CASE REPORT

# Perforated Ileal Duplication Cyst: A Diagnostic Dilemma

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## ABSTRACT

Alimentary tract duplications are rare congenital anomalies. We report a case of a 14-year-old boy, who presented with acute abdomen, with features favouring acute appendicitis. Laparotomy revealed perforated ileal duplication cyst. Resection of ileum along with cyst and anastomosis was undertaken, and proximal ileostomy performed. Postoperative recovery was uneventful and ileostomy was reversed successfully. Histopathology revealed the presence of gastric mucosa at the site of perforation.

**Key words:** *Enteric duplication. Acute abdomen. Ileal duplication cyst. Ectopic gastric mucosa.*

## INTRODUCTION

Enteric duplications are rare congenital anomalies that can occur anywhere in the gastrointestinal tract, with the ileum being the most usual site.[1] They are almost always situated on the mesenteric aspect of the alimentary tract, sharing a common blood supply and muscular coat with the adjacent bowel, while having a separate mucosal lining.[2] These duplications mostly present with abdominal mass, intussusception, bowel obstruction and/or melena,[3] but perforation of duplication resulting in peritonitis is a rare presentation.[4] This report describes a case of perforated ileal duplication that was partly hanging on a mesentery and partly adherent to the adjacent ileum having a common muscular coat.

## CASE REPORT

A 14-year-old boy was admitted in emergency with complains of pain in right iliac fossa, vomiting and low grade pyrexia for 15 days, with history of taking antibiotics and analgesics. He looked toxic and dehydrated, with blood pressure of 90/60 mmHg, heart rate of 100 beats/minute, and body temperature of 99.7$^{o}$F. He had no significant past medical or surgical history.

Clinical examination revealed severe tenderness and guarding in right iliac fossa along with mild tenderness in lower abdomen. Bowel sounds were sluggish.

Complete blood count showed haemoglobin level of 13.3 g/dl and white cell count of 7.95x10$^{9}$/L with neutrophil count of 76.8%. His blood urea level was 43 mg/dl, and serum electrolytes were within normal range. Ultrasonography revealed a small collection with internal echoes in the pelvis and hepato-renal angle, and dilated bowel loops in right iliac fossa. Plain X-ray abdomen showed localized ileus in right iliac fossa.

On clinical assessment, perforated appendicitis was diagnosed, and after resuscitation the peritoneal cavity was entered through grid iron incision. Approximately 200 ml of purulent fluid was aspirated from pelvis and right iliac fossa, but appendix was found to have serosal congestion only, not correlating with the clinical picture. Further exploration revealed a 45 cm long and 3 cm wide tubular structure consistent with small intestine. Its proximal 25 cm hanged on a short mesentery arising from the ileal one, and was blind ended (Figure I), while the distal segment was adherent to the ileum on its mesenteric border sharing a common wall, and opening into it approximately 10 cm proximal to ileo-caecal junction. There was a 4 cm wide perforation, approximately in the middle of the duplication cyst where it joined the ileum.

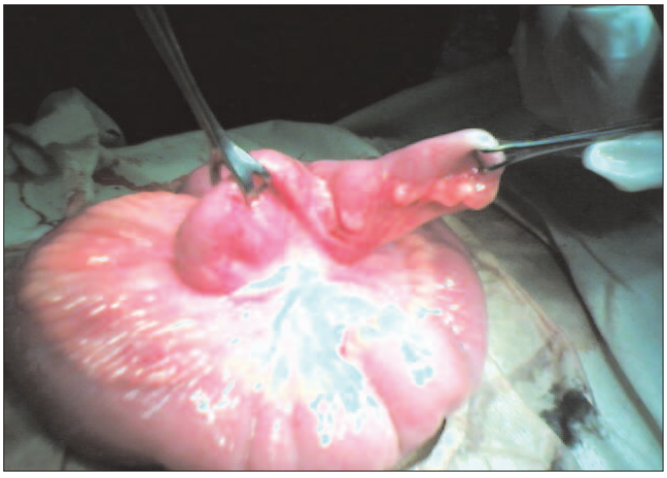

**Figure 1:** Proximal part of duplication, hanging on mesentry.

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The proximal segment was resected free from ileal mesentery without jeopardizing the ileal blood supply. The distal portion of the cyst, adherent to the ileum, was resected along with it, and ileal continuity restored. The presence of thick pus prompted for covering loop ileostomy. The resected ileal loop looked double barrelled, and the surgical diagnosis was perforation of duplicated segment of ileum.

Pus was sent for culture and sensitivity. Postoperatively, the patient was kept on Ceftriaxone and Flagyl. The culture report revealed *E. coli* sensitive to the given antibiotic. Recovery was smooth and event free.

Histopathological findings revealed that the duplicated segment had a muscular layer similar to that of the small intestine, and was lined partly by intestinal and partly by gastric mucosa, showing areas of ulcerations (Figure 2). The area around the perforation contained gastric mucosa, suggesting that ectopic gastric mucosa caused ulcerations that led to perforation.

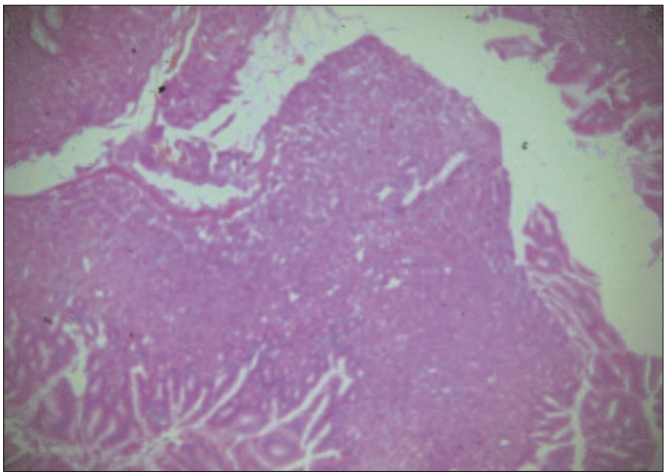

**Figure 2:** Microscopy, showing gastric and intestinal mucosa.

## DISCUSSION

Enteric duplication cysts are rare congenital abnormalities that can occur any where in the gastrointestinal tract, from tongue to anus, the ileum being the most common site. They are located adjacent to some part of the gastrointestinal tract, having smooth muscle in their wall, and lined by mucosa similar to that or some other part of the alimentary tract.[4,5] They may contain more than one type of mucosa including ectopic gastric mucosa, or pancreatic tissue.[2]

Morphologically, they may be spherical or tubular; the spherical type being more common in the small intestine.[6] Each type is further categorized as: communicating, where its lumen opens into the adjacent intestine; and non-communicating. Tubular duplications are often of the communicating type, connecting at proximal and/or at distal end.[6] In this case it was a tubular duplication of ileum that communicated at its distal end.

Most of these duplications manifest as abdominal mass, intussusception, bowel obstruction and/or melena.[3,7] However, a few reports of perforations leading to peritonitis have been made,[3] as seen in this patient.

Ulcerations due to ectopic gastric mucosa frequently result in melena,[7] while asymptomatic progression to perforation is rarely seen, as in this case. Peptic ulceration of ectopic tissue frequently results in unusual and misleading symptoms, making the diagnosis difficult. $_{99}Tc_m$-pertechnetate scanning can detect the ectopic gastric mucosa,[8] but cannot exclude Meckel's diverticulum. Moreover, the time limitation in acute abdomen makes this procedure impossible to employ. Abdominal sonography, though a useful diagnostic tool in detecting spherical duplications, cannot detect the tubular type.[4]

With regard to the management of enteric duplications, two important concerns are a common blood supply between duplication and adjacent bowel to avoid excessive resection of normal bowel, and the presence of ectopic gastric mucosa negating the internal drainage of the duplication, due to a risk of ulcerations and haemorrhage.[7] Resection of duplication alone is difficult due to a common blood supply shared with the normal bowel as well as being adherent to it. The procedure of choice is removal of duplication along with resection of a short segment of normal intestine.[4] When short gut syndrome is feared from extensive resection due to long tubular duplication, mucosectomy is an excellent alternative, as the gastric mucosa does not regenerate.[7]

This case shows that, though rare, gut duplication anomaly may be encountered while dealing with an acute abdomen, posing diagnostic as well as management confusion.

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