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Case report

Three familial cases of fundic gland polyposis without polyposis coli

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Abstract. We report three cases of fundic gland polyposis in the stomach identified in three patients who were related. Grossly the numerous polyps covered an area limited to the body and fundus of the stomach, no polyps were found in the antrum, duodenum, colon, or rectum, and histologically, the gastric lesions consisted of numerous hamartomatous polyps, characterized by proliferation of the fundic and cystic glands. The gastric lesions were identified in families without polyposis coli. This type of fundic gland polyposis has never been documented before in the literature.

Key words: Stomach – Fundic gland polyposis – Gastric polyp

Introduction

Fundic gland polyposis/polyps (FGP) is marked by the presence of varying numbers of small and sessile polyps restricted to the body and fundus of the stomach. Histologically, FGP is characterized by proliferation of the fundic glands and cystically dilated glandular ducts. This finding was initially described by Japanese investigators as gastric lesions of a familial polyposis coli/Gardner's syndrome (FPC) (Ohsato et al. 1974; Utsunomiya et al. 1974; Watanabe et al. 1978) and was deemed peculiar to patients with FPC (Watanabe et al. 1978). Later, FGP showing no familial inheritance was identified in patients in the absence of FPC and interpreted as a separate clinicopathological entity (Sipponen and Siurala 1978; Iida et al. 1980; Tatsuta et al. 1980; Katz et al. 1982; Iida et al. 1984; Lee and Burt 1986; Venkataseshan et al. 1987; Sato et al. 1988). Lee and Burt (1986) found no histological differences between FGP with and without FPC.

We report three familial patients with FGP without FPC, a novel finding.

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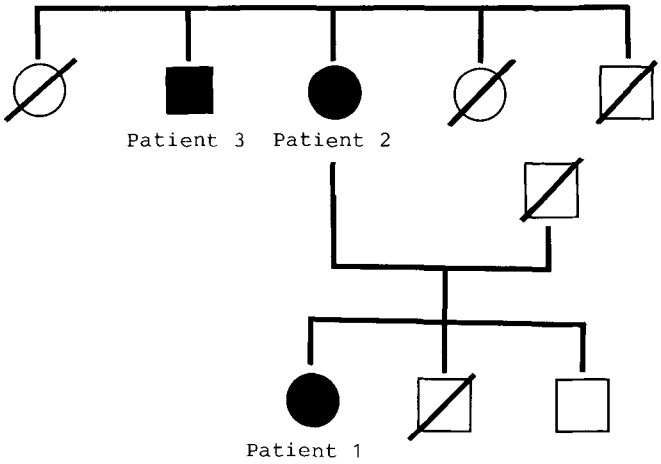
Case reports

A 16-year-old woman (patient 1) was admitted to our hospital in April 1981, with complaints of abdominal pain and nausea. Double-contrast radiograph and gastroscopy revealed numerous polyps extending from the fundus to the body of the stomach. No polyps were found in the cardia, antrum, pylorus or duodenum. Her family members were then examined by radiography and as shown in the family pedigree (Table 1), numerous polyps covering the gastric body and fundus were detected in her 42-year-old mother (patient 2; Fig. 1) and 48-year-old uncle (patient 3). No polyps in the alimentary canal or extra-alimentary changes (including pigmentation, nail changes and hair loss) were found in the other family members.

All gastric polyps observed in the three patients had a smooth surface and no peduncle and measured less than 8 mm in diameter. Patients 2 and 3 had no symptoms, and physical examination and laboratory data on admission showed no signs of abnormality, except for elevation of rGTP (r-glutamyltranspeptidase) (132 mU) in patient 3, which reflected a fatty liver revealed by ultrasound examination.

During a follow-up period of 11 years, the polyps showed little or no change in patients 1 and 3. In contrast, in the body of the stomach in patient 2, gastroscopy revealed the appearance of two larger polyps measuring 1.5 × 0.5 cm and 1.5 × 1.5 cm, respectively,

Table 1. Family pedigree



Square, Male; circle, female; black, affected by fundic gland polyposis; slanted line, no polyps detected in the gastrointestinal tract

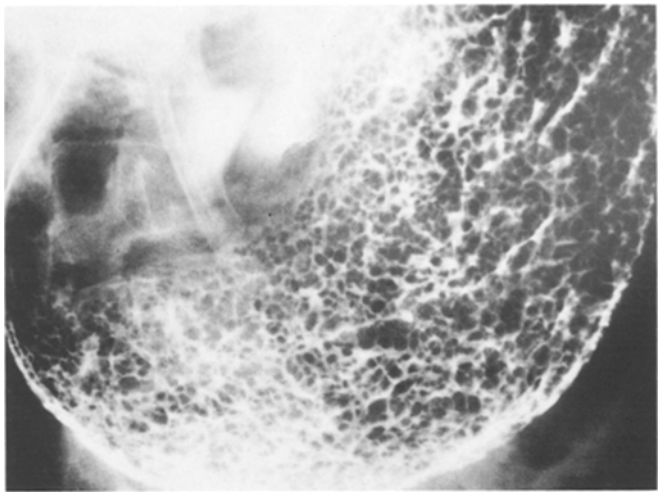


Fig. 1. Double-contrast radiograph of patient 2 shows numerous polyps restricted to the fundus and body of her stomach, but not in the cardia, antrum, pylorus and duodenum

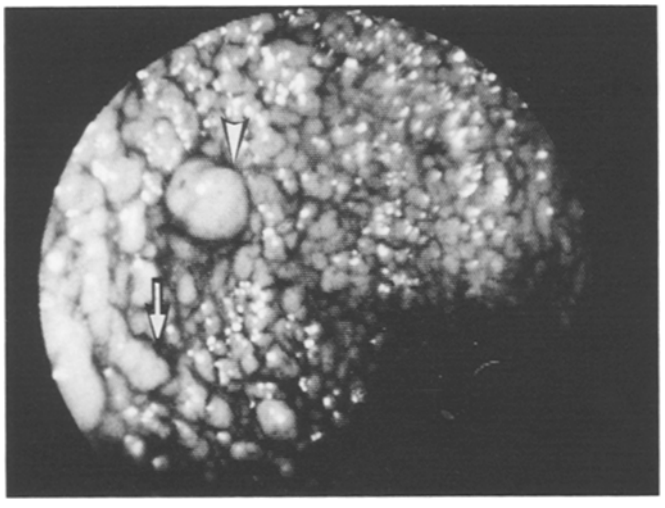


Fig. 2. Gastroscopy of patient 2 shows that numerous hemispherical polyps were closely distributed from the angulus to the fundus of the stomach, covering the entire area. Almost all the polyps are small, less than 8 mm in diameter, and sessile. Hyperplastic polyp (*arrowhead*) measuring 1.5 × 1.5 cm and adenoma (*arrow*) measuring 0.5 × 1.5 cm are also present

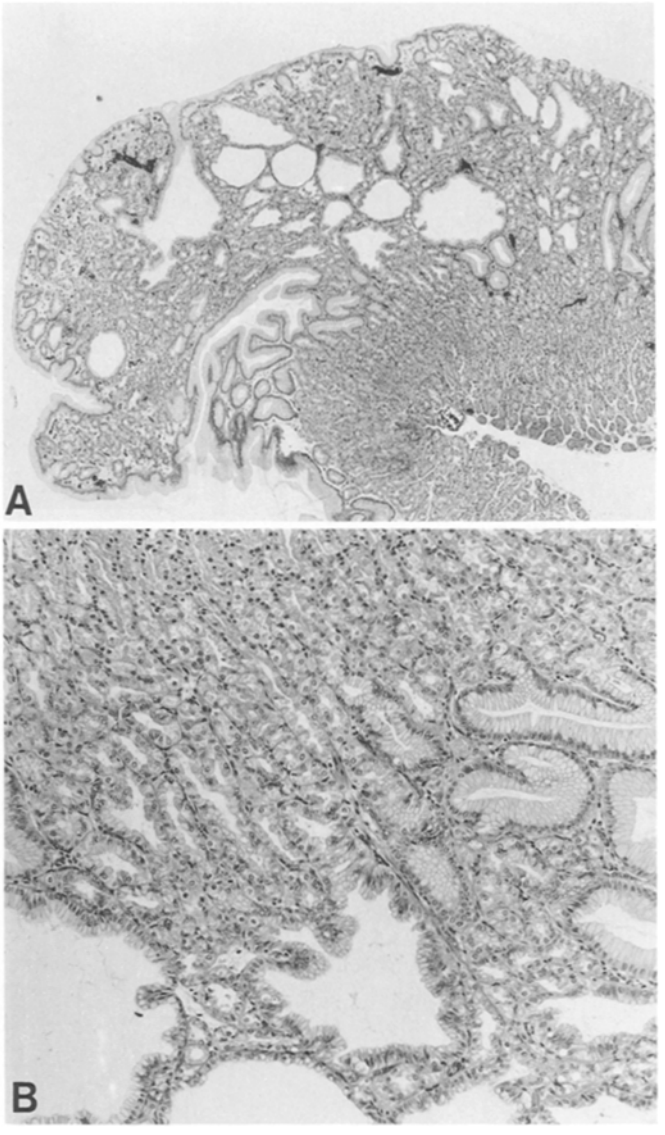


Fig. 3A, B. Photomicrographs of small sessile fundic gland polyp shown in Fig. 2. **A** The polyp is elevated above normal body-type mucosa, characterized by overlying foveolae, proliferative glands and dilated glandular ducts. Haematoxylin and eosin. ×40 **B** The glands are composed largely of mucous neck cells and parietal cells and partly of chief cells, and the dilated ducts are lined by foveolar epithelium. Haematoxylin and eosin, ×120

in August 1986 (Fig. 2) and of two larger ones 1 and 2 cm in diameter, respectively, in June 1988.

Histologically, the small polyps biopsied from patient 1 and 2 showed slight elevation above the normal body-type mucosa (Fig. 3A). The overlying foveolae were shortened, beneath which there were glands composed largely of mucous neck cells and parietal cells and partly of chief cells with slightly basophilic cytoplasm; the proliferated glands were irregular, tortuous and sometimes branching (Fig. 3B). In addition, microscopic examination revealed the presence of dilated glandular ducts lined by surface type epithelium with an occasional glandular-type epithelial proliferation (Fig. 3B). The polyps of patient 3 were not biopsied. A large polyp measuring 1.5 × 0.5 cm in patient 2 had an adenomatous papillotubular architecture, which consisted of closely packed and irregular tortuous tubules characterized by prominent glandular budding (Fig. 4A). The lamina propria between the tubules consisted largely of a delicate fibrovascular stroma. The lining epithelium was composed of tall mucus-producing cells, similar to foveolar epithelium, with mild to moderate atypia; the oval nuclei were enlarged and hyperchromatic, contained a prominent nucleolus, and exhibited a tendency for stratification with loss of nuclear polarization (Fig. 4B). Their cytoplasm contained mucin reactive to both periodic acid-Schiff and alcian blue (pH 2.5), suggesting that the polyp may be an adenoma. The other three large polyps were characterized by elongated or cystically dilated crypts consisting largely of foveolar cells without atypia and some parietal cells. The deeper part of the polyp contained a focus of fundic glands. These findings indicate that the polypoid lesions are a hyperplastic polyp of foveolar type.

During the follow-up period of 11 years, no polyps were detected in the antrum, pylorus, duodenum and colon in the three patients by gastroscopy, sigmoidoscopy and barium enema. No extra-alimentary manifestations (pigmentation, nail changes and hair loss) suggestive of FPC were detected in these patients.

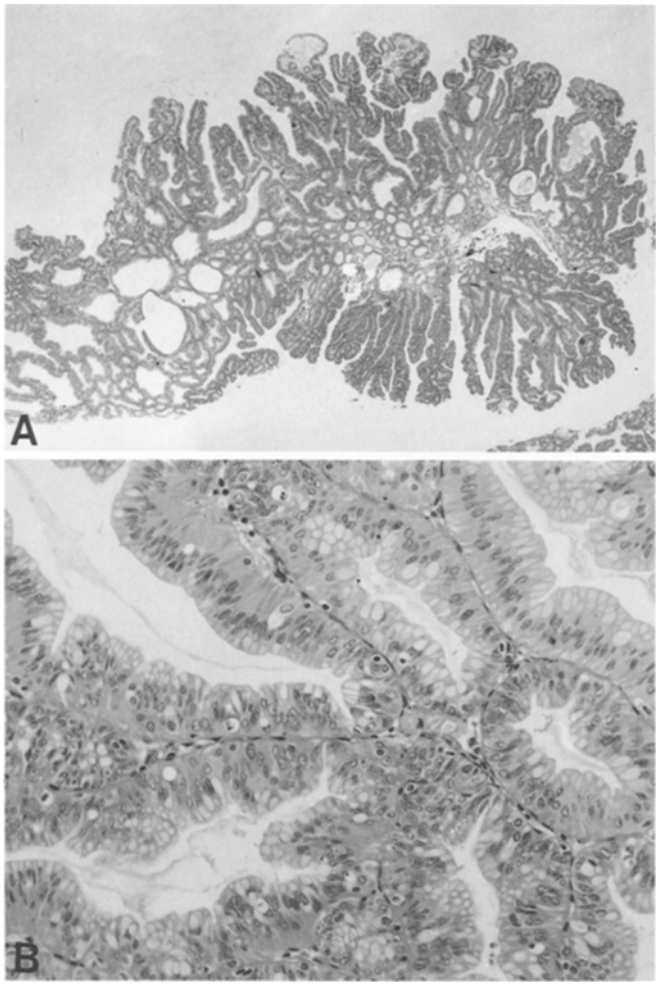


Fig. 4A, B. Photomicrographs of an adenoma shown in Fig. 2. **A** The polyp is characterized by a papillotubular architecture. Haematoxylin and eosin, ×12 **B** The epithelial lining is composed of tall mucus-producing cells with mild to moderate atypia. Haematoxylin and eosin, ×240

Discussion

FGP may occur with or without FPC. Histologically, FGP is characterized by proliferation of the fundic glands with cystically dilated glandular ducts and there are no histological differences between FGP with and without FPC (Lee and Burt 1986). They appear to be non-adenomatous polyps with hamartomatous (Sipponen and Siurala 1978; Watanabe et al. 1978; Tatsuta et al. 1980; Katz et al. 1982; Iida et al. 1984) or hyperplastic (Jarvinen et al. 1983; Burt et al. 1984) features and with an undefined histogenesis. FGP was observed in 27.3–55% of individuals with FPC by endoscopic examination (Watanabe et al. 1978; Jarvinen et al. 1983; Burt et al. 1984; Iida et al. 1984; Iida et al. 1985). Sipponen and Siurala (1978) and Iida et al. (1984) have found 9 and 23 cases of FGP without FPC in 15,000 and 27,000 routine gastroscopic studies, respectively. Our cases may be considered to be a variant of FGP without FPC, a variant of FGP with FPC, or a distinct disease of the stomach.

The lesions of FGP without FPC, unlike our cases, show no familial tendency and usually number less than 100 (often 15–30). The appearances of adenoma or hyperplastic polyp have not been reported in FGP without FPC, while those of FGP with FPC are linked to a positive familial inheritance. Numerous polyps covering the whole gastric body and fundus have been found in FGP with FPC (Iida et al. 1984, 1985). In addition, adenomas are the most common form of gastric lesions accompanying FPC (Ohsato et al. 1974; Utsunomiya et al. 1974; Watanabe et al. 1978; Denzler et al. 1979; Ranzi et al. 1981; Jarvinen et al. 1983; Iida et al. 1988). Hyperplastic polyps have also been described in the gastric body and antrum of some FPC patients (Utsunomiya et al. 1974; Denzler et al. 1979; Ranzi et al. 1981). Although similar to FPG with FPC in these aspects, polyps in the antrum, duodenum, colon and rectum were excluded in our three cases, and no extra-alimentary manifestations were detected in the cases or other family members. It has been reported that in a great majority of FPC patients, the polyps become manifest in the colon before the age of 30, but, in some cases, polyps do not develop in the colon until the fifth decade or later (Asman and Pierce 1970). These data suggest the possibility that the FGP in our case might have occurred before the manifestation of FPC in this family. Further observations may be required to determine whether our cases belong to FGP with FPC.

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