Case report

Asymptomatic gastric heterotopia in the rectum with *Helicobacter pylori* infection

Jarosław Swatek[1], Lech Wronecki[1], Roman Ciechanek[2], Justyna Szumiło[1]

[1]Department of Clinical Pathomorphology, Medical University of Lublin, Lublin, Poland
[2]Gastromed Ltd., Lublin, Poland

Gastric heterotopia is very rare in the rectum – less than 50 cases have been reported so far. Only in six of them *Helicobacter pylori* has been observed in heterotopic mucosa. We report a case of a 58-year-old woman with asymptomatic gastric heterotopia in the rectum, incidentally revealed during colonoscopy as a small, sessile polyp. The presence of *H. pylori* was confirmed by immunohistochemistry. This finding supports the opinion that *H. pylori* may pass along the gastrointestinal tract in a viable form and that the fecal-oral route of transmission is possible.

Key words: gastric heterotopia, rectum, *Helicobacter pylori*.

Introduction

Gastric heterotopia is defined as the presence of gastric oxyntic mucosa outside of the stomach and is generally regarded as a congenital malformation [1, 2, 3]. It should be distinguished from the gastric-type surface and foveolar epithelium and/or non-specialized antral-type mucosa that may occur in various parts of the digestive system in the course of various chronic inflammatory conditions, e.g. inflammatory bowel disease, duodenal peptic ulcer disease or chronic cholecystitis. These findings represent an acquired, reactive, adaptive change in the direction of cellular differentiation and therefore are regarded as metaplastic lesions [1, 2, 3]. The exception is Barrett esophagus, in which the presence of oxyntic mucosa is nowadays not interpreted as heterotopia, but as an example of metaplasia.

Gastric heterotopia is most common in the upper esophagus, duodenum and Meckel diverticulum, but can be found in all portions of the digestive tract. It is relatively often observed within intestinal duplications [1, 4, 5]. The location in the rectum is one of the rarest ones [2, 3, 6, 7, 8]. To our knowledge, only in six cases has the presence of *Helicobacter pylori* been revealed in heterotopic mucosa [1, 2, 6, 9, 10, 11]. Gastric heterotopia in the rectum typically presents with painless bleeding [2, 7, 8, 9, 12]. Endoscopic examination most commonly reveals a single, small, sessile polyp [6, 7, 9].

A rare case of asymptomatic gastric heterotopia in the rectum with the presence of *Helicobacter pylori* confirmed by immunohistochemical staining is reported.

Case description

In a 58-year-old asymptomatic woman three polyps were excised during colonoscopy within the screening program for colorectal cancer. In microscopic examination, two of them, excised from the sigmoid colon, proved to be tubular adenomas: one 1.2 cm in diameter, with severe dysplasia, and the other one 0.4 cm in diameter, with mild dysplasia. The third polyp, sessile, measuring 0.6 × 0.5 × 0.4 cm (Fig. 1A), located in the distal rectum about 2 cm above the dentate line, was composed of gastric oxyntic mucosa and contained moderate mononuclear

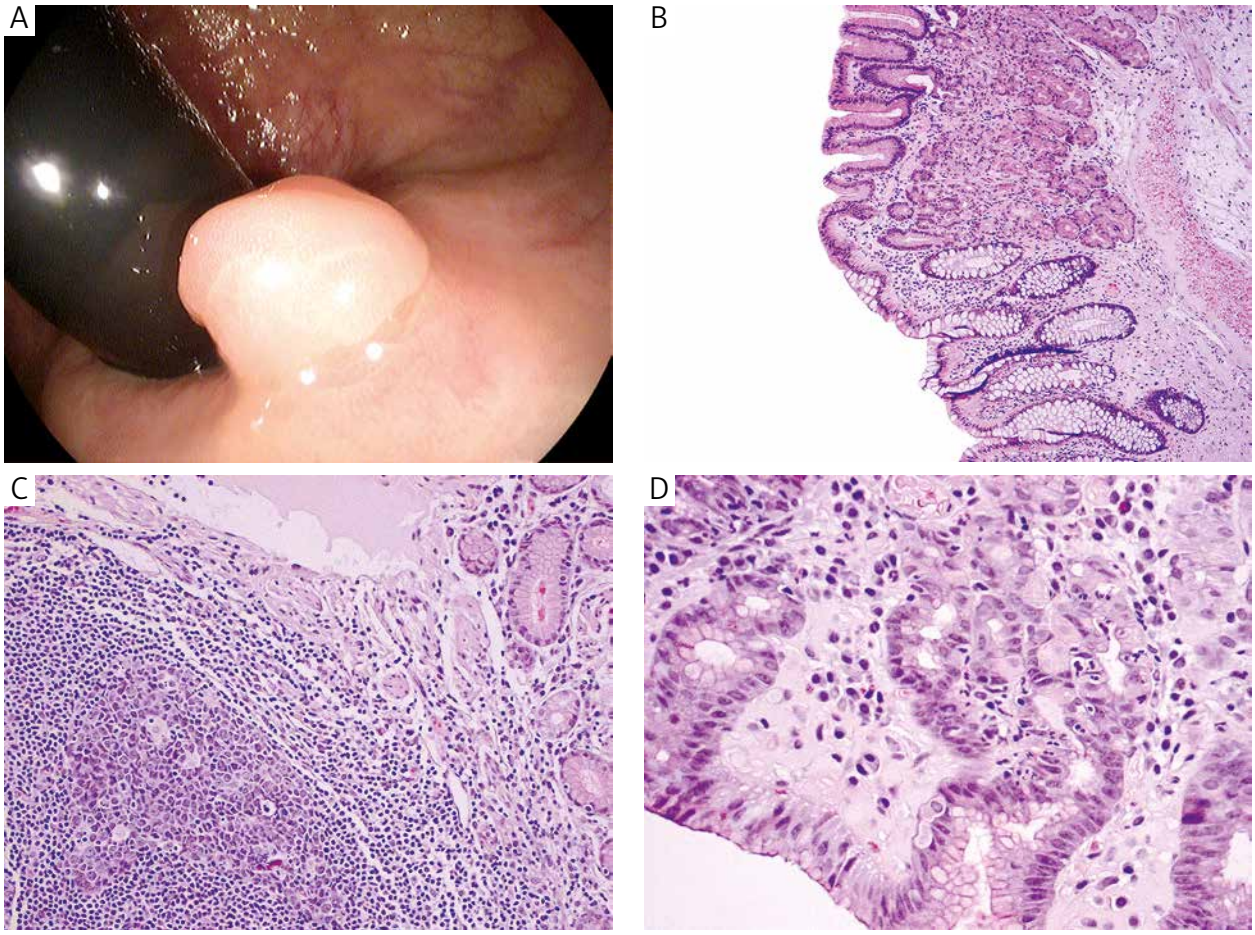

Fig. 1. Gastric heterotopia in the rectum. A) Single, small, sessile polyp revealed during colonoscopy. B) Gastric oxyntic mucosa present in the upper part and some intestinal crypts in the lower part of the photomicrograph (HE, objective magnification 5×). C) Moderate mononuclear inflammatory infiltrate and lymphoid follicle (HE, objective magnification 10×). D) Neutrophils invading the epithelium of fundic glands (moderate activity of inflammation) (HE, objective magnification 20×)

inflammatory infiltrate with some dense aggregates of lymphocytes and with a neutrophilic component (moderate activity) (Fig. 1B, C, D). Because such a picture found in the stomach would suggest *H. pylori* infection, Warthin-Starry staining was performed, and it highlighted the presence of numerous bacteria in a typical distribution on the epithelial surface (Fig. 2A). The bacteria were identified as *H. pylori* by immunohistochemistry (Fig. 2B), using a primary polyclonal rabbit antibody (DAKO, Denmark) in a 1 : 50 dilution, with EnVision+ System-HRP (DAB). Gastroscopy was not performed.

Discussion

Gastric heterotopia is not an uncommon finding in the proximal part of the digestive tract, especially in the upper esophagus, duodenum and Meckel diverticulum, but is very rare in the segments that are derived from the embryonal hindgut, including the rectum [9]. Since the first report by Ewell and Jackson in 1939 [13], less than 50 cases of gastric heterotopia in the rectum have been reported [2, 3, 6, 7, 8]. To the best of our knowledge, only in six of them has *H. pylori* been observed [1, 2, 6, 9, 10, 11] and in only one case has the presence of *H. pylori* been confirmed by immunohistochemistry [10].

In contrast to pyloric metaplasia, gastric heterotopia, composed of oxyntic-type mucosa with glands containing parietal and chief cells, is generally believed to be a developmental anomaly, not associated with chronic inflammatory conditions [1, 2, 3], but its exact origin remains unknown and its congenital nature is sometimes questioned. According to one theory, gastric heterotopia may result from the failure of descent of the embryonal foregut. Other authors postulate a developmental error of differentiation of the pluripotent endodermal cells lining the whole primary embryonal intestinal tube [2, 3, 6, 9, 14]. This may explain the occurrence of gastric heterotopia in sites distal to the foregut derivatives. More recent studies on Cdx2 mutant mice suggest that any pattern of gastric differentiation (including fundic and pyloric) may occur in the colon as a result of de-

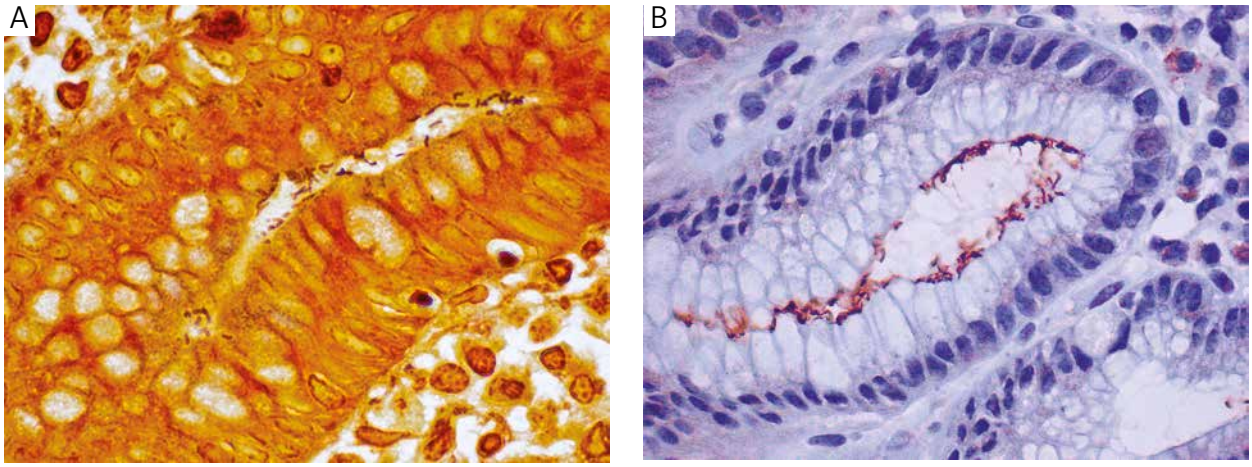

Fig. 2. *Helicobacter pylori* infection in gastric heterotopia in the rectum. A) Warthin-Starry staining (objective magnification 40×). B) Positive immunostaining for *Helicobacter pylori* (EnVision+ System-HRP (DAB), DAKO, objective magnification 40×)

regulation/inactivation of the homeobox genes, triggered by local factors [15]. According to this idea, gastric heterotopia may be an acquired lesion as well, not necessarily congenital.

The range of patients' age at presentation is wide (1 day to 65 years). The literature suggests slight male predominance [3, 6, 7, 9, 16]. The most common presenting symptom is rectal bleeding [2, 7, 8, 9, 12], usually painless, slight, intermittent, of various duration, even up to several years [2, 8, 9]. Occasionally, hemorrhage is not revealed but iron deficiency anemia is the first manifestation [9]. Less common manifestations include perineal ulceration, anal or abdominal pain, and altered bowel habits [1, 2, 3, 4, 6, 7, 9, 12, 16]. The symptoms may suggest irritable bowel syndrome [6, 7]. There are very few reports on asymptomatic gastric heterotopia, recognized in lesions discovered incidentally, as in the case presented here, during screening colonoscopic examination [14, 16, 17].

In most of the reported cases gastric heterotopia was located in the rectum more than 2 cm from the dentate line [1, 7, 16] and presented endoscopically as a single, sessile polyp [2, 6, 7, 8, 9, 12, 17], usually small, but sometimes measuring 4-5 cm in diameter [2, 8, 17]. In our case heterotopia was found about 2 cm above the dentate line, but its endoscopic view was typical (single, small, sessile polyp). Rarely gastric heterotopia was diagnosed in association with a diverticulum or an ulcer [1, 3, 4, 6, 7, 12, 16] or in a flat lesion, described in one case as "an irregular, pale plaque" [1] and in another one as "a sharply demarcated area of congested mucosa" [3]. The lesions typically do not display any distinctive endoscopic features and the definitive diagnosis is possible only on histopathological examination, although some descriptions referring to color and texture of the lesion may be suggestive of gastric-type mucosa: "intense pink color" [2], "salmon in color" [6], "there appeared to be some furrowing within this polyp" [6], or even "lesion with gastric rugae" [8].

The presence of *H. pylori* within gastric heterotopia suggests stomach infection. Dye *et al.* [1] observed *H. pylori* both in heterotopia and in the stomach. Srinivasan *et al.* [9] and Wildemore *et al.* [6] found the bacteria only in heterotopia and did not confirm their presence in gastroscopic biopsies. However, it does not rule out the infection of the stomach, as biopsy samples represent only a very small fragment of gastric mucosa. In our case, gastroscopy was not performed as the patient did not report any complaints. Reports on the presence of *H. pylori* in gastric heterotopia in the rectum support the opinion that these bacteria may pass along the whole length of the gastrointestinal tract in a viable form and that the fecal-oral route of transmission is possible [1, 2]. The inflammation associated with this infection may contribute to ulceration and bleeding or to more serious complications that are sometimes observed, such as perforation of the bowel, fistula formation or severe hemorrhage [2, 6, 17, 18]. Dye *et al.* [1] reported the resolution of chronic active inflammation of heterotopic mucosa after eradication of *H. pylori*.

Due to the small number of reported cases, there are no guidelines for the treatment of gastric heterotopia in the rectum. In most of the cases endoscopic excision was chosen [2, 6, 7, 8, 9, 17]. This is reasonable in view of possible complications. Malignant transformation has not been reported so far, but such a possibility should be considered, because there are reports on adenocarcinoma arising in gastric heterotopia of the upper esophagus [19].

In conclusion, gastric heterotopia in the rectum is a rare condition that may lead to serious complications. It typically presents as a sessile polyp, and it should be included in the differential diagnosis for

rectal bleeding. In a few cases it may be asymptomatic, even if infected by *Helicobacter pylori*.

The authors declare no conflict of interest.

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