV criteria.

**Conclusion** In the absence of anatomical or other functional abnormalities of the gastric conduit, revisional surgery is contraindicated; rather, the diagnosis of rumination syndrome should be considered. The role of behavioral therapy integrated with diaphragmatic breathing training and biofeedback should be investigated in these patients.

**Keywords** Esophagectomy · Rumination syndrome · Diaphragmatic breathing · Functional dyspepsia · Revisional surgery

Severe gastric conduit dysfunction after esophagectomy is a disabling condition that may eventually require revisional surgery. It has been estimated that up to 5% of patients present with dilated/redundant gastric conduit or diaphragmatic hernia 1–6 years after the index operation [1–3], but early inability to eat that requires permanent jejunostomy feeding is extremely rare or perhaps under-reported. Rumination syndrome, an uncommon and poorly recognized functional foregut disorder of unknown etiology, has frequently been associated and sometimes confused with gastroesophageal reflux disease, achalasia, esophageal diverticula, gastroparesis, and eating disorders such as anorexia and bulimia [4]. However, to the best of our knowledge, this syndrome has never been described in patients undergoing esophagectomy for carcinoma or benign disease.

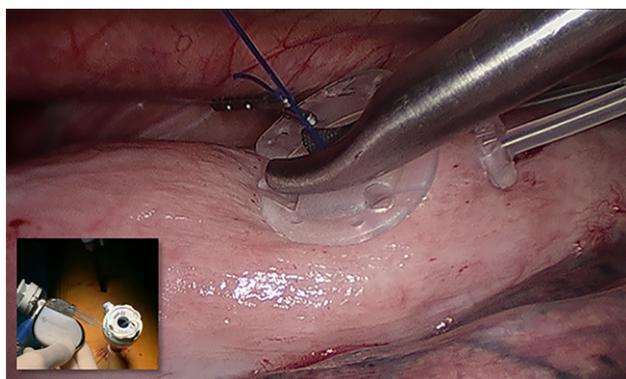
Rumination syndrome is characterized by effortless oral regurgitation of recently ingested food. This phenomenon typically occurs 10–15 min after a meal,

D. Bernardi, MD · E. Asti, MD · L. Barbieri, MD ·  
 L. Bonavina, MD, FACS (✉)  
 Department of Biomedical Sciences for Health, Division of  
 General Surgery, IRCCS Policlinico San Donato, University of  
 Milan Medical School, Via Morandi 30, 20097 San Donato  
 Milanese (Milano), Italy  
[luigi.bonavina@unimi.it](mailto:luigi.bonavina@unimi.it)

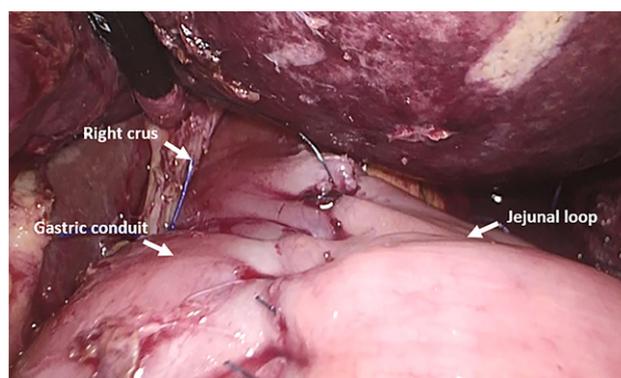
is not preceded by nausea or retching, and the regurgitated material may be swallowed again. Rumination occurs with any food and leads to weight loss in about 40% of patients. From a pathophysiological standpoint, rumination is associated with increased intragastric pressure generated by an unconscious mechanism eliciting contraction of the abdominal wall and relaxation of the diaphragm. Although epidemiologic data are limited, a higher prevalence of rumination has been found in patients with eating disorders, hypochondriasis, depression, fibromyalgia, and rectal evacuation disorders. In most individuals, lower esophageal sphincter pressure and the gastric emptying time are within normal limits [5, 6]. Diagnosis is usually delayed because patients may conceal symptoms. In the majority of patients, the clinical diagnosis of rumination syndrome can be made by using the Rome IV criteria [7]. Objective tests including gastroduodenal manometry, postprandial high-resolution impedance manometry, and abdominal wall electromyography may help to support the diagnosis but are available only in a few specialized centers. Pharmacotherapy is of little or no help in these patients, although baclofen significantly improved symptoms in 63% of patients compared with placebo in a small, randomized, double-blind, placebo-controlled, crossover trial [8]. No controlled studies have demonstrated the efficacy of fundoplication or subtotal gastrectomy in patients with refractory rumination. Instead, techniques of diaphragmatic breathing have been widely recommended and clinical improvement is reported in up to 66% of patients. Biofeedback guided by electromyography of the abdomino-thoracic muscles has the potential to further improve meal tolerance by reducing postprandial muscular tone and increasing lower esophageal sphincter pressure [9].

## Methods

Two female patients, aged 51 and 59 years, were referred to our tertiary-care esophageal center for refractory food regurgitation after esophagectomy



**Fig. 1** Thoracoscopic implant of the Enterra® (Medtronic, Minneapolis, USA) neurostimulator

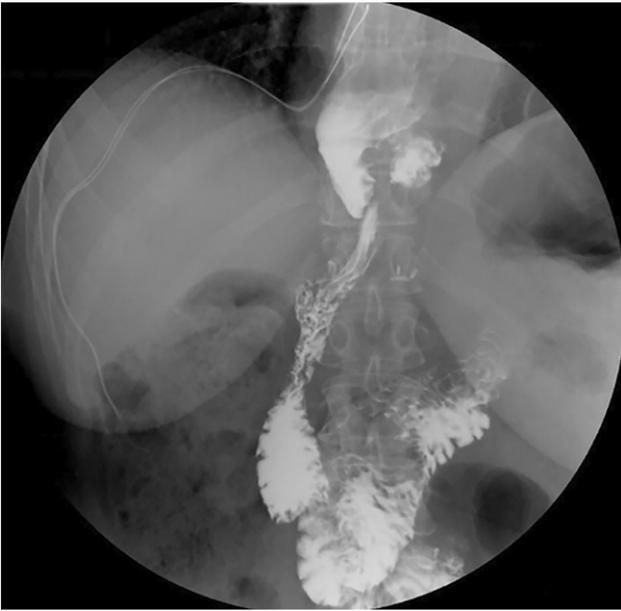


**Fig. 2** Laparoscopic Roux-en-Y gastrojejunostomy

and gastric conduit replacement performed in 2007 and 2012, respectively. Indications for esophagectomy were recurrent achalasia after failure of multiple balloon dilations and Heller myotomy, and T1-N0 squamous cell esophageal carcinoma, respectively. Their previous medical and psychiatric history was apparently unremarkable. The patients complained of persistent food regurgitation and severe weight loss that occurred within the first postoperative year and progressively worsened. Proton pump inhibitors at high dosage, erythromycin, metoclopramide, and antidepressant medications had been totally ineffective. Pneumatic pyloric dilatation and laparoscopic pyloromyotomy failed to relieve symptoms, and both patients eventually required permanent tube jejunostomy for nutritional support. Radiological and endoscopic investigations did not demonstrate trans-diaphragmatic hernia or mechanical obstruction of the gastric conduit. Subsequent attempts to restore oral nutrition consisted of thoracoscopic implant of an Enterra® (Medtronic, Minneapolis, USA) neurostimulator ([10]; Fig. 1), and, as a last resort, laparoscopic Roux-en-Y gastrojejunostomy (Fig. 2).

## Results

The mean follow-up time after the index operation was 7 years. All medical and surgical attempts to relieve symptoms were clinically unsuccessful in these patients in whom radiological and endoscopic investigations did not demonstrate trans-diaphragmatic hernia or mechanical obstruction of the gastric conduit (Fig. 3). Retrospective analysis of hospital charts and further talks with the patients revealed a clinical history highly suggestive of rumination syndrome according to the Rome IV criteria. In addition, we suspected that regurgitation was present several years before the index esophagectomy and was possibly associated with an eating disorder. Both patients refused behavioral therapy and are currently fed by jejunostomy.



**Fig. 3** Gastrografin swallow study after laparoscopic Roux-en-Y gastrojejunostomy in a patient previously implanted with a neurostimulator. The image shows that the contrast medium is injected into both the duodenum and the jejunal loop without any obstruction, but the patient still complained of food regurgitation

## Discussion

It is likely that the permissive thoraco-abdominal pressure gradient may have worsened after esophagectomy owing to loss of lower esophageal sphincter barrier and enlargement of the cross-sectional area at the crural diaphragm [11]. Therefore, in the presence of a widened hiatus, an increased intra-abdominal pressure coupled with the negative intrathoracic pressure may have triggered a repetitive retrograde flow of ingested food within the gastric conduit. This is similar to the typical rumination phenomenon that has been described in patients with an intact upper digestive tract. Unfortunately, although all the Rome IV criteria were fulfilled in our patients, a postprandial high-resolution impedance manometry could not be performed to objectively support the diagnosis.

Best available evidence suggests that a gastric tube provides superior functional outcomes compared with the whole stomach as an esophageal substitute, whereas the benefit of upfront pyloric drainage remains unproven [12]. Nonetheless, gastroparesis and duodenogastric reflux are considered the most common pathophysiological abnormalities causing symptomatic sequelae of esophagectomy and gastric conduit replacement [13]. True gastric conduit obstruction, although less frequent, can necessitate surgical revision to reduce a para-conduit diaphragmatic hernia, to remodel a dilated conduit, or to ensure pyloric transit. This approach has been shown to be beneficial in improving quality of life and preventing aspiration pneumonia in selected patients [3]. It is,

however, plausible that factors other than gastroparesis or mechanical conduit obstruction are involved in the pathogenesis of symptoms, and rumination may be one of these. Intuitively, the threshold of gastric pressurization (>30 mmHg) required for rumination to occur in patients with normal anatomy [5, 14] may be less in the gastric conduit after esophagectomy owing to the enlarged hiatus and the absence of the lower esophageal sphincter barrier.

## Conclusion

We have reported for the first time the diagnosis of rumination syndrome in post-esophagectomy patients. The diagnosis and treatment of rumination syndrome remain a challenge. The important take-home message is that this diagnosis should indeed be suspected in a subgroup of patients complaining of persistent regurgitation after esophagectomy. After an extensive clinical work-up to rule out anatomical or other functional abnormalities, patients fulfilling the Rome IV criteria should be advised to learn diaphragmatic breathing and should be referred to a behavioral psychologist, to a physiotherapist, or to a yoga instructor. The question of whether behavioral therapy integrated with diaphragmatic breathing training and biofeedback can correct the thoraco-abdominal pressure gradient and benefit post-esophagectomy patients with rumination syndrome remains to be answered.

**Acknowledgments** Supported by AIRES (Associazione Italiana Ricerca Esofago).

## Compliance with ethical guidelines

**Conflict of interest** D. Bernardi, E. Asti, L. Barbieri, and L. Bonavina declare that they have no competing interests.

**Ethical standards** All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975 (in its most recently amended version). Informed consent was obtained from all patients included in the study.

## References

1. Kent MS, Luketich JD, Tsai W, Churilla P, Federle M, Landreneau R, et al. Revisional surgery after esophagectomy: an analysis of 43 patients. *Ann Thorac Surg.* 2008;86:975–83.
2. Ganeshan DM, Correa AM, Bhosale P, Vaporciyan AA, Rice D, Mehran RJ, et al. Diaphragmatic hernia after esophagectomy in 440 patients with long-term follow-up. *Ann Thorac Surg.* 2013;96:1138–45.
3. Rove JY, Krupnick AS, Baciewicz FA, Meyers BF. Gastric conduit revision postesophagectomy: management for a rare complication. *J Thorac Cardiovasc Surg.* 2017;154:1450–8.
4. Halland M, Pandolfino J, Barba E. Diagnosis and treatment of rumination syndrome. *Clin Gastroenterol Hepatol.* 2018;16:1549–55.

5. Halland M, Parthasarathy G, Bharucha AE, Katzka DA. Diaphragmatic breathing for rumination syndrome: efficacy and mechanism of action. *Neurogastroenterol Motil.* 2016;28:384–91.
6. Bredenoord AJ, Chial HJ, Camilleri M, Mullan BP, Murray JA. Gastric accommodation and emptying in evaluation of patients with upper gastrointestinal symptoms. *Clin Gastroenterol Hepatol.* 2003;1:264–72.
7. Stanghellini V, Chan FK, Hasler WL, Malagelada JR, Suzuki H, Tack J, et al. Gastroduodenal disorders. *Baillieres Clin Gastroenterol.* 2016;150:1380–92.
8. Pawels A, Broers C, Van Houtte B, Rommel N, Vanuytsel T, Tack J. A randomized double-blind, placebo-controlled, cross-over study using baclofen in the treatment of rumination syndrome. *Am J Gastroenterol.* 2018;113:97–104.
9. Barba E, Burri E, Accarino A, Malagelada C, Rodriguez-Urrutia A, Soldevilla A, et al. Biofeedback-guided control of abdominothoracic muscular activity reduces regurgitation episodes in patients with rumination. *Clin Gastroenterol Hepatol.* 2015;13:100–6.
10. Asti E, Lovece A, Bonavina L. Thoracoscopic implant of neurostimulator for delayed gastric conduit emptying after esophagectomy. *J Laparoendosc Adv Surg Tech.* 2016;26:299–301.
11. Klein WA, Parkman HP, Dempsey DT, Fisher RS. Sphincterlike thoracoabdominal high pressure zone after esophagogastrectomy. *Baillieres Clin Gastroenterol.* 1999;105:1362–9.
12. Akkerman RDL, Haverkamp L, van Hillegersberg R, Ruurda JP. Surgical techniques to prevent delayed gastric emptying after esophagectomy with gastric interposition: a systematic review. *Ann Thorac Surg.* 2014;98:1512–9.
13. Bonavina L, Anselmino M, Ruola, Bardini R, Borsato N, Peracchia A. Functional evaluation of the intrathoracic stomach as an oesophageal substitute. *Br J Surg.* 1992;79:529–32.
14. Kessing B, Bredenoord A, Smout A. Objective manometric criteria for the rumination syndrome. *Am J Gastroenterol.* 2014;109:52–9.