

Retrograde gastroesophageal intussusception: an exceedingly rare complication of Heller myotomy in a patient with achalasia cardia

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To the Editor,

We present the case of a patient with achalasia cardia who developed retrograde gastroesophageal intussusception (GEI) after Heller myotomy.

A 50-year-old woman presented with epigastric pain, few episodes of vomiting, and dysphasia since 1 month. She underwent Heller myotomy 6 months ago for achalasia. Barium swallow and manometric studies performed at initial diagnosis revealed aperistaltic and dilated mid and distal esophagus (maximum diameter, 5.5 cm), with low amplitude contractions and smooth tapering at the gastroesophageal junction that showed absence of relaxation in response to swallowing. She underwent a laparoscopic technique, in which myotomy was performed from the gastroesophageal junction approximately 6 cm upward on the lower esophagus and extending approximately 2.5 cm to the stomach after releasing gastrohepatic and cardiophrenic ligaments. Antireflux procedure was not performed. Her symptoms were alleviated shortly after the procedure, and she remained apparently asymptomatic before the current episode of illness. The patient denied any history of strenuous physical activity, trauma, or any event that predisposed to increased intraabdominal pressure. Physical examination results were unremarkable. Routine biochemical and hematological investigation results and abdominal ultrasound were normal. A diagnosis of recurrent disease was considered, and contrast enhanced CT (CECT) of the chest and upper abdomen was performed with oralcontrast given in form of "on-table" bolus. CECT revealed gross dilatation of the mid and distal esophagus, and a hypoattenuating, well-defined intraluminal lesion in the distal-most esophagus and GEI junction (Figure 1a). Sagittal reformatted images better characterized the intraluminal mass to be a retrograde invagination of the proximal

stomach into the distal esophagus (Figure 1b). Lung fields and visualized abdominal sections revealed no other significant abnormality. An endoscopic reduction of intussusception was planned and explained to the patient. However, the patient refused to undergo treatment and any further work-up at our center and was eventually lost to follow-up.

Retrograde GEI, a rare occurrence in human beings, is sporadically encountered in veterinary medicine and is most often reported in young dogs and cats that have congenital megaesophagus (1). Only few case reports regarding GEI occurrence in humans exist in the English literature (2-5).

Gastroesophageal intussusception etiology is not completely understood. In animals, congenital megaesophagus and other congenital esophageal defects have been implicated as the underlying cause of GEI (1). However, in humans, most cases have been reported in adults with acquired abnormalities of the gastroesophageal junction. In an isolated pediatric case report by Lukish et al. (2), GEI was preoperatively detected in a 3-year-old child who presented with acute esophageal obstruction. In a systematic review of 42 cases of GEIs and retrograde gastric mucosal prolapse, Gowen et al. (3) revealed that increased abdominal pressure and gastroesophageal junction abnormalities such as abnormal gastroesophageal sphincter relaxation, redundant gastric mucosa, and retrograde peristalsis might be etiological factors. They also identified five risk factors that increased abdominal pressure and caused GEI, i.e., sudden sustained exertion, small-bowel obstruction, acid bile peptic disease, pregnancy, and severe vomiting, particularly in alcoholics. Association between GEI and chronic achalasia has been described in an isolated case report by Wong et al. (4). Although the literature indicates the role of increased gastroesophageal relaxation in causing GEI, only an isolated GEI case that

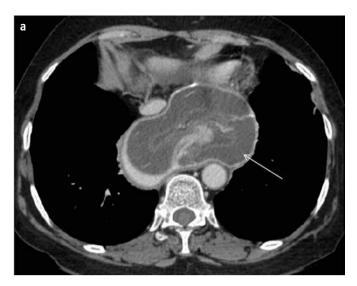




Figure 1. a, b. Axial CECT of the thorax shows a well-defined, hypodense, intraluminal mass in the distal esophagus (a), sagittal reconstructed image shows the invaginated proximal stomach into the distal esophagus, suggesting the diagnosis of GEI (b). The invaginated stomach appeared as mass on axial images

GEI: gastroesophageal intussusception

occurred as a complication of Heller myotomy has been previously reported (5). GEI can clinically present with acute esophageal obstruction, vomiting, upper gastrointestinal bleeding, and severe epigastric or retrosternal pain that radiates to the neck and shoulder and that can mimic cardiac pathology (6).

Imaging, particularly computed tomography (CT), plays an important role in diagnosing GEI. Multiplanar reconstructions in modern day multidetector scanners enable a confident delineation of GEI. In humans, CT reveals telescoping of the fundus into the distal esophagus, as observed in the current case. However, in animals, there are reports showing dislocation of the entire stomach, gastrosplenic ligament, and spleen within the esophagus (1). Treatment options include fundoplication or gastropexy and thoracotomy, followed by manual reduction and esophageal myotomy. Preoperative diagnosis using CT can facilitate minimally invasive surgeries or non-operative reduction by pressure with the esophagoscope (3).

Our case highlights the fact that Heller myotomy is a risk factor for retrograde GEI that occurred because of increased post-procedural relaxation of the gastroesophageal junction. To the best of our knowledge, this is the second report regarding such an association and the third case report to describe preoperative imaging findings of GEI in the English literature.

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