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CASE REPORT | ENDOSCOPY

Pyloric Gastric Adenoma: Endoscopic Detection, Removal, and Echoendosonographic Characterization

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ABSTRACT

Pyloric gland adenomas (PGAs) are rare neoplasms found not only in the gastrointestinal tract but also in other extragastrointestinal organs. They have potential for malignant conversion, and early detection and removal is imperative to prevent invasive disease. PGAs prove difficult in management and surveillance given their rarity. However, increasing familiarity with histological appearance and use of advanced tools such as echoendosonography can bring greater understanding of their clinical history. We describe a unique case of a PGA detected within a hiatal hernia sac characterized with echoendosonography and highlight the need to develop surveillance protocols for these types of lesions.

KEYWORDS: pyloric gland adenoma; echoendosonography; gastric polyps; surveillance

INTRODUCTION

Pyloric gland adenomas (PGA) are rare precancerous neoplasms that exhibit gastric pyloric gland differentiation. They present a diagnostic challenge for the North American endoscopy community because they account for roughly 2% of all gastric adenomas.[1] Correct diagnosis is important because they have a reported advancement from low-grade intraepithelial neoplasia to adenocarcinoma at a carcinogenesis rate of 12%–47%.[1] Arising in chronically inflamed mucosa, they are gastric predominant but have also been found in the gallbladder, duodenum, bile duct, pancreas, and the esophagus.[2] We present an exceedingly rare case of a female patient with gastroesophageal reflux disease (GERD) who have concern for a subepithelial lesion within a hiatal hernia sac with resection revealing a PGA.

CASE REPORT

A 62-year-old woman with a medical history of rheumatoid arthritis, anemia, and GERD presented as a referral for endoscopic ultrasound (EUS) after she was found to have a 10-mm esophageal subepithelial nodule on a previous esophagogastroduodenoscopy. She had an unremarkable physical examination, vital signs, and laboratory studies.

The repeat upper endoscopy performed revealed a small hiatal hernia and distal grade A esophagitis. In the stomach, diffuse erythema was compatible with gastropathy. A single-sessile 10-mm polyp of benign appearance was detected within the hiatal hernia sac on the gastric side of the gastroesophageal junction (GEJ) (Figure 1). EUS was performed and revealed a thickened mucosal layer, however no discrete lesions (Figure 2). As the lesion was predominantly on the gastric side, it was able to be removed safely with a hot snare polypectomy and no additional lift method was necessary. Histologically, the polyp revealed glandular proliferation of back-to-back mucinous glands, consistent with a pyloric adenoma with no signs of dysplasia (Figure 3). She was advised to return in 12 months for a surveillance endoscopy.

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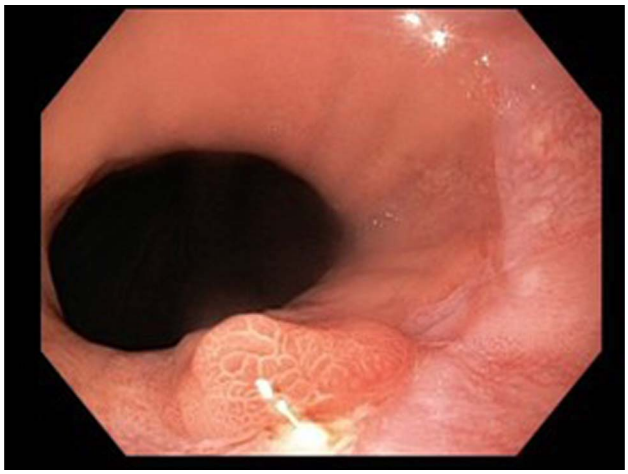

Figure 1. Upper endoscopy view of the pyloric gland adenomas found within the hiatal hernia on the gastric side of the gastroesophageal junction.

Figure 2. Endoscopic ultrasound (radial 10 mHz) image taken at the region of the polyp before excision revealing a thickened esophageal wall but no clear mucosal/submucosal lesion.

DISCUSSION

We present a highly unusual case of a PGA found within the hiatal hernia sac of a patient with a history of chronic GERD. On our review of the English literature, less than 1,000 cases of PGAs have been reported since originally described in 1976.[3] Esophageal/GEJ PGAs like that of our patient are particularly rare, with less than 20 reported cases, none of which seem to have been detected within a hiatal hernia sac. In Table 1, we summarize several of the largest case series of PGAs available and their regions of origin.

Detection of PGAs is of key importance because they carry an increased risk of malignant transformation. Histologically, PGAs are composed of back-to-back cuboidal to low columnar epithelial cells, with pyloric gland appearance, and eosinophilic ground-glass cytoplasm. Their recognition as a pathologic lesion in the United States was delayed until 2009 with the publication of a large case series by Chen et al.[5] This case series was able to corroborate characteristics of PGAs previously noted in European and Asian studies, including predominance in female patients, prevalence in gastric body, and association with autoimmune gastritis. The authors also discussed that the delay in recognition of PGAs in North America was at least partly due to pathologists' relative unfamiliarity with the full range of gastric neoplasms compared with pathologists in other regions in the world. Not only is this due to the overall reduced incidence of gastric neoplasms in North America but also due to skewed detection frequency, given the Asian Pacific region still conducts the largest volume share of upper endoscopies globally at 43.1%.[9,10] Grossly, PGAs appear as polypoid lesions, although some can be irregular mucosal, flat, ulcerative, or even submucosal tumor-like lesions.[1] With the increasing availability of EUS, locoregional characteristics for these lesions can now be studied as well. We were able to identify 6 other studies that used EUS for characterization and removal of PGAs (Table 2).

PGAs, as a subtype of adenomatous polyps, should be resected. Generally, adenomatous polyps are considered to have malignant potential and are recommended to be surveilled. In the United States, the American Society of Gastrointestinal Endoscopy recommends repeat endoscopy 1 year after adenomatous polyp resection, followed by surveillance endoscopy

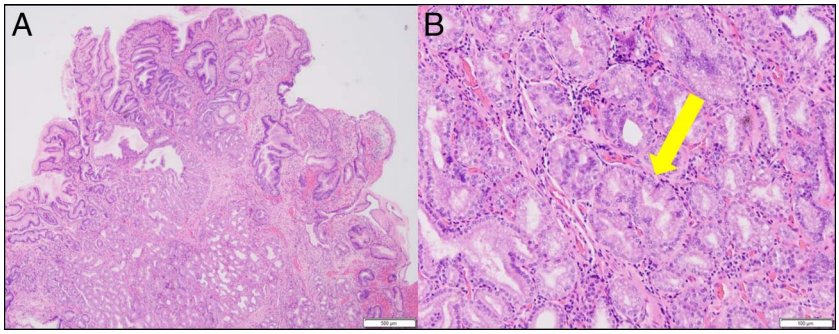

Figure 3. (A) A low-power view of the polyp shows a polypoid fragment of stomach mucosal epithelium with glandular proliferation. (B) A high-power view of the interior of the polyp shows back-to-back mucinous tubular glands surrounded by a monolayer of small round nuclei without noticeable atypia, consistent with a pyloric adenoma. Although many pyloric adenomas can have dysplastic features, such as cribriform formation, with nuclear crowding, nuclear enlargement, loss of nuclear polarity, enlarged nucleoli, and/or hyperchromasia, this polyp did not. Yellow arrow points to back-to-back mucinous glands.

Table 1. Case series of PGAs and their respective regional origin

Author/yr	Cases	Mean age (yr)	Sex (F:M %)	Site	Region of origin
Vieth 2003[4]	90	73	61:39	Gastric	Europe
Chen 2009[5]	41	73	70:30	Gastric	North America
Choi 2018[6]	67	66	52:48	Gastric gastroesophageal junction	North America, Europe, Asia
Vieth 2010[7]	60	71	50:50	Gastric	Europe
Miller 2020[8]	57	74	63:37	Duodenum	North America/Europe

F = female; M = male; PGA, pyloric gland adenomas.

every 3–5 years, although there is unrobust evidence to support this interval.[17] Adjunctively, the background mucosa of patients with PGAs are often found to have gastric intestinal metaplasia, which in isolation does not warrant surveillance, but does justify interval surveillance when grouped with other high-risk features. The length of surveillance is unknown but should take into consideration the relative cancer risk of the individual by weighing several factors including family history of gastric cancer, migrant status from high incidence area, Operative Link on Gastritis Assessment stage 3–4, and history of familial polyposis syndrome.[18]

In 2 multicenter clinicopathological studies of PGAs, the 21 pooled cases of intramucosal/invasive adenocarcinoma were associated exclusively with histological findings of high-grade dysplastic PGAs. This finding really underscores the risk of progression through the stepwise carcinogenic sequence of low-grade dysplasia, high grade, and adenocarcinoma, making resection of these precancerous lesions with curative intent imperative to curb the natural progression to malignancy.[6,8] In addition to the individual cancerous risk of PGAs, the common backdrop of atrophic gastritis and gastric intestinal metaplasia surrounding these lesions may make it reasonable to use long-term surveillance intervals.

In our case, the initial reason for referral was the subepithelial appearance of the polyp. EUS was used and useful for evaluating the originating mucosal layer, echogenicity, vascularity, size, and absence or presence of adjacent lymph tissue.[19] Knowing whether the neoplasm was of mucosal or submucosal origin helped with strategizing the ultimate resection technique, that is, standard single-piece polypectomy, endoscopic mucosal resection, or endoscopic submucosal dissection. There have been no reports that specifically compare the recurrence rates of PGAs based on the removal technique used. Local recurrence is low, 10% on 3-year follow-up in 1 multicenter study, and similar despite high-grade or low-grade dysplasia.[8] This low recurrence makes local resection with endoscopy appropriate, provided that the lesion can be completely resected.

Currently, the focus on PGAs is mostly on the histological grade relative to their evolution to adenocarcinoma. EUS adds an additional diagnostic component to endoscopy by elucidating the depth of mucosal invasion and thus delivers information about staging. With the increasing availability of EUS, we propose that topographical information and locoregional staging should be included along with histological grading in the development of longitudinal surveillance algorithms for gastric lesions such as PGAs.

In conclusion, our case contributes to the ongoing effort to build a more robust North American database of PGAs. We were able to highlight an unusual occurrence of this lesion at the GEJ within a hiatal hernia sac. This case highlights the need for robust evidence-based surveillance guidelines incorporating both histological and topographical characteristics of these lesions to build surveillance algorithms for prevention of malignant progression in this gastric polyp subset.

Table 2. Cases of PGAs characterized through echoendosonography

Author/yr	Cases	Age	Sex (M/F)	Site	Country
Park 2022[11]	1	86	M	Esophagus	Asia
Yamamoto 2018[12]	1	81	M	Gastric	Asia
Ichikawa 2021[13]	1	37	F	Gallbladder	Asia
Sooklal 2020[14]	1	—	—	Gastric	North America
Kim 2021[15]	1	52	F	Gastric	Asia
Min 2020[16]	1	69	M	Gastric	Asia

F = female; M = male; PGA, pyloric gland adenomas.

DISCLOSURES

Author contributions: A Liyen Cartelle, E. Holzwanger, S. Igbinedion, HJ Rosenberg, DK Pleskow: participated in design of the work, acquisition, analysis, or interpretation of data for the work. A Liyen Cartelle, E. Holzwanger: drafting of work. TM Berzin, MS Sawhney, M. Gabr, DK Pleskow: revised article critically for important intellectual content and provided final approval of the version to be published. DK Pleskow is the article guarantor.

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Informed consent was obtained for this case report.

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