

## EXTRALUMINAL MASS TYPE OF HETEROTOPIC GASTRIC MUCOSA IN THE ILEUM CAUSING HEMORRHAGE

Hirosada SHIGEMOTO, Yoshihiro HORIYA\*, Yasuhisa YAMAMOTO\*, Hisao MASAKI\*, Yoshihiko TAKAO\*, Kaiso SANO\*, Yasuhiko ITO\*\*, Isamu NARABAYASHI\*\* and Wataru FUJITA\*

*Department of Primary Care Medicine, \*Department of Surgery,  
\*\*Division of Nuclear Medicine, Department of Radiology  
Kawasaki Medical School,  
Kurashiki 701-01, Japan*

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### Abstract

Two infants with heterotopic gastric mucosa in the ileum causing hematochezia are presented. Both showed a hot spot by preoperative abdominal scanning with technetium 99m pertechnetate. Operations revealed that each of these cases had a solid tumor on the antimesenteric side of the ileal serosa. Serial microscopic examination showed that the tumor consisted of gastric fundal gland and there was a small cavity with a few narrow communicating channels between this cavity and gastric mucosa protruding into the ileal cavity. These cases are considered to be the extraluminal mass type of congenital gastric heterotopia.

Heterotopic gastric mucosa in the gastrointestinal tract may be either congenital or acquired. The congenital variety is most often found in the esophagus, duodenum, Meckel's diverticulum, or in the remnants of the vitelline duct. It is rare in the jejunum and ileum<sup>1)</sup>.

We present two cases of infants who had bloody bowel motions due to heterotopic gastric mucosa in the ileum. Both had tumors proliferating extraluminally from the wall of the ileum and showed a hot spot with preoperative abdominal scanning with technetium 99m pertechnetate.

### CASE REPORTS

Case 1 : A 4-month old female infant was admitted to the Kawasaki Medical School Hospital in October 1978, with a three-day history of bloody stools and anemia. On examination, she was well-nourished and healthy. Abdominal and rectal examinations were negative except for the presence of

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重本弘定, 堀谷喜公, 山本康久, 正木久男, 高尾良彦, 佐野開三, 伊藤安彦, 榎林 勇, 藤田 涉

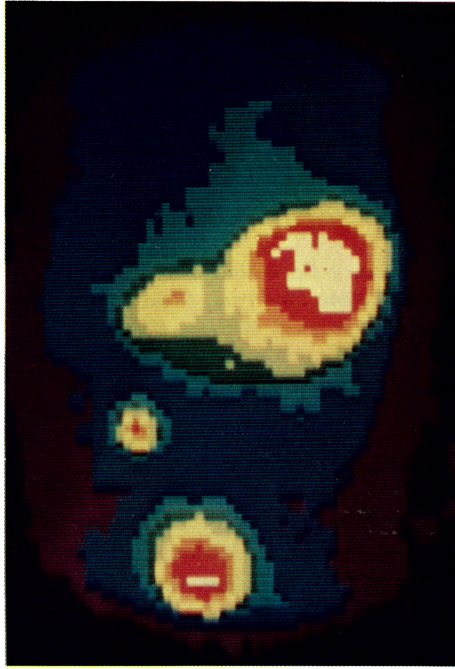


Fig. 1. Abnormal uptake of  $^{99m}\text{Tc}$  in heterotopic gastric mucosa in the case 1. Normal uptake of radionuclide in stomach and bladder seen in upper and lower abdomen.

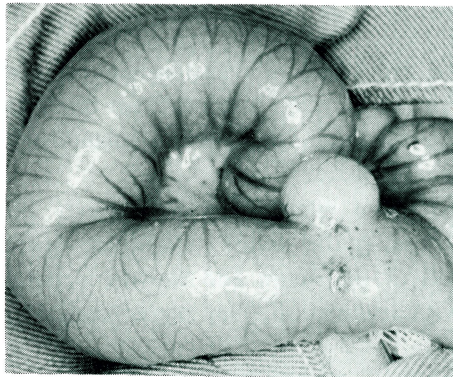


Fig. 2. A 13 mm spherical mass is found on the serosa of the antimesenteric side of the ileum about 32 cm from the ileocecal valve.

guaiac-positive stools. Blood examination showed a hematocrit of 30.8% hemoglobin 10.6 g/dl, white blood count 11,100 and 200,000 platelets. Roentgenograms of the gastrointestinal tract including the small bowel and a barium

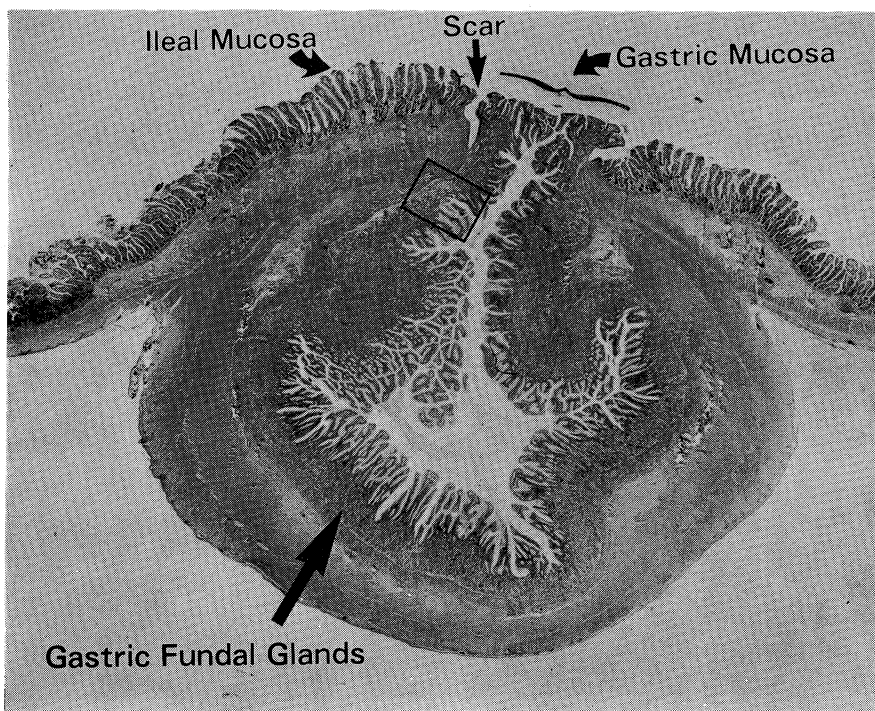


Fig. 3. Microphotograph of the tumor in the case 1 demonstrates the small cavity and the gastric mucosa in the tumor ( $\times 4$ , H&E stain).

enema, showed no abnormalities. Abdominal scan with technetium 99m pertechnetate showed abnormal uptake in the right lower quadrant at 15 to 120 minutes after injecting the isotope (Fig. 1). A Meckel's diverticulum with ectopic gastric mucosa was suspected. At surgery, a 13 mm spherical mass was found on the serosa of the antimesenteric side of the ileum. It was located about 32 cm from the ileocecal valve (Fig. 2). Ileal resection was performed. Postoperative technetium 99m pertechnetate scanning was negative and the postoperative course was uneventful. The mucosal surface of the resected ileum had a small erosion, but there was no visible communicating fistula into the tumor. Serial microscopic examination showed that the tumor consisted mostly of fundal type with some pyloric type gastric mucosa (Fig. 3,4). There was a small cavity with a few narrow communicating channels between this cavity and gastric mucosa protruding into the ileal cavity. There was an erosion between gastric mucosa and ileal mucosa (Fig. 5).

Case 2 : A 22-month old female infant was admitted to the Kawasaki Medical School Hospital in December 1978, with a history of passing bloody

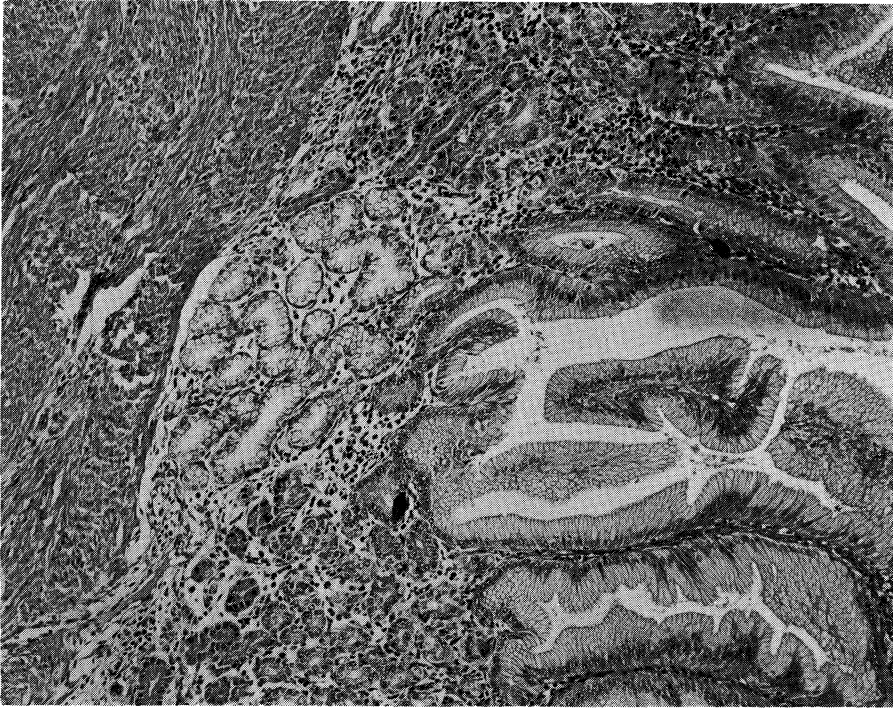


Fig. 4. The magnified view of the frame in Fig. 3 demonstrates that the tumor consists mostly of fundal type (dark region) with some pyloric type gastric mucosa (clear region) ( $\times 40$ ).

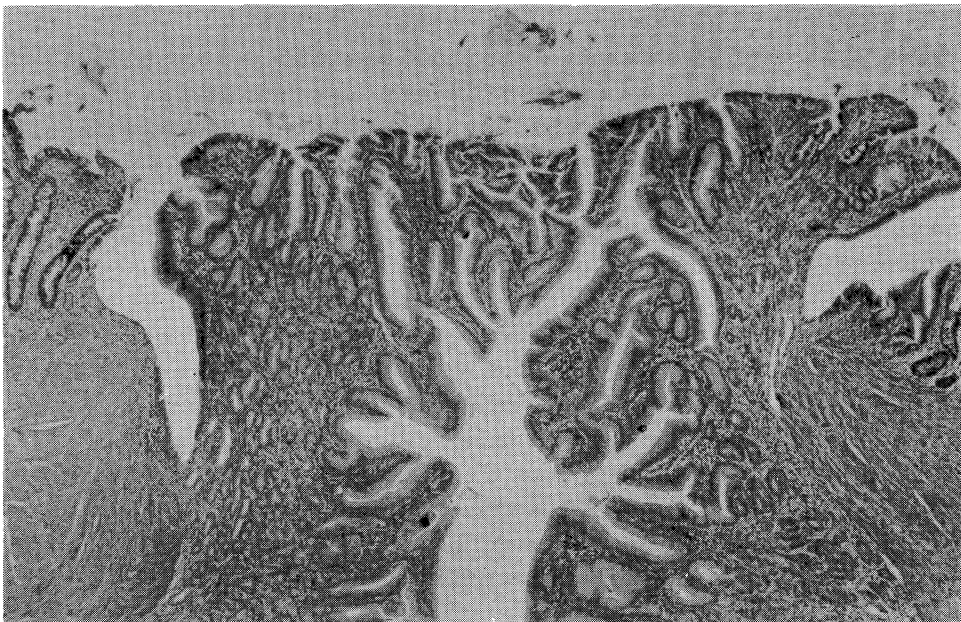


Fig. 5. The magnified view of the gastric mucosa protruding into the ileal cavity in Fig. 3 demonstrates that this tumor has a few narrow communicating channels between this cavity and gastric mucosa protruding into the ileal cavity and there is an erosion between gastric mucosa and ileal mucosa ( $\times 23$ ).

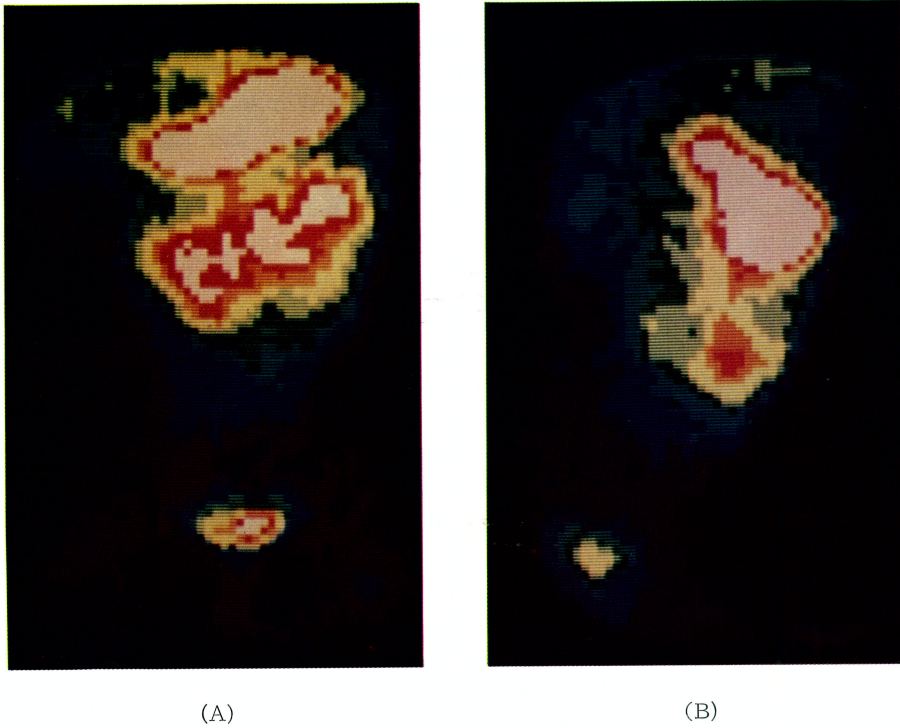


Fig. 6. Abnormal uptake of  $^{99m}\text{Tc}$  in heterotopic gastric mucosa adjacent to the normal uptake of the stomach in the case 2 : (A) anterior view, (B) right lateral view.



Fig. 7. A hemispherical mass  $8 \times 7 \times 5$  mm is found on the serosa of the antimesenteric side of the ileum about 75 cm from the ileocecal junction.

stools. She had an episode a month earlier which disappeared spontaneously. General appearance showed a well-nourished, but pale infant with obvious profuse bleeding from the rectum. Blood examination showed a hematocrit of 29.9%, hemoglobin 9.8 g/dl, white blood count 8,200 and 150,000 platelets. The test for occult blood was highly positive. Roentgenograms of the gastrointestinal tract including the small bowel and a barium enema, were normal. A technetium 99m pertechnetate scan showed abnormal increased uptake in the upper midabdomen adjacent to the uptake of the stomach (Fig. 6). A preoperative diagnosis of Meckel's diverticulum with ectopic gastric mucosa was made and operation was performed. A hemispherical mass  $8 \times 7 \times 5$  mm was found on the serosa of the antimesenteric side of the ileum about 75 cm from the ileocecal junction (Fig. 7). Postoperative technetium 99m pertechnetate scanning was negative, and the postoperative course was uneventful. The mucosal surface of the resected ileum had a peptic ulcer  $10 \times 2 \times 1$  mm, but there was no visible communicating fistula into the tumor. Microscopic serial sections showed that this tumor had a small cavity and its wall consisted of the gastric fundal gland (Fig. 8). There were a few narrow communicating channels about 0.1 to 0.2 mm in diameter between the cavity of the tumor and gastric mucosa protruding

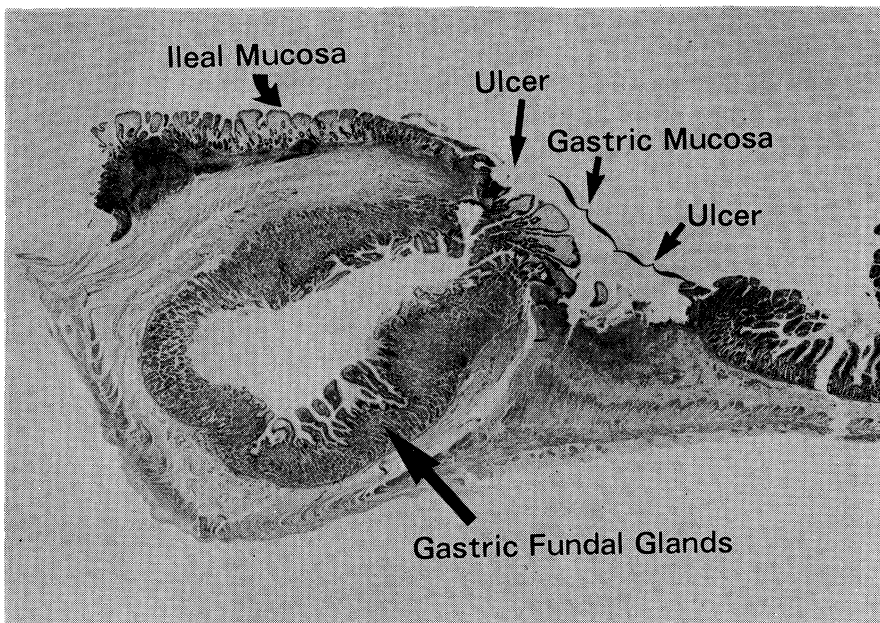


Fig. 8. Microphotograph of the tumor in the case 2 demonstrates the small cavity and the fundal type of gastric gland in the tumor ( $\times 4$ , H&E stain).

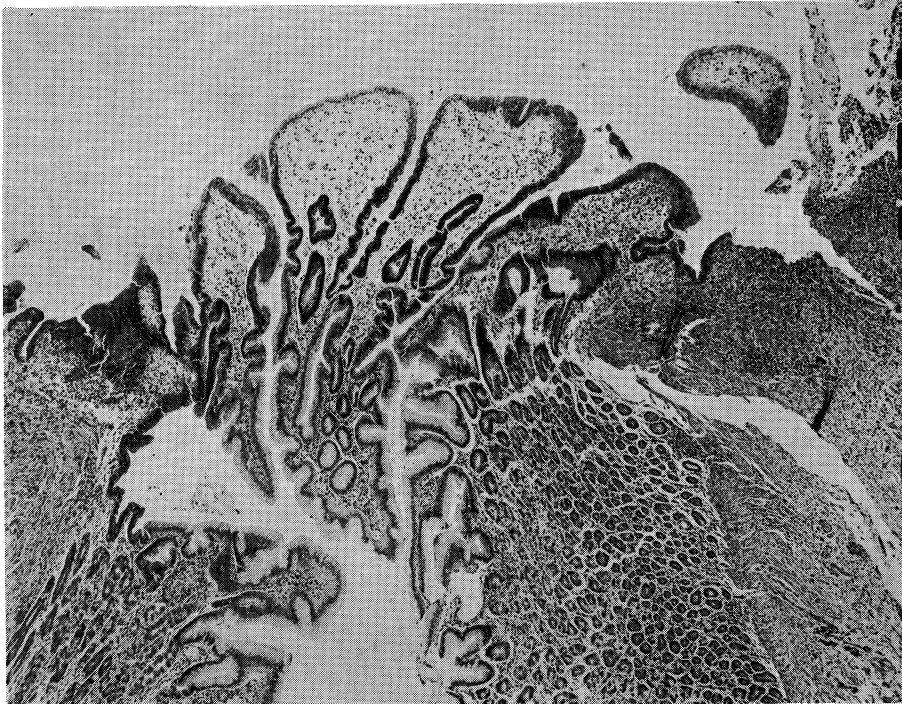


Fig. 9. The magnified view of the gastric mucosa protruding into the ileal cavity in Fig. 8 demonstrates that there are a few narrow communicating channels between the cavity of the tumor and gastric mucosa protruding into ileal cavity, and ulcers between the gastric and ileal mucosa ( $\times 19$ )

TABLE 1. Gastric Heterotopia

Congenital origin (Fundal type)	Superficial h.	{ Esophagus Duodenum Small intestine Meckel's diverticulum Duplication Umbilicus
	Deep h.	
{ Acquired origin (Regenerative process in chronic inflammatory lesion) (Pyloric type; Metaplasia)		

into ileal cavity. There was a lineal ulcer between the gastric and ileal mucosa (Fig. 9).

**DISCUSSION**

Taylor<sup>1)</sup> reported that superficial gastric heterotopia fell into two groups (Table 1). Those of acquired origin are common in the jejunum and ileum

TABLE 2. Summary of Previously Reported and Present Cases of Tumorous Heterotopic Gastric Mucosa in the Jejunum and Ileum

Author	Patient No.	Sex	Age	Location and Side	Symptoms and Duration	Appearance	Size
Poindecker <sup>2)</sup> (1912)	1	M	9 yr	Ileum mesenteric	Obstruction Since infant	Polypoid	—
Taylor <sup>1)</sup> (1927)	2	M	1 yr 5 mo	Ileum mesenteric	Hematochezia 4 mo	Rugosal with ulcer	—
Barták <sup>3)</sup> (1932)	3	M	50 yr	Jejunum antimesenteric	Asymptomatic	Extraluminal	1×2 cm
Kimpton <sup>4)</sup> (1938)	4	F	7 yr	Jejunum	Obstruction 4 yr	Polypoid	0.4 cm long 3 cm diameter
Ramsay <sup>5)</sup> (1954)	5	F	9 yr	Jejunum mesenteric	Obstruction 2 mo	Rugosal	3.8×5 cm
Soule <sup>6)</sup> (1959)	6	M	28 yr	Jejunum antimesenteric	Obstruction 4 yr	Polypoid (2)	2×1×5 cm 2.5×2×2 cm
	7	F	25 yr	Jejunum mesenteric	Obstruction 4 mo	Polypoid with ulcer	1.5 cm diameter
	8	F	6 yr	Ileum antimesenteric	Obstruction 3 weeks	Polypoid	1.5 cm diameter 1 cm stalk
Lee <sup>7)</sup> (1970)	9	M	11 yr	Ileum mesenteric	Obstruction 3-4 yr	Polypoid	3×3.5×0.8 cm
	10	M	25 yr	Jejunum lateral	Obstruction 3 weeks	Rugosal (2)	8×3 cm 6×3 cm
Nawaz <sup>8)</sup> (1974)	11	M	43 yr	Ileum circular	Anemia 40 yr	Rugosal with ulcer (2)	4 cm long 7 cm long
Hinkamp <sup>9)</sup> (1974)	12	M	4 yr	Ileum antimesenteric	Peritonitis	Ulcer with perforation	2.5 cm diameter
Chandrakamoj <sup>10)</sup> (1978)	13	M	11 mo	Ileum mesenteric	Hematochezia 1 mo	Polypoid with ulcer	2×2×1 cm
	14	M	12 mo	Ileum mesenteric	Hematochezia 6 hr	Polypoid with ulcer	0.5×1×2 cm
Case 1	15	F	4 mo	Ileum lateral	Hematochezia 3 days	Extraluminal with scar	1.3×1.1×1.0 cm
Case 2	16	F	1 yr 10 mo	Ileum antimesenteric	Hematochezia 2 mo	Extraluminal with ulcer	0.8×0.7×0.5 cm



in areas of mucosal regeneration accompanying inflammatory lesions such as regional enteritis. In this situation, the gastric mucosa can be considered as metaplasia and consists mainly of mucus-secreting cells without parietal or chief cells.

A review of the literature with our 2 cases revealed only 16 cases of congenital heterotopic gastric mucosa beyond the ligament of Treitz during the past 70 years except for its occurrence in Meckel's diverticulum and duplication (Table 2). In the pediatric age group, the number reduces to 11 in the jejunum and ileum. The first case of the congenital heterotopic gastric mucosa was presented by Poindecker<sup>2)</sup> in 1912, as an instance of gastric heterotopia in the ileum of a 9-year old boy who had evidence of intestinal obstruction. Taylor<sup>1)</sup> reported the first case of a 17-month old boy causing hemorrhage in the ileum. Of these 16 cases, eight presented a small bowel obstruction resulting from intussusception of the mass of gastric mucosa six melena or hematochezia, one perforation due to ulcer and one an incidental surgical finding (Table 3). All 6 cases of bleeding from the rectum have been reported to be in the ileum and 5 were under 2 years of age.

TABLE 3.  
Reported Complications of Congenital Gastric Heterotopia  
in the Jejunum and Ileum

Complication	Number of Patients (Child)	
Intussusception	8	(5)
Melena or Hematochezia	6	(5)*
Perforation	1	(1)
Incidental Surgical Finding	1	(0)
Total	16	(11)

\*containing our 2 cases

Gryboski<sup>11)</sup> divided the congenital superficial gastric heterotopia into two groups macroscopically : polypoid type which grows like a polyp in the intestinal cavity and rugosal type which has thickened mucosal folds. The previously reported 14 instances are 8 of polypoid type, 5 of rugosal type and one of Barták's case which had a thickened serosal mass on the jejunum and normal jejunal mucosa in the jejunal cavity. Each of our two cases presented hematochezia and had a solid tumor on the antimesenteric side of the ileal serosa. Microscopic serial sections showed that the tumor consisted of gastric fundal glands with a small cavity and gastric mucosa protruding into the ileal cavity with a few channels 0.1 to 0.2 mm in diameter between the cavity and gastric mucosa in the ileal cavity. In view of the serial microscopic studies,

TABLE 4.  
Type of Congenital Gastric Heterotopia Reported  
in the Jejunum and Ileum

Type	Number of Patients (Child)	
Polypoid type	8	(6)
Rugosal Type	5	(3)
Extraluminal Type	3*	(2)**
Total	16	(11)

\*Barták's case (M, 50yr) found at gastric surgery

\*\*our 2 cases

these cases are considered to be the "extraluminal mass type" of congenital gastric heterotopia, rather than Meckel's diverticulum or duplication (Table 4).

We agree with Taylor<sup>1)</sup>, Willis<sup>12)</sup> and Gray<sup>13)</sup> that the primitive intestinal mucosa is totipotent and capable of differentiating into any type of intestinal epithelium at any other level. Thinking from the locations in the vicinity of the omphalomesenteric remnant and their histologic appearance, these lesions are probably vestigial omphalomesenteric ducts in the wall of ileum.

#### Acknowledgment

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