Ileal duplication with extensive gastric heterotopia in a girl

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Background: **Gastrointestinal duplications are rare congenital abnormalities known to occur at any level of the alimentary tract from the mouth to the anus. The cause of intestinal duplication has not been established. Several theories have been put forward to explain different types of duplications. Some of these duplications are large sized and giant, and only 4 cases have been reported.**

Methods: **A 4-year-old girl was referred to our hospital with a history of abdominal pain, abdominal distension, and diarrhea mixed with black blood for 20 days. Technetium-99m scintigraphy identified heterotopic gastric mucosa at the middle and lower abdominal region. Enteric duplication was suspected.**

Results: **Operatively, duplication was found to be located at the ileum with abnormal hypertrophy in shape, 50 cm of the ileum was resected, and an ileoileal end-to-end anastomosis was made. Stomach-like mucosa and some ring structures were identified instead of the normal intestinal mucosa when opening this ileal duplication. Microscopically, most of mucosa showed gastric corpus-fundic glands.**

Conclusions: **This is an unusual case of enteric duplication. Ultrasonography, computed tomography and technetium-99m scintigraphy are helpful in the diagnosis of duplication.**

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Key words: **children;**
enteric duplications;
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Introduction

Enteric duplications are rare congenital abnormalities. They are usually spherical or tubular in shape. Duplicated parts usually share a common muscularis and a common blood supply with the adjacent gut. They may contain ectopic gastric mucosa or pancreatic tissue. Peptic ulceration caused by ectopic gastric mucosa in the duplication can lead to gastrointestinal bleeding or peritonitis.[1] We report a case of perforated ileal duplication, which presented as a stomach-like duplication. This is a very unusual manifestation in enteric duplication and only 4 cases have been reported.

Case report

A 4-year-old girl presented to our unit with a history of abdominal pain, abdominal distension and diarrhoea mixed with black blood for 20 days. Medical history showed ileal perforation with a simple suture closure 40 days ago in another hospital. On admission, the patient was afebrile with blood pressure of 110/70 mmHg, a respiratory rate of 30, and a pulse rate of 140. She was anemic (hemoglobin, 65 g/L; platelet count, 525 g/L), but other laboratory findings were normal. Mild tenderness and distention were found in the middle abdomen. Bowel sounds were present and hyperactive. Ultrasonographic examination and computed tomography (CT) of abdomen revealed a little bit free fluid in the peritoneal cavity. Technetium-99m scintigraphy identified heterotopic gastric mucosa at the middle and lower abdominal region (Fig. 1). Operatively, duplication was found to be located at the ileum with abnormal hypertrophy in shape (Fig. 2), 50 cm of the abnormal ileum was resected with an ileoileal end-to-end anastomosis. Opening this duplication, we could only identify stomach-like mucosa and some ring structures instead of the normal intestinal mucosa (Fig. 3). Microscopically, most of the mucosa showed gastric corpus-fundic glands (Fig. 4).

Discussion

Gastrointestinal duplications are rare congenital abnormalities known to occur at any level of the alimentary tract from the mouth to the anus. The cause

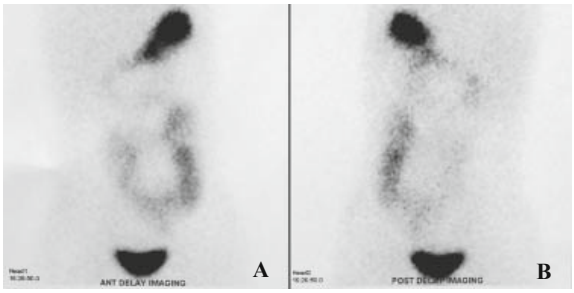

Fig. 1. Technetium-99m scintigraphy (**A**) showing abnormal tracer accumulation in the middle and lower abdominal region appearing at the same time as gastric mucosal uptake and progressively increasing in intensity. 60-minute delayed images (**B**) also showing abnormal tracer accumulation and remaining the same shape of radioactive accumulation at the same position compared with dynamic imaging.

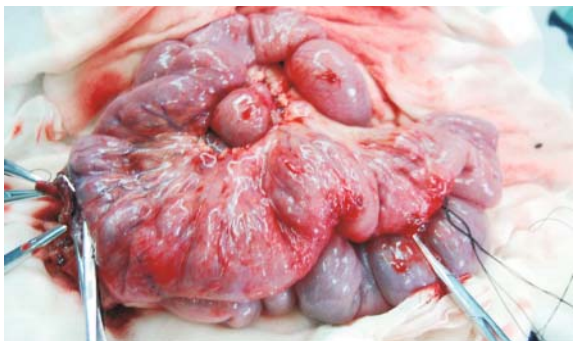

Fig. 2. Intraoperative picture showing duplication located at the ileum with abnormal hypertrophy in shape.

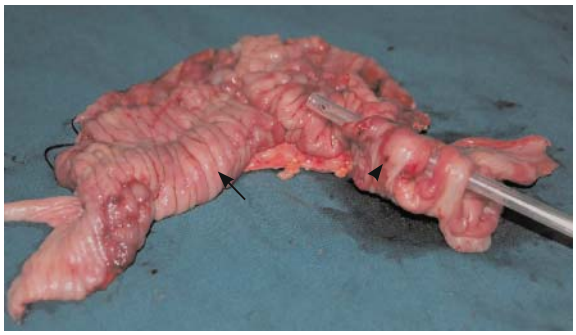

Fig. 3. Stomach-like mucosa (arrow) and some ring structures (arrow head) instead of the normal intestinal mucosa when opening the ileal duplication.

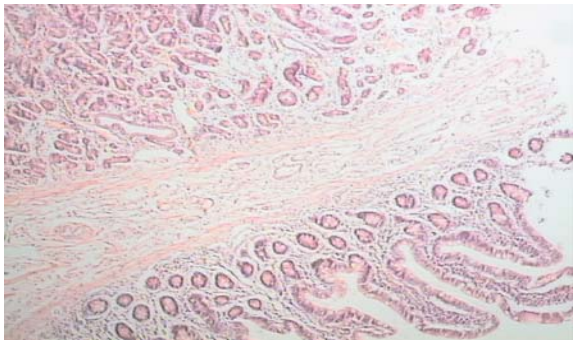

Fig. 4. Microscopically, most of gastric mucosa showed corpus-fundic glands (HE, original magnification × 10).

of intestinal duplication is unclear. Several theories have been put forward to explain the cause of duplications. Of these, persistence of fetal gut diverticula, defects in re-canalization of the solid stage of primitive gut, partial twining and the split notochord theories are popular.[2]

Duplicated parts usually share a common muscularis and a common blood supply with the adjacent gut. There are two types of duplication: cystic and tubular. The length of a tubular type of duplication varies from few millimeters to 90 cm and rarely is almost as long as the normal intestine. Tubular type of duplication usually communicates with the lumen of the adjacent normal intestine either caudally, distally or at several points in between. When there is no communication they are filled with mucous secretion and cause pain, usually presenting with a mass in the abdomen. When a tubular type of duplication communicates with the normal bowel at its cephalic end, the lumen greatly distends with intestinal secretions causing obstruction or even perforation. On the other hand the duplicated part empties readily in a caudal type of communication. Cystic type of duplications usually has no communication with the lumen of adjacent bowel. Gastric type of mucosa usually lines part or the whole of the length of the tubular duplication. Secretion of acid peptic juice from this ectopic gastric mucosa can cause peptic ulcer, bleeding and even perforation with peritonitis.[3-6] Our case presents as a special type. Opening this duplication, we could identify stomach-like mucosa and some ring structures instead of normal intestinal mucosa.

We also could not identify the gut from the duplication. On microscopic examination, intestinal mucosa was unremarkable and most of the mucosa showed gastric corpus-fundic glands. The pathologic results support that it is duplication.

The clinical diagnosis of duplication may be difficult before surgery. Abdominal ultrasonography and CT scan can detect duplication. Technetium-99m scintigraphy demonstrates ectopic gastric mucosa in duplication and is helpful in cases of unexplained gastrointestinal bleeding. The management of symptomatic duplication is surgical. Infants with subclinical intestinal duplication discovered incidentally on scan are commonly subjected to surgery to prevent the potential risk of complication by obstruction or perforation.[7]

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