Early gastric cancer associated with gastritis cystica polyposa in the unoperated stomach treated by endoscopic submucosal dissection

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Gastritis cystica polyposa (GCP) is an uncommon hyperplastic benign lesion, which is histologically characterized by hyperplastic foveolar epithelia and multicystically dilated glands inside the lesion. GCP is also called gastritis cystica profunda when the cystic lesion locates within the submucosa. GCP was at first described as polypoid tumors developed in the gastric mucosa adjacent to Billroth-II stoma.[1] This relatively rare lesion usually develops in patients who previously underwent gastroenterostomy, especially for treatment of a benign lesion, eg, gastric ulcer.[2-5] GCP is infrequently found in an unoperated stomach.[6-8] Because GCP is usually found in the operated stomach, its etiology and pathogenesis have been considered to be chronic inflammation after surgical treatment. Mucosal prolapse or duodenogastric reflux of intestinal contents into the gastric remnant may be inferred to be a cause of GCP.[4,9,10] Subsequent reports brought up some similar findings in various synonyms, and some of the cases reported were with accompanying carcinoma.[2,11,12] However, the mechanism of GCP developing in the unoperated stomach is unclear.

In some cases reported, it was suggested that GCP at a gastroenterostomy site may be a precancerous lesion because carcinoma is frequently found at old gastrojejunostomy stoma and the histologic features of GCP resemble the experimental stomal polyps preceding carcinoma after partial gastrectomy in rats.[13-15]

Herein, we report 2 cases of early gastric cancer (EGC) that accompanied GCP, which occurred in patients who had never undergone stomach surgery. The patients were treated with the endoscopic submucosal dissection (ESD) technique. The carcinoma cells were confined to the mucosal layer.

CASE REPORTS

Case 1

A 73-year-old woman visited our clinic for further evaluation of multiple gastric polyps and a large protrusion of mucosa, which was found at another clinic while performing an upper-GI radiographic contrast study as a periodic medical checkup. At our clinic, a large, 3 × 2.5-cm, nodular elevation was found at the posterior wall of the gastric antrum (Fig. 1A). The rapid urease test was positive. The biopsy specimen revealed tubular adenoma with focal carcinoma. Neither an enlarged lymph node nor distant metastasis was noted on CT. An EUS, performed to evaluate the depth of tumor invasion, revealed an inhomogeneous low-echogenic mass with underlying very small cystic lesions and intact submucosal layer (Fig. 1D). We clinically concluded that the tumor was confined to the mucosal layer, and ESD was performed (Fig. 1B and C). The specimen was found to be EGC of well-differentiated tubular adenocarcinoma, with underlying GCP, and the lesion was truly limited to the mucosal layer (Fig. 2). The resection margin was not involved with cancer cells, and there was no lymphovascular involvement. No complication was observed in the following endoscopy.

Case 2

This 66-year-old man was formerly diagnosed with EGC, well-differentiated adenocarcinoma, at another hospital. A follow-up endoscopy performed at our hospital showed a 1.5-cm, flat, depressed lesion at the anterior wall side of the upper portion of the body (Fig. 3A). The biopsy specimen obtained at our hospital also revealed well-differentiated tubular adenocarcinoma. A rapid urease test was negative. EUS showed an irregular mucosal layer (Fig. 3B). On CT, no cancer-like mass, enlarged lymph node, or distant metastasis was noted. Therefore, ESD was performed. The specimen from the stomach consisted of a 0.6 × 0.5 × 0.07-cm, pale-brown, soft, mucosal fragment. The specimen revealed EGC, with microscopic invasion to the mucosa lamina propria, which was associated with gastritis cystica profunda (Fig. 4). The resection margin was free of cancer cells. Lymphovascular involvement was not observed. There was no complication.

DISCUSSION

Herein, we describe 2 cases of EGC that accompanied GCP. To the best of our knowledge, this is the first description on gastric adenocarcinoma accompanied by GCP in

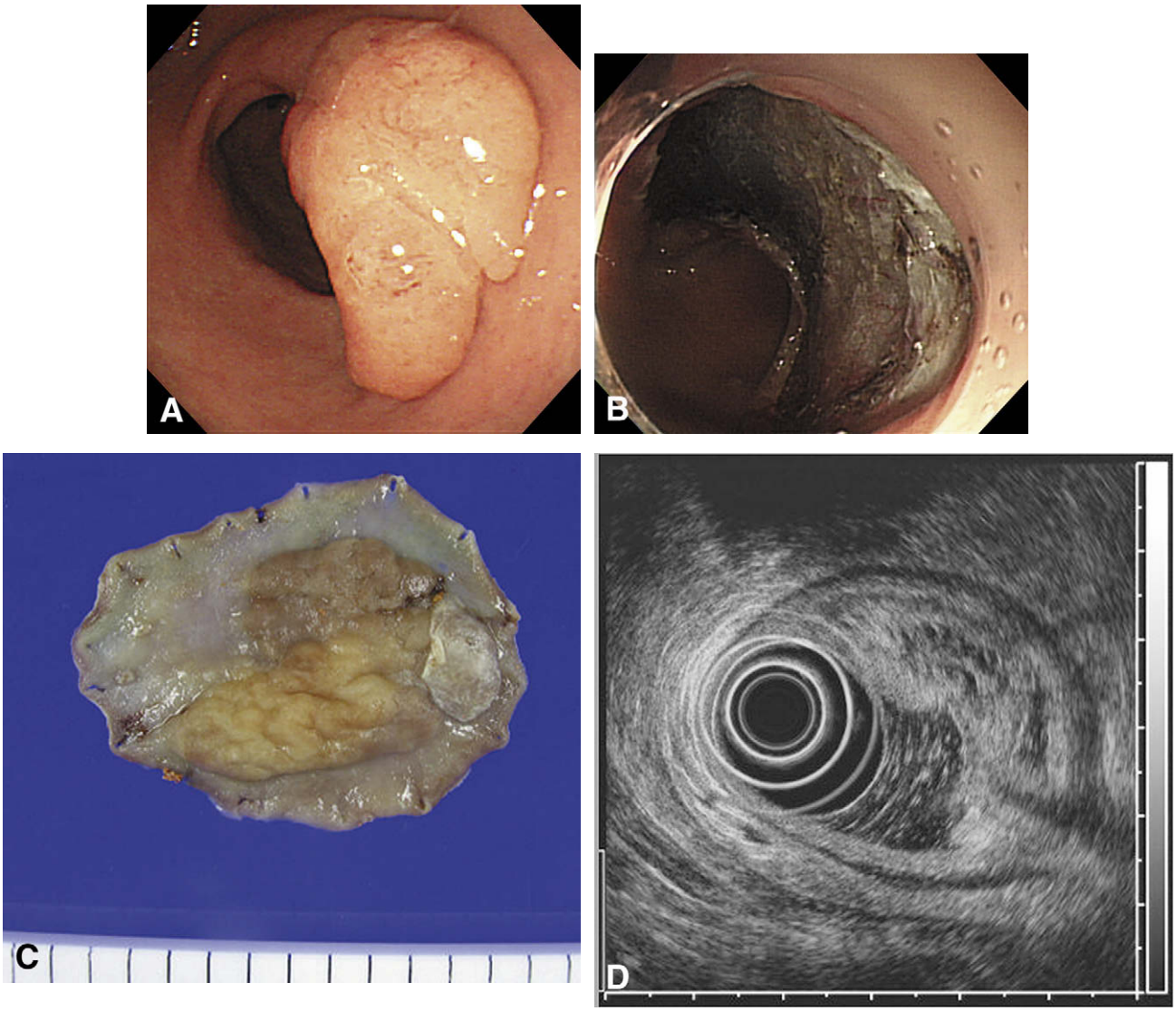

Figure 1. A, Endoscopic view of 3 × 2.5-cm nodular elevation at the posterior wall of the gastric antrum. **B,** Endoscopic view of the resected gastric wall. **C,** The specimen obtained by ESD (5-mm scale). **D,** EUS, showing inhomogeneous low-echogenic mass confined to the mucosa, with underlying multiple cysts.

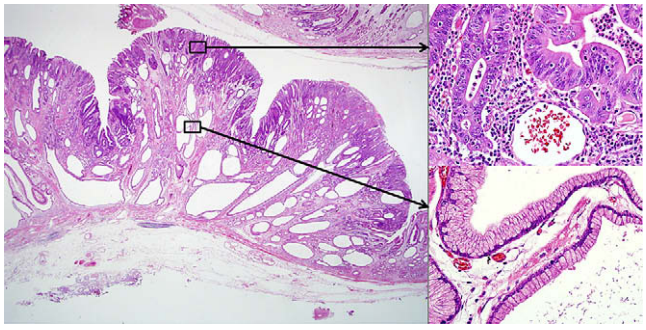

Figure 2. Microscopic photography, showing numerous submucosal cysts of various sizes, with well-differentiated hyperchromatic malignant cells in the mucosal layer (*right upper*) and hyperproliferated glandular cells lining the cystic portion (*right lower*) (H&E, orig. mag. ×12).

patients with no history of previous stomach surgery and was treated with endoscopic dissection, not polypectomy. The patients did not have any GI symptoms or any history of abdominal surgery, including gastrectomy. The pathologic confirmation of EGC was done during a routine medical checkup. Cystic echogenicity of GCP was easily identified by using EUS in both cases. The enlargement of lymph nodes and distant metastasis was not clinically evident, and the patients were treated with ESD, without further surgical treatment.

By using endoscopy, GCP usually shows large polypoid gastric folds or mass-like lesions protruding into the gastric lumen.[6-8] In CT findings, GCP appears to be an intraluminal polypoid mass.[7,8] These unusual findings might easily attract the endoscopist's interest. This would be advantageous for early diagnosis of EGC with GCP as in our cases and other previously reported cases.[13-15] In our first mentioned case, at first, the outgrowing appearance of the mass was suspicious of advanced gastric cancer. EUS