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Gastritis cystica profunda: A challenging disease diagnosed using a novel approach

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Abstract

Gastritis cystica profunda (GCP) is a rare hyperplastic lesion of unclear etiology. Clinical symptoms of GCP are variable and range from nonspecific abdominal pain to gastric outlet obstruction. Often the diagnosis of GCP has been difficult to make as simple esophagogastroduodenoscopy (EGD) guided biopsy can fail to yield the diagnosis. Here, we report a case of GCP that presented in a challenging anatomic location, which required a laparoscopic intragastric surgical technique to make the diagnosis.

Keywords

gastritis cystica profunda; gastric glands; submucosal tumors

Introduction

Gastritis cystica profunda (GCP) is a rare, benign disease characterized by polypoid hyperplasia and cystic dilatation of the gastric glands that extend into the submucosa of the stomach. First described in 1947 by Scott and Payne, it wasn't until 1972 that Littler and Gilbermann suggested that the presence of cystically dilated gastric glands in the submucosa was a reactive, postsurgical condition for which they coined the term “gastritis cystica polyposa” [1]. This would eventually change to the now preferred term “gastritis cystica profunda” because it resembled the similarly named condition in the colon [1]. Clinically, patients can present with upper abdominal pain, acid reflux, nausea, anorexia, or bleeding; although some patients may experience no symptoms [2]. In severe cases, patients can experience massive upper gastrointestinal hemorrhage and gastric outlet obstruction [2]. GCP is often seen as giant gastric folds, submucosal tumors, or isolated polyps [2]. The lesion has been described primarily in the operated stomach, leading to the hypothesis of prior gastric wall injury as the predominant cause of GCP, [2] although it has been described in the unoperated stomach as well. Although the exact pathogenesis mechanism remains largely unknown, GCP has been found to be associated with ischemia and chronic inflammation [3]. It is also thought to be a possible precancerous lesion since a few early gastric cancers have been associated with it [4]. Interestingly, there is also an increased association in cancers with GCP to Epstein-Barr virus (EBV) positive patients [5] and Menetrier disease (MD) [6]. Here, we describe a case of a difficult to diagnose gastric mass that was found to be GCP.

Case Presentation

Our patient was a 72 year old male with a past medical history of a non-small cell lung cancer

treated with chemotherapy and radiation, coronary artery disease, and cardiomyopathy. Previous esophagogastroduodenoscopy (EGD) performed at an outside facility in February of 2014 for symptoms of reflux showed circumferential Barrett's esophagus and a small to moderate hiatal hernia. At this time, he was referred to our institution for continued surveillance of the Barrett's esophagus.

In October of 2015, a surveillance EGD showed a new mass in the gastric cardia measuring 2.6 x 1.4 cm. Fine needle aspiration (FNA) performed at this time was nondiagnostic. The mass was located near the hiatal hernia pinch and was thought to represent a gastrointestinal stromal tumor (GIST) or leiomyoma. Approximately 6 months later, an interval EGD also an increase in the size of the gastric cardia mass to approximately 4 cm (Figure 1). On endoscopic ultrasound (EUS), a heterogeneous mass measuring 3.9 by 3.1 cm in the gastric cardia with small internal cystic spaces was described. He underwent a second FNA of the mass, which showed only inflammation and again failed to yield a diagnosis. A CT scan showed a smooth well marginated mass in the region of the gastric cardia and hiatal hernia, suggestive of a submucosal location (Figure 2).

Given the concern for neoplasm based on its increase in size, surgical resection was recommended. Given the challenging location of the lesion close to the GE junction, a laparoscopic intragastric approach with endoscopic assistance was recommended. This is a novel approach whereby the laparoscopic ports are inserted through the abdominal wall and then into the stomach under direct endoscopic visualization. During the procedure upon manipulation of the mass, purulent drainage was noted from the area of the mass, which resulted in collapse of the mass. Since the mass decompressed, we decided not to resect it. There was no evidence of malignancy on previous biopsies. Thus, the justification for a more radical operation was not present. However, prior to completion of surgery multiple core needle biopsies of the mass were obtained using the laparoscopic intragastric approach. The patient tolerated the procedure well. He was discharged on postoperative day 3 without complications. Surgical pathology showed that the mass was consistent with gastritis cystica profunda (Figure 3).

Discussion

Gastritis cystica profunda is a condition characterized by benign, cystic growth of gastric glands into the submucosa of the stomach [7]. Although GCPs are thought to be associated with previous gastrectomy, this lesion can also occur without previous gastric operations, [8] as observed in our case. GCP is often located in the posterior or anterior wall of the gastric body and in the intermediate zone between the fundic and pyloric glands [9]. In general, laboratory tests are not useful in making a diagnosis of GCP [3]. The pathogenesis of GCP is likely due to chronic ischemia and inflammation. The disruption of integrity of muscularis mucosa causes the migration of epithelial content into the submucosa with subsequent atrophic gastritis, intestinal metaplasia and cystic dilatation of gastric glands [7]. Gastritis cystica profunda is a benign lesion, although a possible precancerous nature has been hypothesized [10].GCP has been shown to occur more frequently in the presence of gastric cancers [11]. In a pathological study of 10,728 patients with gastric cancer, GCP was found in 161 patients [11].

There appears to be a pathogenetic role of Epstein-Barr virus (EBV) in the development of GCP and cancer [8]. It has been demonstrated that there is a delay of apoptosis in EBV-positive gastric carcinomas associated with up regulation of BCL-2 and p53, and a decrease in cellular differentiation associated with

a decrease in E-cadherin expression [8]. EBV infection can alter the normal cell cycle, resulting in disruption in the cellular processes and checkpoints responsible for regulating cell division, which then results in the carcinogenetic effects of EBV [8]. EBV titers have been shown to be significantly higher in dysplastic gastric mucosa versus non-neoplastic gastric mucosa. In a study done by Kim et al, the authors showed that within the transitional area between GCP and gastric carcinoma, there were dysplastic changes positive for EBV [8]. Another group 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%), which suggests that GCP was significantly associated with EBV-positive gastric cancers and that EBV infections may play a role in the dysplastic changes associated with GCP [11].

Menetrier disease (MD) is another premalignant condition which has shown some association with GCP [12]. MD is an uncommon, idiopathic, hyperplastic gastropathy characterized by hyperplasia of foveolar mucous cells, which results in thickening of gastric folds and hypoalbuminemia [6]. It mostly involves the gastric fundus and body [6]. There have been many reports showing close association with MD and GCP as potential precancerous lesions [6]. Further work-up includes testing for cytomegalovirus and H. pylori. Treatment options include proton pump inhibitors (PPI) or H2 blockers, octreotide, monoclonal antibodies to epidermal growth factor receptor (EGFR), and gastrectomy [6].

Regarding the diagnosis of GCP, standard diagnosis by EGD is often difficult because the standard FNA biopsy specimen is usually limited to the mucosa, which cannotprovide sufficient information about the deeper submucosa [13]. In fact, in many cases the preoperative diagnosis of GCP remains challenging despite the current advances in endoscopic techniques and thus patients may have to undergo surgical resection for final pathologic diagnosis to guide treatment [13]. Often times and in our case, CT scan and EUS have been used as a complementary tool to delineate additional characteristics of these lesions [13]. The most common endoscopic features of GCP by conventional white-light endoscopy are nonspecific [3]. In fact, many gastroenterologists now suggest that the diagnostic modality of choice is EUS [3]. GCP on EUS shows primarily 3 major echoic patterns: anechoic (35.3%), mixed heterogeneous with thickened overlying mucosa(50%), and hypoechoic with microcysts (14.7%) [3]. The final diagnosis however still has to be determined by histological exam [3].

There is currently no consensus on the optimal GCP treatment. Due to an insufficient amount of information on GCP, there have been a variety of treatment recommendations, ranging from observation to radical resection [3]. Xu G et al developed a standard protocol to systemically investigate these lesions with EUS before endoscopic submucosal resection/endoscopic submucosal dissection (EMR/ESD) [3]. Given the fact that the diagnosis of GCP mainly relies on histopathology, endoscopic resection serves both a diagnostic and therapeutic procedure. If performed successfully, endoscopic resection with EMR or ESD is less invasive, safer, and more economical than open surgical methods [3]. More importantly, endoscopic resection of GCP better preserves gastric function with minimal injury [3]. It should be noted there were limitations to their proposed protocol including small sample size, single institution experience, and retrospective nature of the study.

In our case report, we presented a patient whose lesion was found to be in a challenging proximal anatomic location associated with a hiatal hernia. It was in close proximity to the GEJ, thus making it unresectable through a standard laparoscopic approach. Thus, a laparoscopic intragastric approach with

endoscopic assistance was attempted. However, the mass decompressed and was no longer amenable to resection. Two previous endoscopic-guided FNAs failed to establish the diagnosis. Therefore, we performed core biopsies through the intragastric port, which successfully made the diagnosis. This approach was found to be diagnostically effective and had the advantage of staying minimally invasive. In conclusion, GCP is a rare and often difficult diagnosis to make, but in certain cases such as ours the diagnosis can be facilitated using a novel surgical method consisting of a laparoscopic intragastric approach with endoscopic assistance.

Figures

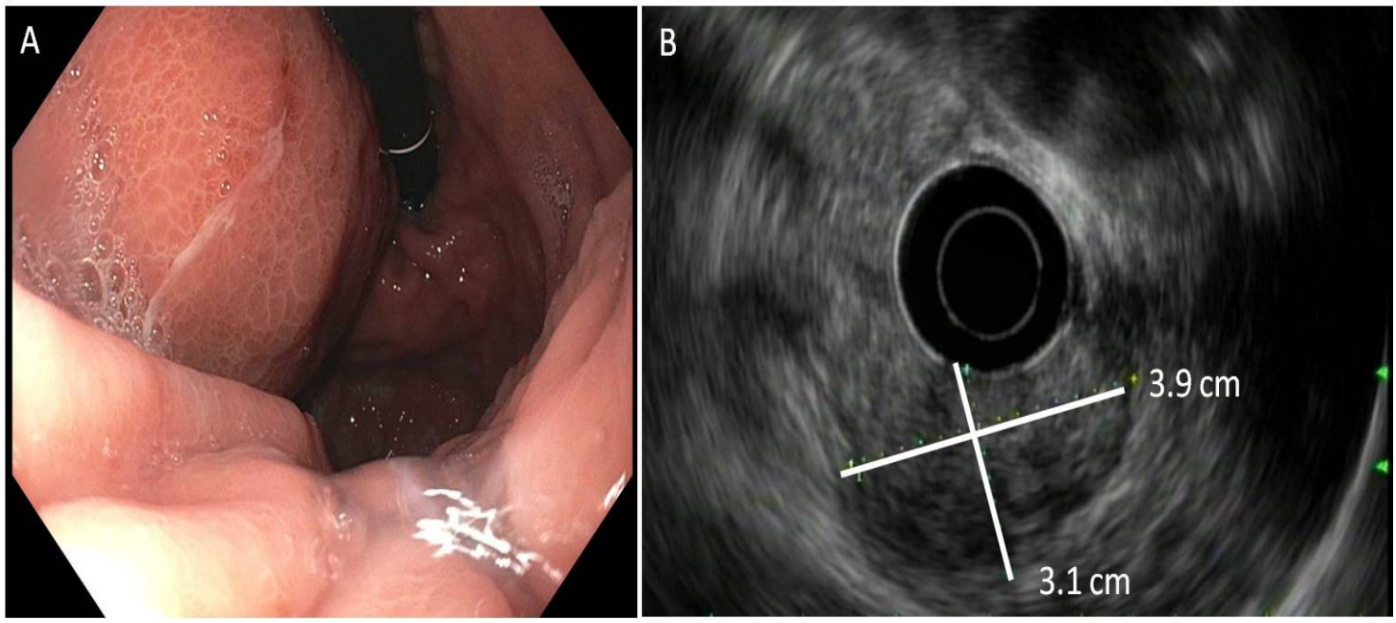

Figure 1: (A) EGD showed a 4 cm subepithelial mass in the gastric cardia at the hiatal hernia pinch located at 40 cm. (B) On limited EUS, there was the heterogeneous mass measuring 39 mm by 31 mm in the gastric cardia with small internal cystic spaces.

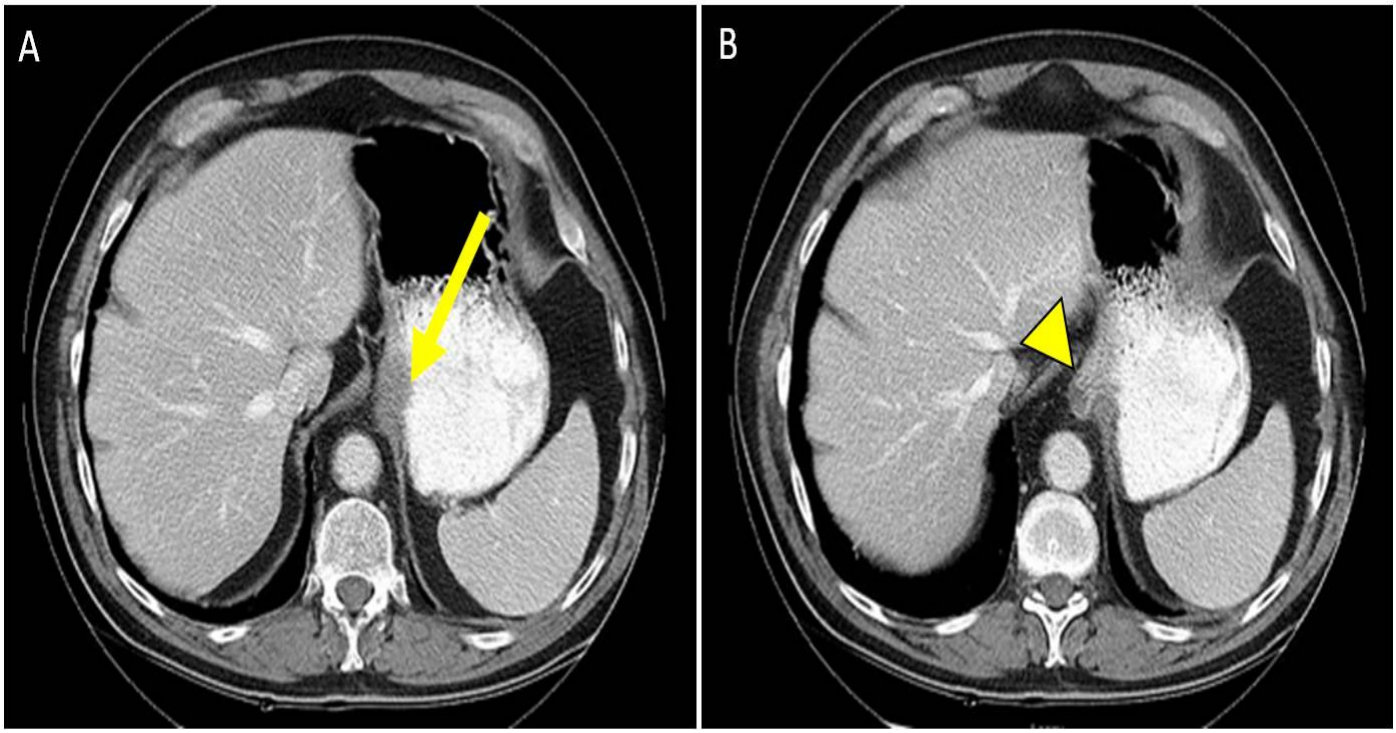

Figure 2: (A) The patient CT scan showeda 3.5 x 1.7 cm smoothly marginated mass adjacent to the gastric wall just beyond the GE junction in the gastric cardia (arrow). (B) There was also a hiatal hernia (arrowhead). Differential diagnosis included gastrointestinal stromal tumor (GIST) and leiomyoma.

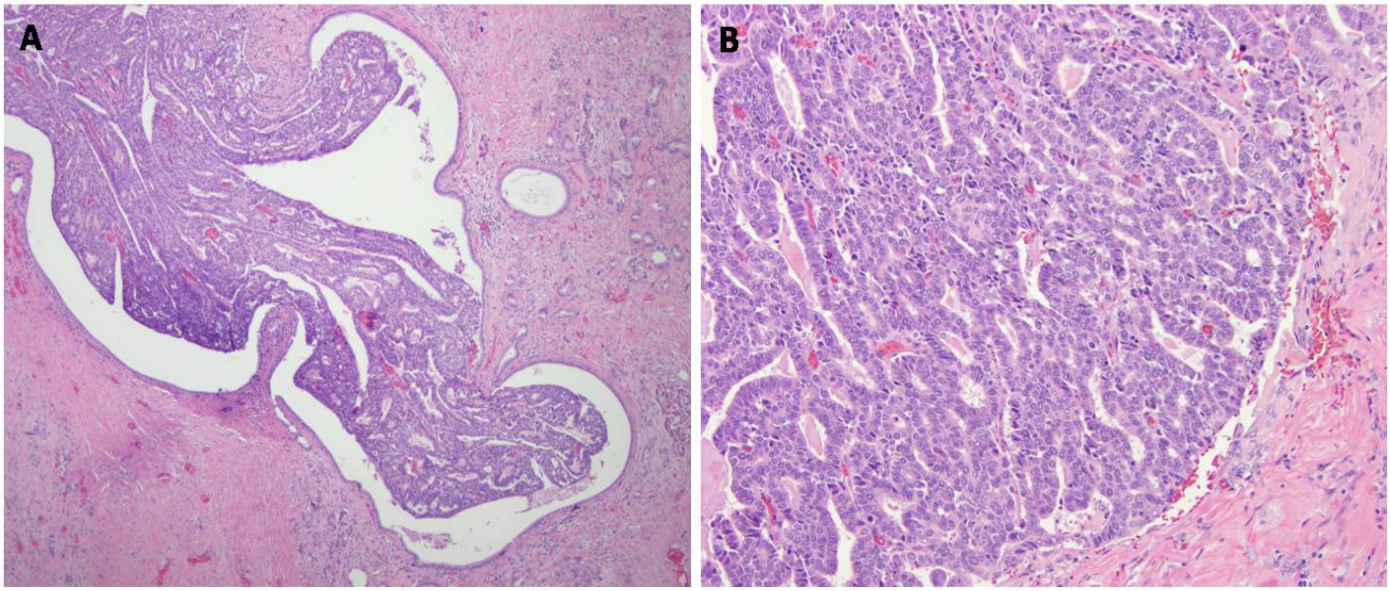

Figure 3: (A) Low power 40x and (B) High power 100x magnification. The laparoscopic intragastric core needle biopsy showed fibrous core tissue with islands of benign glandular cells with variable degree of chronic inflammation. There was no neutrophilic infiltration to suggest acute inflammation. These findings were consistent with gastritis cystica profunda.

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