Case Report

Ulcerated Helicobacter pylori negative gastric heterotopy in the upper esophagus causing foreign body sensation

K.H. Katsanos, S. Kamina[1], D.K. Christodoulou, V. Mitsi[1], E.V. Tsianos

SUMMARY

Heterotopic gastric mucosa patches are congenital gastrointestinal abnormalities and have been reported to occur anywhere along the gastrointestinal tract from mouth to anus. We describe herein the diagnosis and treatment of a patient with esophago-bronchial fistula caused by an heterotopic gastric mucosa patch in the mid esophagus. A 35-year old patient was referred to our department for further investigation of foreign body sensation in the upper esophagus for the last week. Upper gastrointestinal endoscopy showed an ulcer below the upper esophageal sphincter area and subsequent biopsies were compatible with Helicobacter pylori negative ulcerated heterotopic gastric mucosa. Further clinical and laboratory investigation was unremarkable. Patient was started on proton pump inhibitors and had resolution of symptoms within the first week of treatment. This is a rare case of foreign body sensation in the upper esophagus caused by ulcerated heterotopic gastric mucosa.

Key words: Gastric heterotopy, foreign body sensation, therapy, ectopic gastric mucosa, helicobacter pylori, esophagus.

There is no conflict of interest in this study.

INTRODUCTION

Ectopic gastric mucosa (EGM) can occur in the fore-, mid-, and hindgut and, conceivably, at any of their derivatives.[1] The origin of EGM is either heterotopic (congenital) or metaplastic (acquired). Congenital heterotopias differ from metaplastic lesions because they usually retain a normally organized structure whereas metaplastic process tend to consist of a single cell type lacking normal tissue patterns. The reported incidence of EGM in the endoscopic literature ranges from 0.29 to 10% but an incidence of up to 70% has been reported in autopsy studies.[2]

Heterotopic gastric mucosa (HGM) patches are congenital gastrointestinal abnormalities and have been reported to occur anywhere along the gastrointestinal tract from mouth to anus.[1-4] In a prospective study of the frequency and clinical importance of heterotopic gastric mucosa (HGM) in the upper esophagus in 643 consecutive veteran patients 10% of them had HGM patches varying in size from 0.2-3x5cm, which were often located immediately below the upper esophageal sphincter.[5]

Developmentally, at the 40mm fetal stage, the esophagus is lined by ciliated columnar epithelium and at about the 130mm fetal stage the columnar epithelium in the mid esophagus is replaced by pseudostratified squamous epithelium spreading in cephalad and caudad directions.[6] HGM in esophagus is thought to arise from gastric precursor cells that remain after incomplete replacement of the original stratified columnar epithelium lining the embryonic esophagus by stratified squamous epithelium.[7-8]

Diagnosis of HGM is many times difficult and requires experience and a high degree of suspicion. At endoscopy, the HGM appears as a mainly flat or slightly raised, well circumscribed red-orange salmon-colored patch. This is mainly a solitary patch but can be multiple measuring from a few millimeters to several centimenters.[9]

Complications of HGM patches include dysphagia, upper gastrointestinal bleeding[10], stricture[11] and fistula[12] formation, upper esophageal ring[13] and adenocarcinoma.[14-15]

1st Department of Internal Medicine & Hepato-Gastroenterology Unit [1]Department of Pathology, University Hospital of Ioannina, Greece

Author for correspondence:

Prof. Epameinondas V. Tsianos, MD, Ph.D, AGAF, Professor of Internal Medicine, Department of Internal Medicine, Medical School, University of Ioannina, Leoforos Panepistimiou, 45 110 Ioannina, Tel:++30-26510-97501, Fax:++30-26510-97016, email: etsianos@ uoi.gr

Interestingly, HGM in the duodenum may also be manifested as dyspepsia.[16]

We describe herein the diagnosis and treatment of a patient with foreign body sensation in the upper esophagus caused by ulcerated heterotopic gastric mucosa.

CASE REPORT

A 35-year old man was referred to our department for further investigation of foreign body sensation in the upper esophagus. Patient had foreign body sensation during the last week and was not recalling any recent episode of food impaction or any kind of trauma in the area. Patient family history was unremarkable. Patient never smoked and he was not an alcohol abuser.

Patient was thoroughly investigated in the Ear-Nose-Throat Department for these symptoms and routine evaluation was negative. As symptoms insisted, patient was referred to our Department for upper gastrointestinal endoscopy and further investigation.

Patient clinical examination was negative and laboratory tests did not reveal something remarkable. Upper gastrointestinal endoscopy was performed with mild sedation. In the upper esophagus an ulcer below the upper esophageal sphincter area was identified surrounded by a 'salmon-like' color appearing mucosa (Figure 1). There were no signs of gastro-esophageal hernia, reflux esophagitis or Barrett's esophagus and overall gastric and duodenal mucosa were normal. Targeted biopsies were taken from the esophageal ulcer, and random biopsies were taken from the stomach and the duodenum.

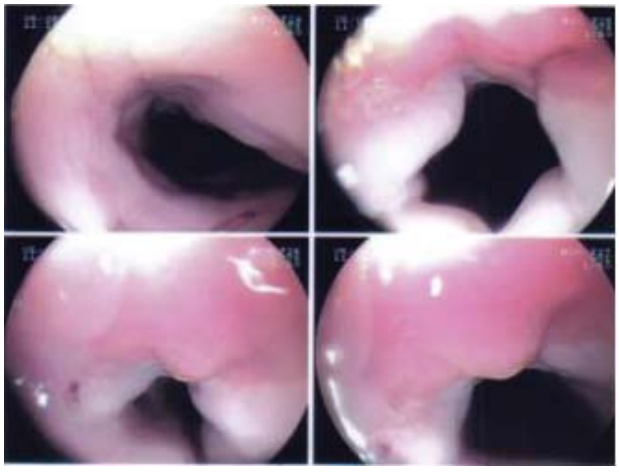

Figure 1. Endoscopic view of the ulcer in the upper esophagus caused by heterotopic gastric mucosa.

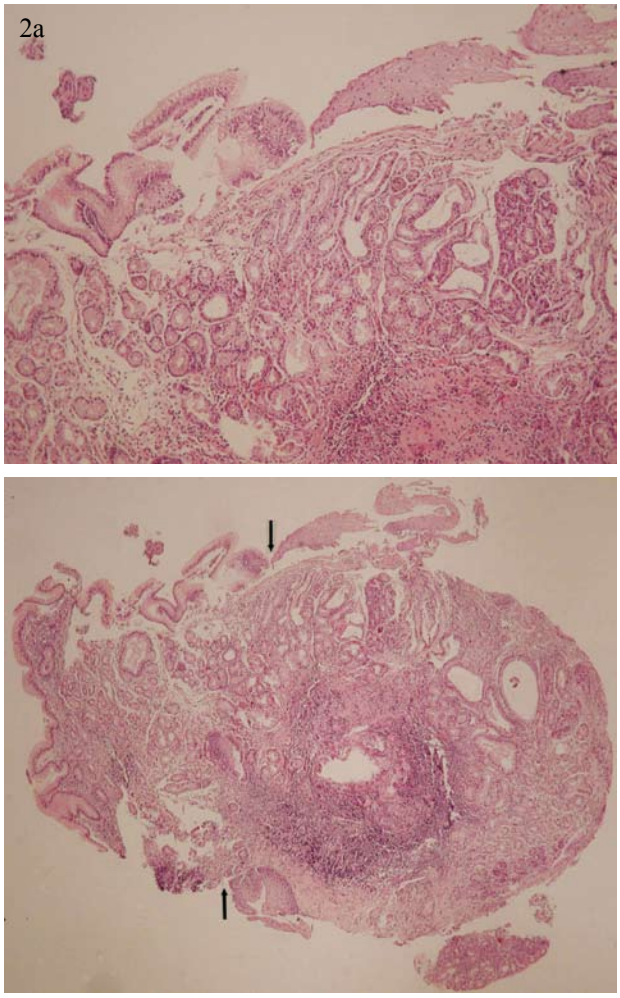

Figure 2. a. Heterotopic gastric mucosa in upper esophagus. Arrows denote the transition between the columnar foveolar epithelium and the esophageal stratified squamous epithelium. (H&E, X 40). **b.** Heterotopic gastric mucosa in upper esophagus. Columnar epithelium merges with stratified squamous epithelium. At the lamina propria cardiac and fundic-type glands are evident (H&E, X 200)

Microscopic examination of the biopsies taken from the esophageal ulcer revealed the presence in the lamina propria of mucus secreting cardiac-type glands and of fundic-type glands, containing both parietal and chief cells. Furthermore, the mucosa was covered by tall, columnar foveolar epithelium, which at the edges merged with the adjacent esophageal stratified squamous epithelium. Goblet cells were not identified. There was no evidence of dysplasia. Microorganisms with the morphological characteristics of Helicobacter pylori were not observed with Giemsa special stain (Figures 2a, 2b).

Patient was diagnosed with H. pylori negative ulcerated heterotopic gastric mucosa in the upper esophagus and was started on therapy with proton pump inhibitors and had complete resolution of symptoms within the first week of treatment. Follow up endoscopy after two months of continuous treatment showed complete ulcer healing. In order to avoid ulcer relapse of other complications, patient was decided to receive six-month maintenance therapy with proton pump inhibitors at full dose and then undergo a follow up endoscopy.

DISCUSSION

To the best of our knowledge this is a rare case of ulcerated heterotopic gastric mucosa in the upper esophagus causing foreign body sensation.

It is noteworthy that HGM diagnosis in our patient was possible during investigation of a torturing symptom, such foreign body sensation is. In fact, HGM patches have been associated with a broad spectrum of symptoms such as ulceration, bleeding, perforation as well as malignant transformations.[2] If asymptomatic, the clinical importance of esophageal HGM patches is debatable.[5]

The asymptomatic nature of these HGM patches, simple oversight and sometimes-technical difficulty can make endoscopists unfamiliar with these lesions. Except of the characteristic 'salmon-colored' appearance of the HGM patch – however, not evident in the fistula opening of our case -, and in order to confirm diagnosis Congo red dye application (1% in distilled water) to the suspected area has been suggested; according to authors[9], after 5 minutes small punctuate areas within the patch turn from red to black, confirming a fall in pH and that this patch is acid-producing. Although pertechnetate scintigraphy (Tc-99m) has been suggested[17] in order to confirm HGM diagnosis as well as to localise other possible patches it proved of no help in our case.

Our patient had no gastro-esophageal hernia and no evidence of concomitant Barrett's esophagus. Traditionally, esophageal HGM is considered a distinct entity from Barrett's esophagus [18] while the presence of esophageal HGM patch has been associated with gastroesophageal reflux disease.[8] Of interest, immunohistological studies suggested a similarity between Barrett epithelium and HGM patch. [18-19] These studies have shown that Barrett epithelium and HGM have the same mucin core protein expression and cytokeratin pattern (cytokeratins 7 and 20)[19] thus, suggesting a pathogenetic link between these two diseases. However, the 30cm distance of HGM from the gastro-esophageal junction and the absence of concomitant Barrett's lesions exclude this probability in our patient. Furthermore, histological changes of the esophageal squamous epithelium distally adjacent to the HGM area in our patient did not show changes consistent with reflux esophagitis.

Despite the fact that HGM patch in this patient was Helicobacter pylori negative we cannot exclude the probability that Helicobacter pylori may also have previously colonized HGM patch contributing to ulcerogenesis. As HGM is able to secrete HCL and pepsinogen, peptic lesions may be induced[20-21] and the synergistic role of H. pylori infection[22] in ulcerogenesis looks probable in our case. In fact, symptomatic ulceration of an acid producing esophageal HGM patch colonized by helicobacter pylori has been previously reported.[9]

We decided to treat our patient the same priciples applied for a gastric ulcer. Treatment of symptomatic HGM is necessary in order to relieve symptoms and to further prevent the development of complications. Efficient treatment can be successfully offered by the use of proton pump inhibitors. If appropriate and when it seems that medical therapy fails to promote regression of symptoms, transcervical or endoscopic biopsy and/or excision are warranted.[1] Endoscopic laser ablation is an acceptable treatment modality because of the rarity of malignant transformation. However, if a small focus of malignancy is suspected, complete local excision with narrow margins is the treatment of choice in order to exclude further progression.[23] Mucosectomy of the patch aided with chromoendoscopy with 0.5% methylene blue has been also suggested[4] and long-term follow up of these patients seems of great importance. However, all these modalities must be allpied with caution in upper esophageal HGM as severe complications related to dysphagia may occur more frequently due to the complicated anatomy of the area.

To conclude, we presented herein a rare case of a patient with foreign body sensation in the upper esophagus caused by ulcerated heterotopic gastric mucosa, which was successfully treated with proton pump inhibitors.

REFERENCES

1. Ishoo E, Busaba N. Ectopic gastric mucosa in the cervical esophagus. Am J Otolaryngol 2002; 23:181-184
2. Chong V-H, Jalihal A. Cervical inlet patch: case series and literature review. South Med J 2006; 99:865-869
3. Wetmore RF, Barlett SP, Papsin B, et al. Heterotopic gastric mucosa of the oral cavity: a rare entity. Int J Pediatr Otorhinolaryngol 2002; 66:139-142
4. De Angelis P, Trecca A, Francalanci P, et al. Heterotopic gastric mucosa of the rectum. Endoscopy 2004; 36:927
5. Borhan-Manesh F, Farnum JB. Incidence of heterotopic gastric mucosa in the upper esophagus. Gut 1991; 32:968-972
6. Johns BAE. Developmental changes in the esophageal epi-

thelium in man. J Anat 1952; 86:431-442
7. Lessels AM, Martin DF. Heterotopic gastric mucosa in the duodenum. J Clin Pathol 1982; 35:591
8. Sanchez-Pernaute A, Hernando F, Diez-Valladares L, et al. Heterotopic gastric mucosa in the upper esophagus (’inlet patch’): a rare cause of esophageal perforation. Am J Gastroenterol 1999; 94:3047-3050
9. Byrne M, Sheehan K, Kay E, et al. Symptomatic ulceration of an acid producing esophageal inlet patch colonized by helicobacter pylori. Endoscopy 2002; 34:514
10. Bataller R, Bordas JM, Ordi J, et al. Upper gastrointestinal bleeding: a complication of “inlet patch mucosa” in the upper esophagus. Endoscopy 1995; 27:282
11. Steadman C, Kerlin P, Teague C, Stephenson P. High esophageal stricture: a complication of ‘inlet patch’ mucosa. Gastroenterology 1988; 95:521-524
12. Kohler B, Kohler G, Rieman JF. Spontaneous esophagotracheal fistula resulting from ulcer in heterotopic gastric mucosa. Gastroentrology 1988; 95:828-830
13. Weaver GA. Upper esophageal web due to a ring formed by a squamocolumnar junction with ectopic gastric mucosa (another explanation of the Patterson-Kelly-Plummer-Vinson syndrome. Dig Dis Sci 1979; 24:959-963
14. Carrie A. Adenocarcinoma of the upper end of the esophagus arising from ectopic gastric epithelium. Br J Surg 1950; 37: 474
15. Takagi A, Ema Y, Horii S, Morishita M, Kino I. Early adenocarcinoma arising from ectopic gastric mucosa in the cervical esophagus. Gastrointest Endosc 1995; 41:167-170
16. Spiller RC, Shousha s, Barrison IG. Heterotopic gastric tissue in the duodenum: a report of eight cases. Dig Dis Sci 1982; 27:880
17. Emamian SA, Shalaby-Rana E, Majd M. The spectrum of heterotopic gastric mucosa in children detected by Tc-99m pertechnetate scintigraphy. Clin Nucl Med 2001; 26:529-535
18. Sampliner RE. Practice guidelines on the diagnosis surveillance and therapy of Barrett’s esophagus. Am J Gastroenterol 1998; 93:1028-1031
19. Lauwers GY, Mino M, Ban S, et al. Cytokeratins 7 and 20 and mucin core protein expression in esophageal cervical inlet patch. Am J Surg Pathol 2005; 29:437-442
20. Hamilton JW, Thune RG, Morrissey JF. Symptomatic ectopic gastric epithelium of the cervical esophagus. Demonstration of acid production with Congo red. Dig Dis Sci 11986; 31:337-342
21. Jabbari M, Goresky CA, Lough J, Yaffe C, Daly D, Cote D. The inlet patch: heterotopic gastric mucosa in the upper esophagus. Gastroenterology 1985; 89:352-356
22. Carrick J, Lee A, Hazell S, Ralston M, Daskalopoulos G. Campylobacter pylori, duodenal ulcer, and gastric metaplasia: possible role of functional heterotopic tissue in ulcerogenesis. Gut 1989; 30:790
23. Abe T, Hosokawa M, Kusumi T, Kusano M, Hokari K, Kagaya H, et al. Adenocarcinoma arising from ectopic gastric mucosa in the cervical esophagus. Am J Clin Oncol 2004; 27:644-645