

## CASE REPORT

# Jejunal Duplication Cyst - a Rare Cause of Massive Gastrointestinal Bleeding: A Case Report

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### Introduction

Alimentary tract duplication is a relatively rare congenital anomaly. It can be symptomatic or are discovered incidentally. W.E Ladd first introduced the term duplication in 1934. Most duplications are benign, but the presence of ectopic gastric mucosa and the potential for malignant degeneration remain a concern.<sup>1</sup>

Congenital duplication cyst can occur anywhere in the gastrointestinal(GI) tract, although it most commonly occurs in the ileum, oesophagus and colon.<sup>1</sup> Approximately 75% of duplications have been reported to be located within the abdominal cavity, whereas the remaining is intrathoracic (20%) or thoracoabdominal (5%). Ileal lesion are the more common (53%) followed by mediastinal (18%), colonic (13%), gastric (7%), duodenal (6%), rectal (4%), oesophageal (2%), cervical (1%) lesion. They may present as solid or cystic tumours, intussusception, perforation or gastrointestinal bleeding.<sup>2</sup> Most intestinal duplication lie on the mesenteric side of the intestine and share a common muscular wall and blood supply with the native bowel.<sup>3</sup> Appropriate surgical management is required, for this the attending surgeon should be familiar with the pathology and clinical characteristics of these rare cysts.<sup>3</sup> Due to the rarity of this condition the vast majority of literature on enteric duplication cysts is in the form of case reports. Very few case series have been published previously.

### Case report

A 12 months old boy, presented with complaint of melena 1-2 times/day, 2-3 days interval for last 2 months (figure 1). He had no history of fever, vomiting, abdominal pain, abdominal distension or cow's milk allergy, ingestion of offending drugs/ toy/button. He received PRBC transfusion 5 times during the course of illness. On examination baby was sick looking, severely pale, vital: normal, well thriving and abdomen was nontender, no organomegaly, bowel sound present. Other systemic examination reveal normal.

Laboratory parameters showed severe anemia (Hb% 6.8 gm/dL), normal platelet count, on PBF-normocrocytic anemia, feature of colitis on stool routine examination ( RBC: 6-8/HPF, pus cell: 5-6 /HPF), positive occult blood test, ultrasonography of whole abdomen was suggestive of duplication cyst or Meckel's diverticular cyst at right iliac region. Meckel's scan showed heterotopic gastric mucosa in gut wall in hypogastrium. Esophagogas-troduodenoscopy was normal. Barium meal & follow through showed a large globular compressive effect that compressing distal bowel loops with gross narrowing of rectosigmoid region. Barium enema was normal (Table-I).

After correction of anaemia, laparotomy was done. A duplication cyst was seen 10 cm from duodenojejunal junction, measuring about (10x8) cm arising from

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**Received:** 9 January 2018; **Accepted:** 22 February 2018

**Table I**  
*Laboratory Investigations*

CBC	Hemoglobin	6.8 gm/dL
	Total count of WBC	9000/mm <sup>3</sup>
	Platelets	380000/mm <sup>3</sup>
PBF	Normocytic anemia ,otherwise normal.	
Stool routine examination	RBC	6-8/HPF
	Pus cell	5-6/HPF
Occult blood test	Positive	
USG of whole abdomen	Suggestive of duplication cyst or Mickel diverticular cyst intestinal atresia fluid filled dilation area measuring 7.6cm x 5.9cm seen at right iliac region	
Mickel's scan	Heterotopic gastric mucosa in gut wall in hypogastrium	
Esophagogastroduodenoscopy	Normal	
Digital Barium meal of stomach, duodenum & follow through	A large globular compressive effect is seen in lower part of abdomen little towards left, compressing distal bowel loops with gross narrowing of rectosigmoid region. Possibility of cystic mass in lower part of abdomen is to be considered	
Barium enema	No organic lesion in large gut	
Histopathology	Two mucosal layers sharing a submucosal within muscular layer .Gastric mucosal lining is seen. No malignancy found.	

upper part of jejunum. Excision of duplication cyst and adjacent gut with end to end anastomosis was done. Postoperative period was uneventful. He was discharged on the 10<sup>th</sup> postoperative day. The resected intestinal loop was sent for histopathological examination (figure 3).



**Fig 1** (a) Patient (b) Melena



**Fig 2** Barium follow through



**Fig 3** Resected duplication cyst

### Discussion

Jejunal and ileal duplications are the most common alimentary tract duplication and are usually found on the mesenteric border of the intestine.<sup>5</sup> Duplication parts usually share a common muscularis and common blood supply with the adjacent gut. There are two types of duplications- cystic and tubular. 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 fill with secreted mucus and cause pain and usually presents 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 in a caudal type of communication the duplicated part empties readily. Cystic or secular type of duplications usually have 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 the ectopic gastric mucosa can cause peptic ulcer,

bleeding and even perforation with peritonitis.<sup>6-8</sup> Ectopic pancreatic tissue can also be found in the wall of some of these duplications.<sup>9</sup> The sign-symptoms of duplication depends on its type and location. Symptoms usually begin from early childhood.<sup>9</sup> Our patient presented at 12 months of age.

Mass in the abdomen, pain from distension of cyst and intestinal obstruction due to compression of adjacent intestine may be the presenting complaints. Segmental intestinal vascular obstruction can lead to gangrene of adjacent normal bowel. Bleeding from ulceration either within the duplication itself or in the adjacent normal intestine occurs in approximately 20% of patients.<sup>10, 11</sup> Our case presented with painless gastrointestinal bleeding in the form of melena. He was moderately pale with history of 5 unit blood transfusion, which signified massive blood loss.

The causes of intestinal duplication cyst have not yet been established. Several theories have been put forward to explain different types of duplications. Of these persistence of fetal gut diverticula, defects in re-canalization of the solid stage of primitive gut, partial twinning and the split notochord theories are popular.<sup>12,13</sup> Bentley et al suggested that a split notochord that resulted from abnormal adhesions presenting between the ectoderm and endoderm was the primary defect, and that a herniation of the Yolk sac between the two halves of vertebra resulted in the subsequent duplication of the gut. However, the split-notochord theory cannot explain formation of the intra-abdominal duplications that are not associated with spinal deformity.<sup>1</sup>

The clinical diagnosis of duplication may be difficult before surgery. Abdominal ultrasonography and in difficult cases CT scan can detect duplication. Enteric duplication cyst has an echogenic inner lining due to mucosal layer and a surrounding hypoechoic rim due to muscular layer and when identified together preoperatively, combinations of these two layers are highly suggestive of enteric duplication cysts.<sup>14</sup> The presence of these two layers helps to exclude other cystic mass such as mesenteric or omental cyst, choledochal cyst, ovarian cyst, and pancreatic pseudocyst.<sup>15</sup> In our patient ultrasonography of whole abdomen was suggestive of duplication cyst or Meckel's diverticular cyst.

Technetium scan demonstrates ectopic gastric mucosa in duplication and is helpful in cases of unexplained gastrointestinal bleeding. In our case Meckel's scan showed heterotopic gastric mucosa in gut wall in hypogastrium.

The management of symptomatic duplication is surgical. Laparotomy was done in our case and a duplication cyst was found arising from upper part of jejunum. Excision of duplication cyst and end to end anastomosis was done.

Infants with subclinical intestinal duplication discovered incidentally on scan are commonly offered surgery to prevent the future risk of complication by obstruction or perforation.

### Conclusion

Though duplication cyst is a rare cause of gastrointestinal bleeding, jejunal duplication cyst may present with massive gastrointestinal bleeding should be kept in mind during managing of such a case of massive gastrointestinal bleeding.

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