

# Ileal tubular duplication; a rare cause of bowel obstruction in adults

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## ABSTRACT

Gastrointestinal tract duplications (GSD) are rare congenital abnormalities. Eighty percent of GSDs are diagnosed in children less than two years of age. These lesions can be seen anywhere from the oral cavity to the anus, but ileum is the most commonly affected site. GSD can be long and tubular, but are usually in the form of cystic masses. The clinical manifestation of GSD in adults is variable, and they are rarely considered as part of differential diagnosis. In this case report, we presented a 20-year-old patient with ileal duplication. Despite medical tests and radiological examinations, the diagnosis could be made during the operation.

**Keywords:** Ileal duplication, ileus, intestinal obstruction

## INTRODUCTION

Gastrointestinal (GI) duplications are rare congenital anomalies of unknown etiology. Although they can be detected in any part of the digestive tract from the oral cavity to the anus, they are most frequently identified in the small intestine (1). Duplications can be seen in two forms as cystic and tubular types. Approximately 80% of patients clinically present before two years of age (2). The most common symptoms include recurrent abdominal pain, nausea, vomiting and abdominal palpable mass (3). The preoperative diagnosis of GIS duplication is difficult and radiologic examinations may not be sufficient for an accurate diagnosis. Obstruction, bleeding, perforation, volvulus, intussusception and malignancy may be seen in adults with gastrointestinal duplication (4). This case report aimed to evaluate the clinical and histopathologic features of gastrointestinal duplication and its treatment along with the literature.

## CASE PRESENTATION

A 20-year-old single female was admitted to our emergency department with complaints of abdominal pain, nausea, vomiting, and weight loss. Her past medical and family history was unremarkable. Her complaints were present for two weeks. She had experienced ongoing attacks for nearly a year, has applied several times to emergency departments with no diagnosis. She also experienced weight loss during this period. Her pain was located in the umbilicus and left lower quadrant. A year ago, she had had a dark stool and diarrhea.

The patient's abdominal examination was unremarkable except for mild distention and tenderness in the left lower quadrant. Laboratory results showed WBC 9580/mm<sup>3</sup>, 34% hematocrit, platelets 450,000 and CRP of 1 mg/L, and biochemical results were normal. Abdominal X-ray showed a few intestinal gas shadows. Ultrasound (US) was unremarkable. She has previously underwent four abdominal tomography and one contrast-enhanced magnetic resonance (MR) imaging, an enteroclysis and three abdominal US examinations within the past one year. These evaluations reported that there may be a closed fistula between small bowel loops due to Meckel's diverticulitis or inflammatory bowel disease, and mild bowel wall thickening.

She was taken to surgery based on these findings. The abdomen was entered with a supra-umbilical median incision. The exploration revealed thickening of the small bowel mesentery for a segment of 35-45 cm, located approximately 60-70 cm proximal to the terminal ileum, and slight granulation tissue in the bowel serosa in one area (Figure 1). Approximately 40 cm of small bowel loop was resected and an anastomosis was performed along with appendectomy. There were no postoperative complications. The dissection of the specimen revealed a duplication. An intramesenteric located small bowel loop of approximately 22 cm length that was linked to the ileum in its distal portion with a blind ending in its proximal part was detected (Figure 2).

When CT images were retrospectively analyzed, typical tubular ileal duplication was identified (Figure 3, 4).

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Figure 1. Intramesenteric located duplication

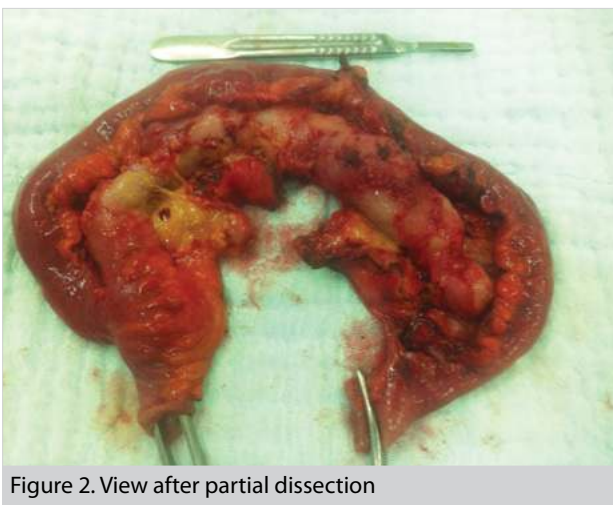


Figure 2. View after partial dissection

The pathology report was reported as tubular small bowel duplication with tissue sections lined with gastric type epithelium and presence of heterotopic pancreas in the per-intestinal fatty tissue. Intestinal metaplasia was not detected.

The patient was informed on the case report to be published in a scientific journal, and a consent document was obtained.

#### DISCUSSION

Fitz (4) defined GI duplication for the first time. In 1937, Ladd defined GI duplication as the presence of a well-developed smooth muscle layer and an inner epithelium similar to digestive tract epithelium in close proximity to the gastrointestinal system. GI duplications are frequent in the small intestine and are most common in the ileum (1, 5). In our case, the duplication was located in the ileum. Its incidence is generally one in every 10,000 live births (4). It may be of the cystic or tubular type, but the tubular type is rarer (5, 6). In our case, the duplication was of the tubular type. Unlike Meckel's diverticulum, they are located within the mesentery. Approximately 80% of patients manifest with symptoms before two years of age, the rest may remain asymptomatic and undiagnosed until adulthood (2). Our case was a 20-year-old adult patient who remained undiagnosed despite a number of evaluations. The incidence rate at autopsy series was reported to be 1/4500 (1).



Figure 3. Duplication in the left lower quadrant on coronal images

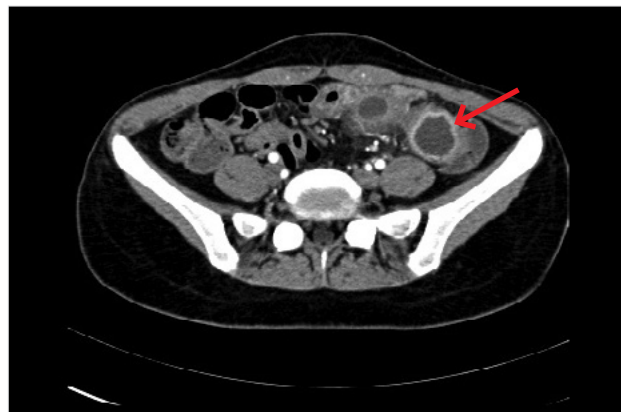


Figure 4. View of the lower left quadrant mass on axial images

Symptoms of gastrointestinal system duplications vary. The most common symptoms are abdominal pain, vomiting, distention, palpable mass, and hemorrhage (2, 4, 6, 7). The diagnosis of GI duplications, especially of the ileum, can be difficult since endoscopic examination of the ileum is challenging. Barium X-ray, US, and CT may play an important role diagnosis. Duplications can be distinguished from other abdominal cystic mass lesions by containing normal gastrointestinal mucosa, nevertheless 1/3 of duplications contain ectopic gastric mucosa (8). Ectopic gastric mucosa can lead to peptic ulceration, bleeding, perforation and fistula formation. <sup>99m</sup>Tc pertechnetate is retained by gastric mucosa and it can display bowel duplications containing ectopic gastric mucosa depending on the width of the mucosa (9). In our case, pathologic evaluation of the specimen revealed ectopic gastric mucosa, and a history of dark stool.

Even if it is reported that laparoscopy can be used in the diagnosis of recurrent abdominal pain, we believe that the role of laparoscopy may be limited in the diagnosis of patients with lesions within the mesentery, as in our case.

Duplications in children are benign, however malignant transformation has been reported in adults, although rare (1, 4).

Despite advances in diagnostic methods, the diagnosis can sometimes be only made after surgical procedures as in our case. GI duplications can cause volvulus, intussusception, bleeding and perforation. Hypertrophic ileal duplication, lymphoma, GIST and Crohn's disease should be considered as part of differential diagnosis of ileum duplications.

The ideal surgical technique is complete resection, while simple excision and partial resection can also be applied. Asymptomatic patients who are incidentally detected can be non-operatively monitored. We completely resected the 22 cm in size duplication along with the accompanying segment of the small intestine. Duplications may also include pancreatic mucosa besides gastric mucosa, as mentioned above. In our case, the pathologic examination revealed heterotopic pancreatic tissue.

### CONCLUSION

We believe that in case of unexplained abdominal pain, gastrointestinal duplications should be kept in mind as part of differential diagnosis even in adults, and once recognized complete resection is the appropriate treatment due to the risk of malignancy.

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**Informed Consent:** Written informed consent was obtained from patient who participated in this case.

**Peer-review:** Externally peer-reviewed.

**Conflict of Interest:** No conflict of interest was declared by the authors.

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