

Retrograde gastroesophageal intussusception after peroral endoscopic myotomy in a patient with achalasia cardia

A case report

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Abstract

Rationale: Retrograde gastroesophageal intussusception (RGEI) is a relatively rare gastrointestinal (GI) disorder in which a portion of the stomach wall invaginates into the esophagus. More recently, peroral endoscopic myotomy (POEM) has emerged as an endoscopic alternative to surgical myotomy for achalasia, and, to the best of our knowledge, our case is the first RGEI after POEM to be reported.

Patient concerns: A 22-year-old male was presented with a history of vomiting, intractable retching and hematemesis for 3 days. He had a history of achalasia and underwent POEM 3 years ago caused by symptoms of severe dysphagia to solid and liquid.

Diagnoses: Initially, the patient was diagnosed with a blood-filled esophagus, and the mid esophagus was occluded with a ball-like mass, however, the final diagnosis of RGEI was made by thoracotomy.

Interventions: A therapeutic strategy of conservative treatment and left transthoracic surgery were applied.

Outcomes: The surgery and post operative course were uneventful, and he remained asymptomatic 1 year after operation.

Lessons: POEM is a reliable and minimally invasive endoscopic method for esophageal achalasia. Early recognition and severity of RGEI are essential to decrease the unwanted complications. Upper GI series, esophagogastroduodenoscopy and computed tomography scan are helpful for diagnostic purposes of RGEI. Conservative treatment, endoscopic intervention, and surgery are the mainstay of treatments for RGEI.

Abbreviations: CT = computed tomography, EGD = esophagogastroduodenoscopy, GI = gastrointestinal, IV = intravenous, LES = lower esophageal sphincter, POEM = peroral endoscopic myotomy, RGEI = retrograde gastroesophageal intussusception.

Keywords: achalasia, case report, peroral endoscopic myotomy, retrograde gastroesophageal intussusception

1. Introduction

Intussusception is the introversion or telescoping of a portion of the gastrointestinal (GI) tract into the luminal cavity of the adjacent portion.^[1] GI intussusceptions are frequently occurring in the region where there is a momentous change in lumen diameter, such as ileocolic and esophagogastric junction. RGEI is a condition characterized by symptoms related to dysphagia, chest pain, vomiting, intractable retching, and rarely GI obstruction. RGEI can result from any predisposing factors,

such as Hiatal hernias, abnormal esophagogastric peristalsis, vigorous contractions, or any condition that increase gastroesophageal peristalsis, motility disorders, or previous gastric surgeries such as, laparoscopic myotomy, and fundoplication, respectively.^[1–3]

2. Methods

The authors provided a written informed consent. This study was approved by the Human Ethics Review Committee of Tianjin Medical University General Hospital, Tianjin, (PR China).

3. Case report

A 22-year-old male was presented with a sudden onset of chest discomfort, vomiting, intractable retching, and hematemesis since 3 days. He had known history of achalasia and underwent POEM 3 years ago caused by symptoms of severe dysphagia to both solid and liquid. But since then no symptoms related to dysphagia, chest discomfort, or intractable retching had developed till the present. The patient denied any history of trauma, vigorous physical activity, or any incident that predisposed to increased intraabdominal pressure.

Upon admission to our digestive department, he appeared wasted, anemic and abdominal examination showed abdominal tenderness. The rest of his physical examinations were essentially

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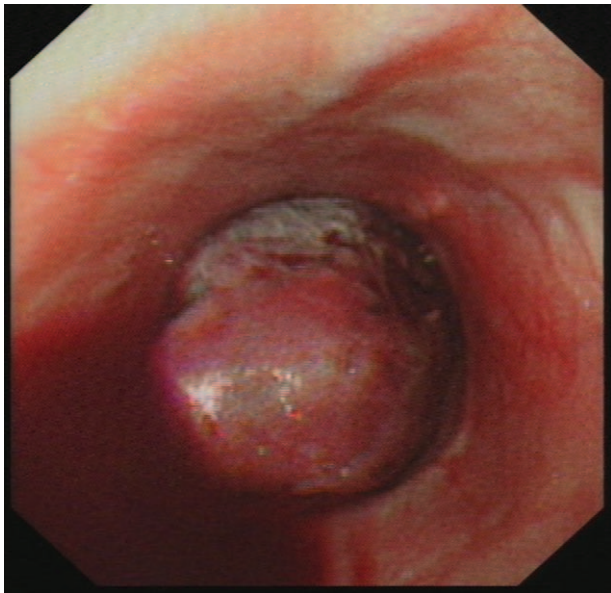


Figure 1. Esophagogastroduodenoscopy revealed a large round intraluminal ball-like mass and massively dilated blood-filled esophagus.

normal. Initial laboratory investigations performed included a complete blood count, renal function tests, liver function tests, electrolytes, coagulation profile, and tumor markers were found completely within normal range except hemoglobin level of 8 g/dL. EGD was performed at initial diagnosis which showed a massively dilated blood-filled aperistaltic esophagus. Within the mid-esophagus, there was a large round intraluminal ball-like mass that was compressible with biopsy forceps (Fig. 1). CT scan showed a soft tissue lesion in the mid-esophagus (Fig. 2). Based on the EGD and CT findings, the patient was suspected of having RGEI.

The patient was started on conservative treatment with intravenous (IV) fluid hydration, IV proton pump inhibitor, and antiemetics, but his symptoms were alleviated shortly after the initial supportive treatment, and he was immediately referred

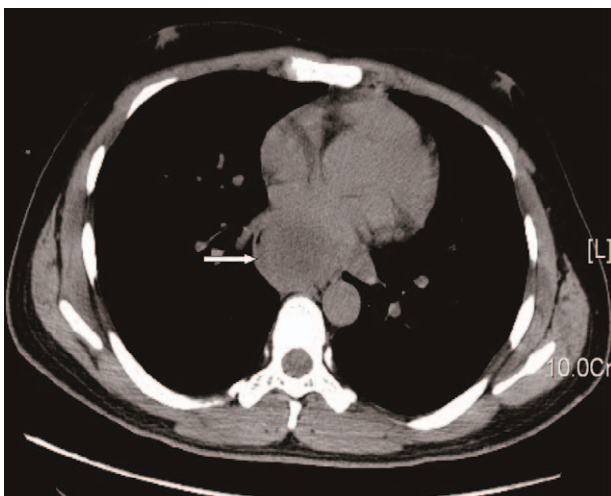


Figure 2. A computed tomography scan demonstrated a soft tissue lesion in the mid esophagus (arrow).

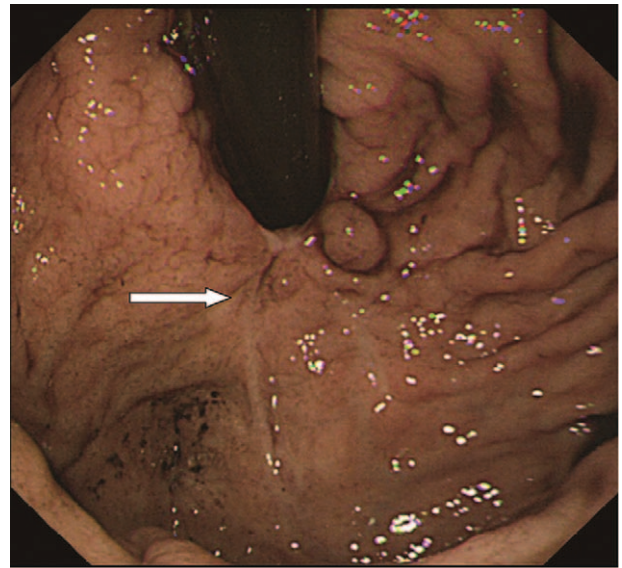


Figure 3. Esophagogastroduodenoscopy revealed normal gastroesophageal lining and scar tissue in distal esophagus and cardia (arrow).

to the surgical department, and the decision for left transthoracic surgery was made. Intraoperatively, the intussuscepted ball-like mass was confirmed as an edematous portion of the gastric fundus and body with erosion and bleeding, and a portion of the stomach was found incarcerated in the mid esophagus which was manually reduced internally and followed by anterior gastropexy. These findings supported a left transthoracic surgical diagnosis of RGEI.

Postoperatively, the patient recovered uneventful, and he was discharged on the 15 postoperative days in a stable condition but was planned to undergo a 3-monthly upper endoscopic examination for the first 1 year after surgery, which showed normal gastroesophageal lining and scar tissue in distal esophagus and cardia (Fig. 3). No complication and recurrence were reported during a 1-year follow-up, and he remained asymptomatic 1 year after the surgical procedure.

4. Discussion

RGEI was first described by Von Rokitansky in 1840 and Lannon and A.Culiner in 1946. RGEI after POEM is a relatively rare gastroesophageal entity, in which a part of the gastric wall invaginates into the esophagus. RGEI usually present with dysphagia, esophageal obstruction, chest pain, vomiting, intractable retching, and rarely upper GI bleeding.^[3] Intussusceptions frequently occur in the region where there is a momentous change in lumen diameter, such as ileocolic and esophagogastric junction result in partial or complete blockade of the GI tract, hypovolemia, dehydration, and shock. All previous cases of RGEI that were diagnosed and reduced by endoscopic procedure and laparotomy are characterized by a loose cardiophrenic ligament, excessively long mesenteric attachments, or lax gastric fixation.^[4] A summary of clinical, etiological factors and management of RGEI are presented in (Fig. 4).^[1-7] We hereby discuss the case of our patient who diagnosed with RGEI after 3 years of POEM procedure for achalasia cardia. His symptoms were markedly improved for 3 years after POEM procedure until he comes with a sudden onset of chest discomfort, vomiting,

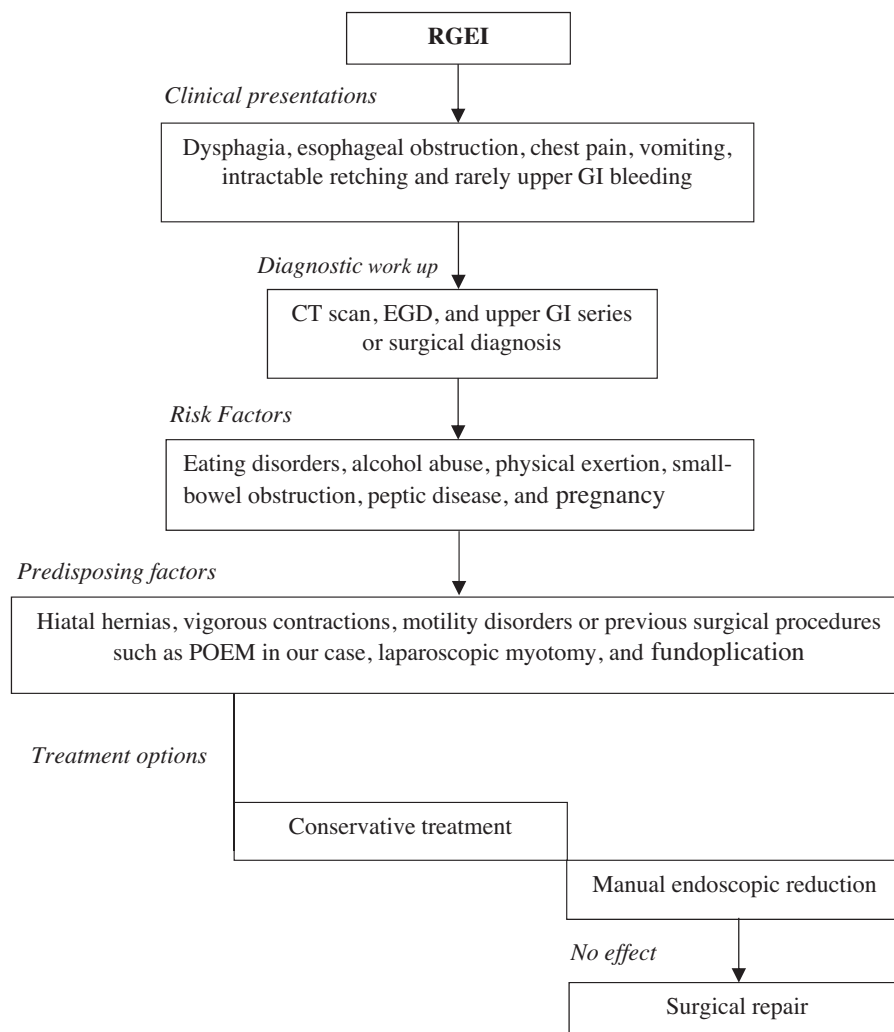


Figure 4. A summary of clinical, etiological factors, and management of RGEI.

retching, and hematemesis. For diagnostic evaluations, we performed CT scan and EGD showed intraluminal ball-like mass with blood-filled obstructed mid esophagus, however, a final diagnosis of RGEI was established by the left transthoracic surgery. Postoperatively, the patient outcome was satisfactory and he remained asymptomatic 1 year after surgical repair.

RGEI might be difficult to differentiate from unreduced gastroesophageal prolapse because endoscopically both appeared as an intraesophageal protrusion of gastric mucosa. POEM is a relatively safe and minimally invasive procedure and is commonly applied to the treatment of achalasia,^[5] but RGEI after POEM never reported before. Recently, Kumar et al^[3] also reported a patient with RGEI after 6 months of Heller myotomy for achalasia. Imaging, particularly CT scan, plays a vital role in diagnosing gastroesophageal intussusception.^[3]

Several mechanisms on the etiology of RGEI remain to be elucidated. Certain risk factors may increase the chance of developing RGEI including eating disorders, alcohol abuse, physical exertion, small-bowel obstruction, peptic disease, and pregnancy.^[6] Many predisposing factors contribute to developing RGEI, including Hiatal hernias, vigorous contractions, motility disorders, or previous surgical procedures such as

POEM in our case, laparoscopic myotomy, and fundoplication, respectively.^[2,3] In the etiology of our case, we assumed 3 possible risk factors that may have the potential to cause RGEI after POEM procedure. POEM has a potential to develop a large gastroesophageal opening and reduce lower esophageal sphincter (LES) pressure. The LES is a specialized thickened region of the circular muscle layer, when a myotomy is performed for the inner circular muscle fibers of LES reduces the LES pressure. The reduced LES pressure has also been reported in patients with RGEI after Heller myotomy for achalasia.^[2,3] Severe vomiting and a sudden increase in intraabdominal pressure might be the possible risk factor for RGEI. According to Post et al,^[7] RGEI can result from a sudden increase in intraabdominal pressure with severe vomiting. The dilation of the esophageal lumen which frequently found in achalasia is likely to be the risk factor in developing RGEI.^[2,5]

Therapeutic approaches for RGEI include conservative treatment, endoscopic, and surgical interventions. Fluid and electrolyte management is paramount to the care of the surgical patient because of surgical procedure. Surgery should perform on patients who do not respond to conservative treatment or endoscopic intervention. However, recurrence of RGEI after

surgical reduction is not very well documented, underscoring the need for close postoperative clinical, radiological and endoscopic follow-up is mandatory. Prognosis depends on the anatomical location, extensiveness, and intussusception period. The prognosis is better if treated with early surgical intervention and supportive care; moreover, the prognosis is poor with perforation and peritonitis.

5. Conclusions

POEM is an effective and minimally invasive endoscopic procedure for esophageal achalasia. Theoretically, completeness of LES myotomy is the key to better outcomes, but delay complication of LES relaxation after POEM may cause gastroesophageal prolapse and RGEI. Identification of RGEI and the relative risk factors remains a challenging task for gastroenterologists. Early recognition and severity of RGEI are essential to decrease the unwanted complications. Upper GI series, EGD and CT scan are helpful for diagnostic purposes of RGEI. Conservative treatment, endoscopic intervention, and surgery are the mainstay of treatments for RGEI. There is currently no standard accepted treatment available for RGEI.

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References

- [1] Ujiki MB, Hirano I, Blum MG. Retrograde gastric intussusception after myotomy for achalasia. *Ann Thorac Surg* 2006;81:1134–6.
- [2] Tessolini JM, Tunis AS, Vlachou P. An unusual cause of recurrent dysphagia. *Gastroenterology* 2016;151:e5–6.
- [3] Kumar I, Chauhan RS, Verma A. Retrograde gastroesophageal intussusception: an exceedingly rare complication of Heller myotomy in a patient with achalasia cardia. *Turk J Gastroenterol* 2017;28:316–8.
- [4] Wong MD, Davidson SB, Ledgerwood AM, et al. Retrograde gastroesophageal intussusception complicating chronic achalasia. *Arch Surg* 1995;130:1009–10.
- [5] Misra L, Fukami N, Nikolic K, et al. Peroral endoscopic myotomy: procedural complications and pain management for the perioperative clinician. *Med Devices (Auckl)* 2017;10:53–9.
- [6] Gowen GF, Stoldt HS, Rosato FE. Five risk factors identify patients with gastroesophageal intussusception. *Arch Surg* 1999;134:1394–7.
- [7] Post PJ, Robben SG, Meradji M. Gastro-oesophageal intussusception after Nissen-fundoplication. *Pediatr Radiol* 1990;20:282.