Heterotopic Gastric Mucosa in Gastric Propria Muscularis Treated by Endoscopic Submucosal Dissection: A Case Report and Literature Review.

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Case Report

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Abstract

Background: Heterotopic gastric mucosa（HGM）is a congenital anatomical variation. It can occur in various tissues and organs of the gastrointestinal tract. Part of the HGM appeared as a solitary, sessile submucosal lesions covered with normal mucosa, and because it was relatively rare, thereby resulting in some HGM to be easily missed or misdiagnosed. We report on a case of heterotopic gastric mucosa in gastric propria muscularis.

Case presentation: A 32-year-old man with abdominal distension for one month. Upper gastrointestinal endoscopy revealed hemispherical lesion covered with smooth mucosa located in gastric antrum. Endoscopic ultrasound (EUS) revealed that it might be derived from the submucosa, with no echogenic nodules, and a partition is visible inside. Endoscopic submucosal dissection (ESD) was performed and histological examination revealed scattered heterotopic mucosal glands (HGG) located in the propria muscularis. Regular follow-up, the patient's abdominal distension was significantly relieved.

Conclusion: HGM of the type of submucosal tumor-like lesion is rare. EUS is a well-established method for submucosal lesion. On EUS, these lesions showed cystic anechoic central core in the submucosa with no solid component,which is similar to gastric cyst. Therefore, this requires us to be alert to the possibility of HGM in order to further evaluate and treat, and if necessary, it can be removed by ESD.

Background

Heterotopic gastric mucosa can be found in all organs of the gastrointestinal tract, particularly in the esophagus, duodenum and ileum[1]. Recent studies observed the lesions endoscopically appeared as a solitary, sessile submucosal tumour-like mass[2, 3]. Morphologically, solitary large HGMs may be difficult to distinguish from other submucosal tumours such as carcinoma, carcinoid tumours or lymphoma. So far, a differential diagnosis for it has yet to be established. With the development of endoscopy, EUS provides valuable information for the diagnosis of submucosal tumours of the gastrointestinal tract. Simultaneously, ESD can achieve complete resection, which has led to the correct diagnosis of submucosal leisions that lacked specific endoscopic presentations.

Case Presentation

A 32-year-old man was admitted to our hospital because of with a history of abdominal distension of one-month duration. No remarkable medical history and family genetic history were observed, and physical examination indicated no positive symptoms. White light endoscopy (OLYMPUS GIF-H290) revealed a hemispherical lesion covered with smooth mucosa located in gastric antrum (Fig. 1A). EUS (OLYMPUS GF-UE260) revealed that it might be derived from submucosal layer, with no echogenic nodules, and a partition is visible inside(Fig. 1B). The size was about 2.0*2.0cm. Combined with endoscopic ultrasound, we think it is likely to be a cyst. However, the size of the lesion was large and located in front of the pyloric duct, causing partial obstruction, which was related to the patient's

abdominal distension symptom. After communicating with the patient and family members, they requested an ESD surgical resection to remove the lesion(Fig. 1C). Eventually, the lesion was completely removed by ESD. Histological examination using hematoxylin–eosin (H&E)-stained sections revealed scattered heterotopic mucosal glands located in the propria muscularis(Fig. 2A, 2B). Immunohistochemistry demonstrated positive staining for Ki67(3%) (Fig. 2C), MUC-6(partial positive) (Fig. 2D), SMA(smooth muscle positive) (Fig. 2E). Furthermore, stains for CD34, CD117 and MUC5AC (Fig. 2F) were negative. The results was consistent with heterotopic gastric mucosa. The patient made an uncomplicated postoperative recovery and was discharged on the 14th hospital day. Presently, the patient's abdominal distension symptom has been considerably relieved.

Discussion

Heterotopic gastric mucosa(HGM)in the gastric was extremely rare. Yamagiwa et al reported the first case of heterotopic gastric glands (HGG) in the submucosa of the stomach [4]. Sugawara et al [5] and Sumida Y et al [6] revealed the cases of adenocarcinoma arising from heterotopic gastric mucosa in the stomach. Hashimoto et al [7] reported a case of gastric adenocarcinoma, as well as HGG within the gastric submucosa and muscularis mucosa in the neighborhood. Recently, Wang H et al [8] reported heterotopic gastric mucosa appearing flat uplift at the esophagogastric junction. The patient eventually underwent ESD to remove the lesion, and histopathological examination identified multifocal heterotopic mucosal glands(HGG) located between the muscularis mucosae and submucosa.

Our case differs from the previously published report in that, in our case, its endoscopic morphology appeared as a submucosal lesions. Moreover, We also performed EUS, which showed it might be derived from submucosal layer, with no echogenic nodules, and a partition is visible inside. Furthermore, our case differed in the pathological findings. In our case, heterotopic mucosal glands located in the propria muscularis. As far as we know ,this is the first report of HGM appearing propria muscularis in stomach.

HGM lacked specific endoscopic presentations, which resulting in such lesions are often overlooked or misdiagnosed. Endoscopic morphology of HGM diverse Previous reports [9–11] described HGM as comprised of solitary or multiple nodules and submucosal tumour-like appearance. Among them, the proportion of the multiple nodules exceed 70%, however, the submucosal leision accounts for the least proportion. In our case, we described the leision of hemispherical HGM with a submucosal tumour-like appearance, which is similar to benign lymphoma, leiomyoma, leiomyosarcoma and carcinoid tumours, ect. So far, literature with regard to the EUS description of gastric HGM was rare. The EUS findings in our cases showed no echogenic nodules, and a partition is visible inside. We believe that the partition may be related to heterotopic mucosal glands.

According to the literature, there are 2 hypotheses about the pathogenesis of HGM. The first hypothesis is that the submucosal heterotopic gastric glands have been considered to be congenital malformation. The other possibility is that an abnormal proliferation associated with inflammation [12]. There is no consensus about it.

Heterotopic gastric mucosa is often considered to be a benign disease, whereas there was still controversy regarding the relationship between HGM and gastric cancer. Rubio and Mandai [13] argued that heterotopic gastric mucosa is related to carcinogenesis, but other professors maintained that heterotopic gastric mucosa is not associated with gastric cancer [4]. Gastric cancer arising from the HGG is often difficult to diagnose preoperatively because the cancer components are often located in the submucosa. Therefore, most of them are confirmed by postoperative pathology. Recently, Itoh K et al [14] suggested EUS can determine the coexistence of tumor and cystic regions, that is, isohyperechoic area may be the adenocarcinoma component, and the low echoic area at the edge of the tumor may be the cystic component, which can make a preliminary assessment of the lesion.

HGM of the type of submucosal tumor-like lesion is infrequent. On EUS, these lesions showed cystic anechoic central core in the submucosa with no solid component, which is similar to gastric cyst. Therefor, this case suggested that EUS suggested a cystic anechoic submucosal mass, which we could not simply diagnose it as a cyst. It might be heterotopic gastric mucosa. Thus, close follow-up through endoscopy is recommended, and complete resection is required if carcinogenesis occurs or the lesion is large in size.

Abbreviations

HGM
Heterotopic gastric mucosa
EUS
Endoscopic ultrasound
ESD
Endoscopic submucosal dissection
HGG
heterotopic mucosal glands.

Declarations

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Author contributions

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Conflict of Interest

The authors of this work have nothing to disclose.

Availability of data and materials

Not applicable.

Ethics approval and consent to participate

Not applicable.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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Figures

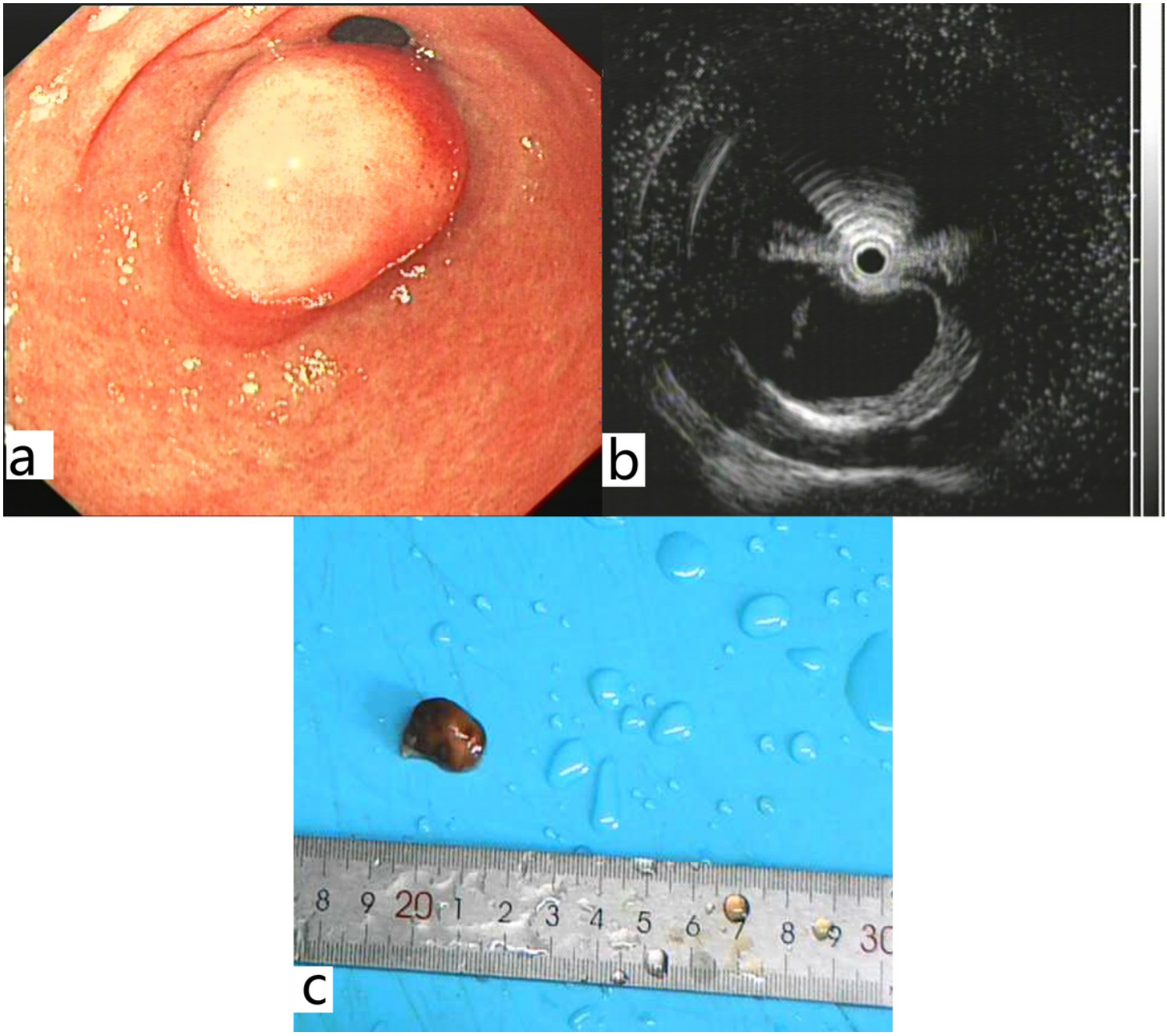

Figure 1

A: White light endoscopy revealed a hemispherical lesion covered with smooth mucosa located in gastric antrum. B: Endoscopic ultrasound revealed that it might be derived from submucosal layer, with no echogenic nodules, and a partition is visible inside. C: The resected specimen, and the mucosa was intact.

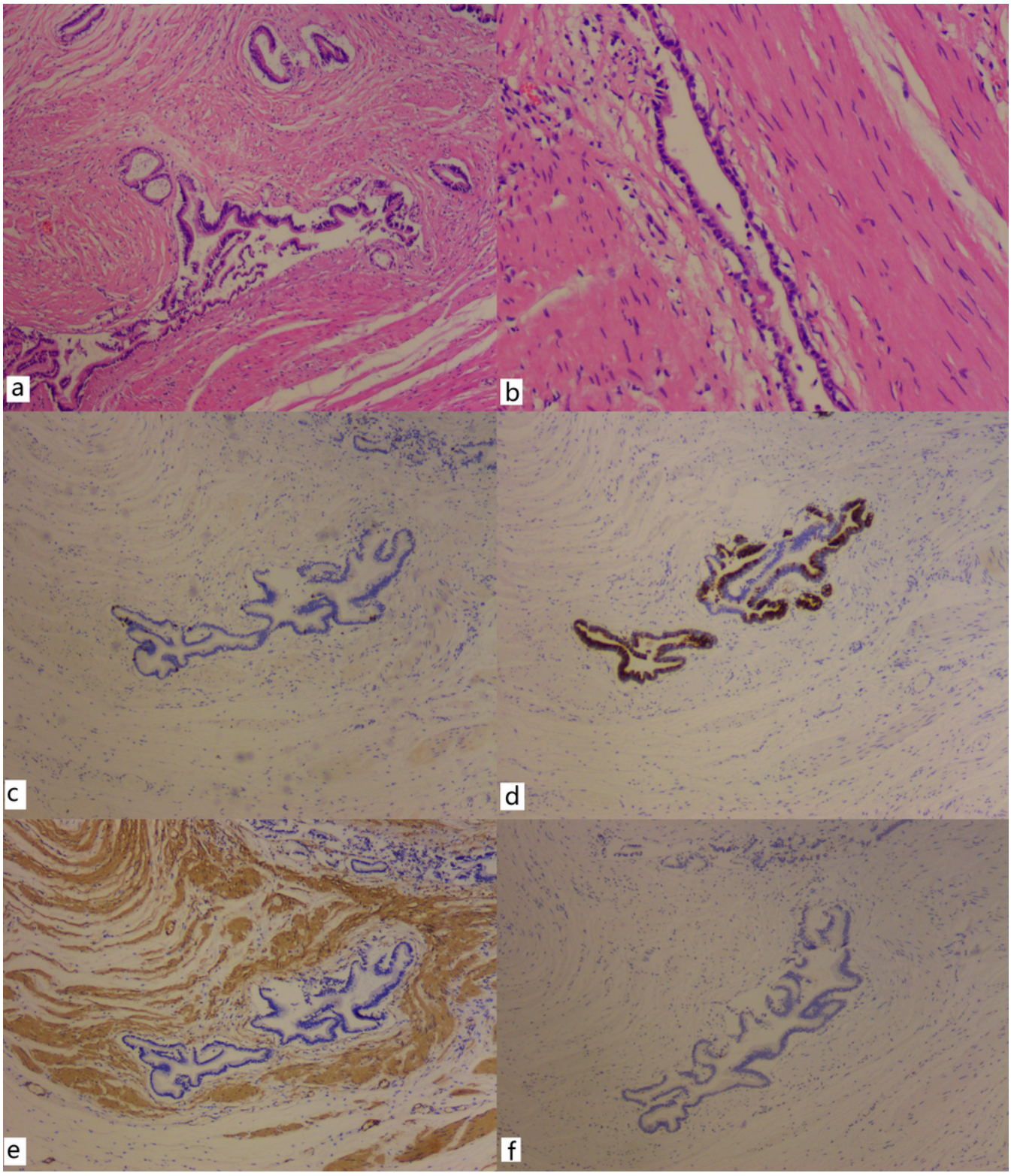

Figure 2

A: Heterotopic gastric glands located in the muscularis propria (H&E, ×40); B: Heterotopic gastric glands located in the muscularis propria (H&E, ×100); C: Positive immunohistochemical staining for Ki67; D: Positive immunohistochemical staining for MUC6; E: SMA positive in smooth muscle around heterotopic gastric gland; F: Negative immunohistochemical staining for MUC5AC.