A Rare Endoscopic Lesion; Gastritis Cystica Polyposa

Nadir Bir Endoskopik Lezyon: Gastritis Cystica Polyposa

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ABSTRACT

Gatsritis cystica polyposa is a rare nodular, polypoid lesion that usually develops at the gastroenteric anastomosis site after gastric surgery. The lesion usually has a strong vascular structure and it can also be a cause of occult or obvious upper gastrointestinal bleeding. This report of a 50-year-old man documents gastritis cystica polyposa developing at a gastroenteric anastomosis site.

Key Words: Gastritis cystica polyposa, Gastric surgery, Epithelial hyperplasia, Iron deficiency anemia, Endoscopy

ÖZ

Gastritis cystica poliposa, genellikle gastrik cerrahi sonrası gastroenterik anostomoz hattında nadir olarak görülen nodüler, polipoid bir lezyondur. Lezyon genellikle güçlü vasküler yapıda olup gizli ya da aşikar üst gastrointestinal sistem kanamasına yol açabilmektedir. Yazıda, 20 yıl önce peptik ülser hastalığı nedeni ile opere edilen 50 yaşında bir erkek hastada gastro-enterik anostomoz hattında gelişen gastritis sistika poliposa lezyonunu anlatılmaktadır.

Anahtar Sözcükler: Gastritis sistika poliposa, Mide cerrahisi, Epitelyal hiperplazi, Demir eksikliği anemisi, Endoskopi

INTRODUCTION

Gastritis cystica polyposa (GCP) is a rare inflammatory lesion of the gastric remnant that usually develops after partial gastrectomy. It is defined by the presence of polyps on anastomotic gastric mucosa, and by the presence of mucosal and submucosal cysts with foveolar hyperplasia on the histopathological examination (1). Littler and Gleibermann first reported GCP as a sessile polypoid lesion that occurred at the site of gastroenteric anastomosis (2). Since this first report, several case reports have been published in the literature for GCP. Recently, GCP was considered to be a precancerous lesion at the anastomosis site of the remnant stomach. Here, we present a new case with a giant polypoid lesion at the gastroenteric anastomosis site.

CASE REPORT

A 50-year-old man was referred to our gastroenterology department from another hospital due to his endoscopic findings. The patient had no history of smoking or alcohol intake but he had iron deficiency anemia findings. His hemoglobin value was 10.8 g/dl and his MCV value was 71 fl on complete blood count. Physical examination showed an old laparotomy scar that was caused by the gastrectomy operation but there was no palpable abdominal mass. Abdominal magnetic resonance imaging (MRI) and gastroscopy were planned as the

Received \ Geliş tarihi : 16.05.2017
Accepted \ Kabul tarihi : 07.08.2017
Elektronik yayın tarihi : 17.04.2018
Online published

DOI: 10.17954/amj.2018.131

first step. The lesion had a heterogeneous hyperintense view on T1-weighted images and it also had multiple milimetric high signal intensity foci on T2 weighted images with MRI. The lesion had intense peripheral enhancement and moderate central enhancement on dynamic contrast MRI. In addition, milimetric foci had no enhancement in the postcontrast phase of dynamic contrast MRI. This MRI finding was compatible with a cystic structure. Our gastroscopic examination showed a giant polypoid lesion at the greater curvature of the anastomosis site that was about five centimeters in diameter (Figure 1). We decided to perform partial polypectomy to obtain accurate biopsy material and performed superficial polypectomy after the sclerotherapy with adrenaline around the anastomosis site. The lesion showed massive arterial bleeding despite sclerotherapy. We used four hemoclips and succeeded in stopping the massive bleeding. The patient was discharged without another clinical problem after one day of hospitalization. On the pathology examination, the lesion grossly resembled a hyperplastic polyp. Its base was broad and it was located near the gastroenteric anastomosis site. The lesion contained foveolar or glandular epithelium in the submucosa or muscularis propria and also submucosal multiple cysts surrounded by disorganized smooth muscle. There was increased mixed inflammatory cells in the lamina propria. Due to these findings, the pathological report was GCP (Figure 2,3). We prescribed high dose proton pump inhibitor and also high dose sucralfate treatment for four weeks to the patient and then performed a second upper gastrointestinal endoscopy. The lesion had shrunk and nearly vanished. We took multiple biopsies from the polypectomy area for follow-up pathology assessment, which showed normal findings. The patient was consulted with the radiology and general surgery departments with his endoscopy, pathology and abdominal MRI findings for re-operation or follow-up. We decided to monitor the patient by gastroscopy at quarterly intervals

DISCUSSION

GCP is a rare, idiopathic, polypoid endoscopic lesion (1). Littler and Gleibermann first reported GCP as a sessile polypoid lesion that occurred at the site of gastroenteric anastomosis (2). Since this first report, several case reports have been published in the literature for GCP and various names such as gastritis cystica polyposa (2), diffuse submucosal cysts (3), submucosal heterotropic gastric glands (4), and stomal polypoid hypertrophic gastritis (5) have been used. In 1963, before the first report of GCP in 1972, a unique gastric disorder with the same histological features of GCP as heterotopic submucosal cysts was described in an unoperated stomach (6). With this report, GCP was defined in an unoperated stomach for the first time and other reports followed it. It has been suggested that GCP

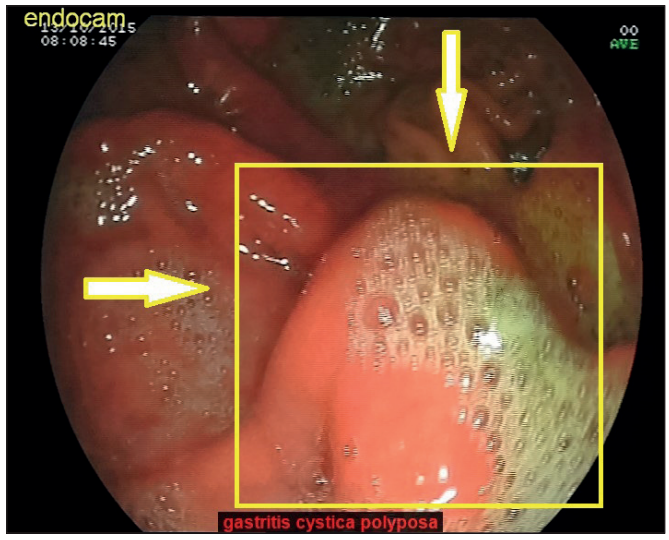

Figure 1: Endoscopic appearance of the lesion.

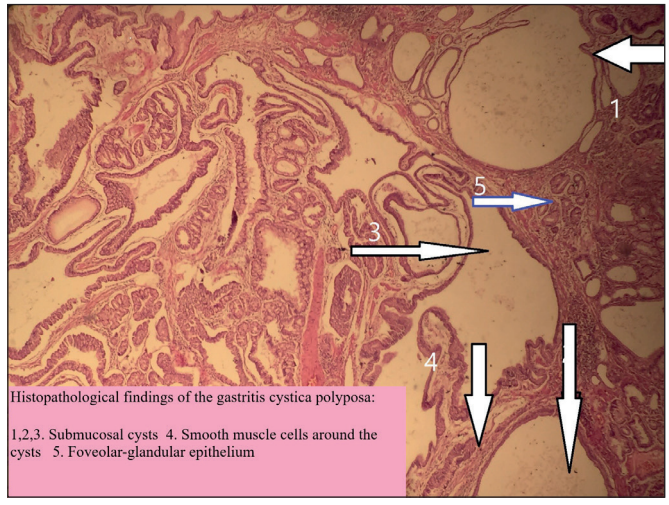

Figure 2: Histopathological findings of the lesion.

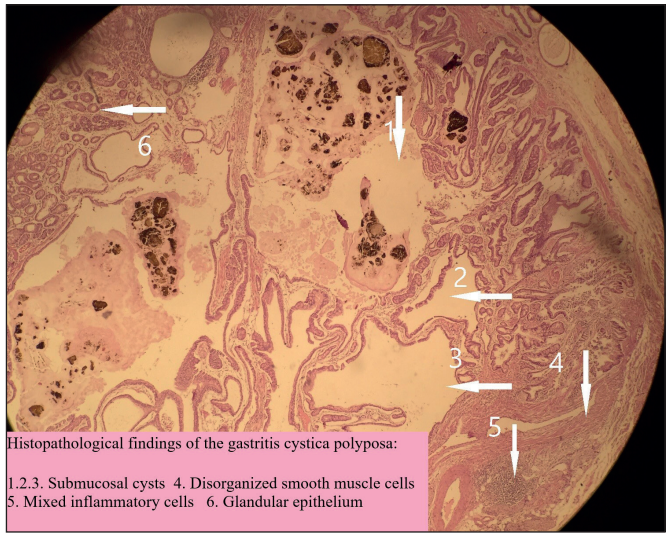

Figure 3: Histopathological findings of the lesion.

is a precancerous lesion, especially when it develops on the gastroenterostomy site. Although the pathophysiology of GCP is not completely known, it is thought to develop due to chronic inflammation, foreign body reaction and ischemic injury of the stomach. Some interruptions of the muscularis mucosa allows migration of epithelial cells to the submucosal layer and subsequent cystic dilatation.(7,8). Mucosal prolapse and duodenal reflux seen after gastric surgery appear to be the predisposing factors for GCP development. Although 65% of the cases in the literature occurred in patients with a history of gastric surgery, GCP can also develop in unoperated patients (6,8). The pathogenesis of GCP in the unoperated stomach is not clear. It has generally been assumed to be of congenital origin, mainly because of the lack of documented prior gastric ulceration or trauma, but it can also develop in consequence of the inflammation in the unoperated stomach. Thus, further studies on the pathogenesis of GCP in an unoperated stomach are necessary (7,8).

The macroscopic appearance of the GCP is similar to hyperplastic polyps (HP). It is characterized by elongation of the gastric foveola with hyperplasia and cystic dilatation of the gastric glands extending into the gastric submucosal layer. GCP in an unoperated stomach has generally been assumed to be of congenital origin, mainly because of the lack of documented prior gastric ulceration or trauma (9). GCP usually cannot be discriminated from inflammatory fibroid polyp and hyperplastic polyp on pathology examination. It is not yet clear whether GCP develops from inflammatory fibroid polyp or HP, or arises independently (10).

GCP has a large differential diagnosis spectrum. The differential diagnosis of a submucosal GCP includes hyperplastic polyp, inflammatory polyp, gastric adenocarcinoma, gastrointestinal stromal tumors (GISTs), neuroendocrine tumors, inflammatory myofibroblastic tumors, schwannomas, heterotopic pancreas, lipomas, sarcomas, cysts, lymphomas, and leiomyomas (11).

Clinical manifestations of GCP are variable and include abdominal pain, occult or obvious gastrointestinal bleeding, anemia, abdominal mass and occasionally gastric outlet obstruction .Endoscopic ultrasound (EUS) can be helpful for the diagnosis. Multiple homogeneous hypo-anechoic cysts in the submucosa are the most frequent EUS finding of GCP. Heterogeneously enhancing polypoid lesions with cystic components should also raise suspicion for GCP (11-13).

Dysplastic changes observed in submucosal glands, especially in selected cases of GCP, suggest that the lesion may be the precursor of malignancy. Mitomi et al. found increased expression of Ki-67, p53 and p21 in GCP lesions as a marker of increased epithelial proliferation and increased DNA repair that could be linked to the malignant progression (8,14). Furthermore, there is another study about the parietal cell K channels subunit called KCNE2. In this study, it was determined that targeted deletion of KCNE2, a subunit of K channels in parietal cells, in mice, and associated with increased rates of GCP and KCNE2 disruption was associated with increased risk of gastric neoplasia (15).

Compared to gastric adenocarcinoma, GCP occurs in younger male patients and rarely presents with weight loss or abdominal complaints. In regards to distinguishing GCP from GIST, GCP develops in a similar age range, but is almost four times more common in men, and only rarely associated with iron deficiency anemia or gastrointestinal bleeding (16%) compared to GIST (72%) (8).

Gastroscopic biopsy materials usually include superficial mucosal tissues. Because GCP is develops in the submucosal layer, superficial endoscopic biopsy materials are generally inadequate for diagnosis. Thus, GCP diagnosis usually requires EUS fine needle aspiration biopsy (EUS-FNA), endoscopic polypectomy or endoscopic submucosal dissection (ESD) procedures (15). Some of these cases may go to surgery for definite diagnosis despite wide polypectomy or ESD. The combined use of EUS and ESD seems to be necessary to avoid unnecessary surgical interventions in GCP (7,8,11,12).

There have been assertions that Epstein-Barr virus (EBV) may be associated with GCP in recent years (16). Choi MG et al. found that the EBV-positive rate was significantly higher in a GCP gastric cancer group (31.1%) than in a non-GCP gastric cancer group (5.8%). This suggests that GCP is closely related to EBV-associated gastric cancers and that EBV infection may play a role in dysplastic changes associated with GCP (17).

There have been no definitive reports describing the relationship between GCP and Helicobacter pylori but in a recent study H. pylori was found to be associated with GCP, gastric ulcer and focal dysplasia in Mongolian gerbils. Furthermore, the H. pylori cag-pathogenicity island-dependent immunological response may trigger GCP according to the same report (18). In our case, the patient had no proton pump inhibitor usage history and the Helicobacter pylori test was negative.

Screening for GCP is controversial. There is no clear consensus on the lesion as GCP does not have a pathognomonic, endoscopic or radiographic appearance (8). The common recommendation is endoscopic follow-up of the lesion at 3-6 month intervals (19).

GCP may have serious vascular structure. There have been some GCP cases recognized after melena and anemia in the literature as in Littler and Gleibermann's case in 1972. In our case, we saw acute massive arterial bleeding after the polypectomy procedure.

In conclusion, this case emphasizes that it is important to keep GCP in mind as one of the reasons of intermittent or occult upper gastrointestinal bleeding in patients who have a history of gastric surgery and anemia. GCP could also be a predisposing factor for gastric adenocarcinoma.

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