



Enteric Duplication Cyst Presenting as Volvulus: Two Case Reports

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Enteric duplication cysts are congenital malformations of the gastrointestinal tract. As the name duplication cyst suggests, which mimics the native gastrointestinal tract, having all the three layers of the gastrointestinal tract. Enteric duplication cyst most frequently occurs in the small bowel, particularly the ileum, but can occur anywhere along the gastrointestinal tract from tongue to rectum. An incidence of 1 in 4500 births, with a slight male preponderance. 80 % of the duplication cysts are symptomatic within first 2 years of life. Due to the varied clinics presentation, radiological evaluation is required for the diagnosis.

Herein, we are reporting two cases of enteric duplication cyst. The first case of 25 years old male patient presented to our center, with the features of acute intestinal obstruction due to volvulus. He was having a duplication cyst at the terminal ileum, causing volvulus. Treated by explorative laparotomy and second case of a 7 years old boy, diagnosed on CT scan, as an enteric duplication cyst, was treated by laparoscopic surgery.

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1. INTRODUCTION

The first case of enteric duplication cyst was reported by Calder in 1733. After that Ladd proposed or coined the term duplication cyst of the alimentary tract in 1930. Clinically, there are two types of intestinal duplication cyst.

1. Cystic duplication cyst is 80% of the cases, they are spherical in shape and not communicating with the bowel lumen and contains 20% gastric ectopic tissue in it.
2. Tubular duplication cysts account for 20% of the cases and communicate directly with bowel lumen [1,2,3,4].

Duplication cysts are attached to the normal alimentary tract and sharing three layers of bowel.

1. The cyst contains mucosa and submucosa.
2. The cyst is surrounded by smooth muscle layer.
3. The cyst must have a common wall and common blood supply.

Enteric duplication cysts are more common in the ileal mesentery 44 %, mediastinum 18%, colon 13%, gastric 7%, duodenal 6%, rectum 4% and oesophagus 2%. Enteric duplication cysts may present with abdominal pain, palpable mass, vomiting, bleeding due to heterotopic mucosa and acute intestinal obstruction due to volvulus or intussusception [5,6,7]. Ultrasonography showing the “gut signature signs” of a duplication cyst. The definitive treatment is open laparotomy or laparoscopic surgery which has excellent results [1,2,8,3].

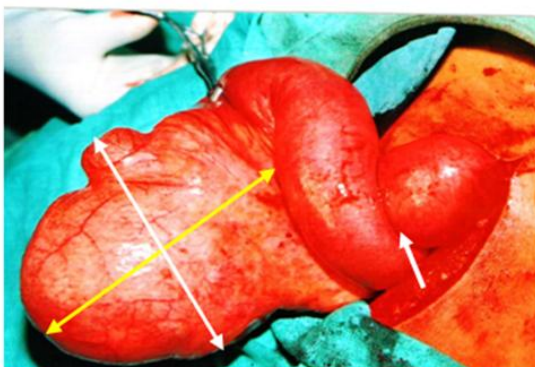
2. CASE REPORTS

2.1 Case 1

A 25 years old male patient was admitted as emergency case on 25/09/2009 at our centre, with complaints of severe abdomen pain, distension of abdomen, vomiting and constipation for 2 days. Physical examination of the patient, the abdomen was diffusely distended with exaggerated bowel sounds. All laboratory investigations were normal, including kidney function tests. Plain abdominal x-ray revealed multiple air-fluid levels suggestive of small bowel obstruction.

After receiving informed consent, patient underwent explorative laparotomy with a provisional diagnosis of acute small bowel obstruction. Abdominal exploration revealed dilated small bowel loops with a cystic mass measuring about 10x10x8 cm in size and spherical in shape at terminal ileum, causing twisting and volvulus of small bowel. After untwisting the cystic mass, there was no ischemia or gangrene of the small bowel. Resection of the cystic mass along with ileal segment with end to end anastomosis was performed.

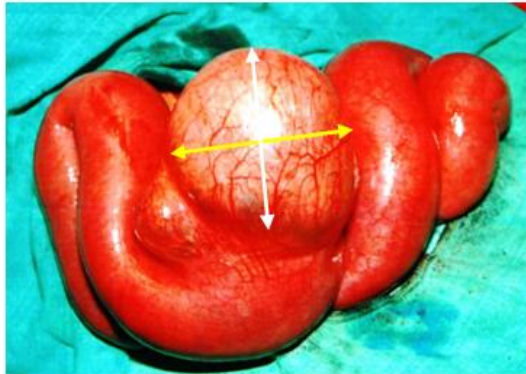
On gross examination of specimen, a big spherical cystic mass 10x10x8 cm of size. The histopathology showed a blind cystic mass lined by mucosal epithelium suggesting an intestinal duplication cyst. The patient was discharged from the hospital on 8th postoperative day with uneventful postoperative course. The patient was followed up after one year and he remained asymptomatic (Case 1. Figs. 1, 2, 3, 4).



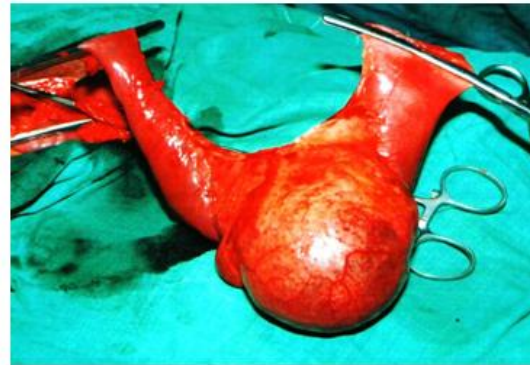
Case 1 Fig-1 Intraoperative photographs showing Volvulus of small bowel due to duplication cyst



Case 1 Fig-2 Intraoperative photographs showing untwisting of volvulus of small bowel



Case 1 Fig-3 Intraoperative photographs showing enteric duplication cyst of size 10x10x8 cm



Case 1 Fig-4 photographs showing total excision of enteric duplication cyst

2.2 Case 2

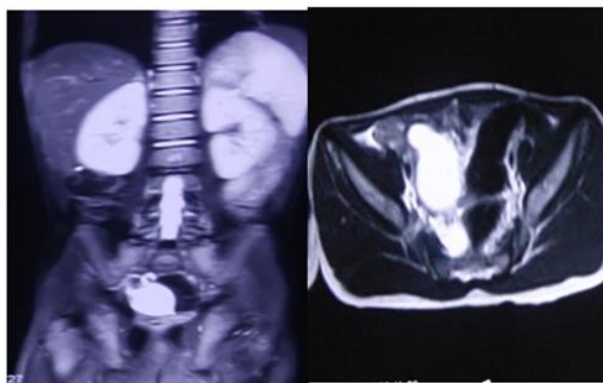
A 7 years' boy was admitted to our centre on 29/03/2017 with complains of abdominal pain and his CT abdomen showed the possibility of intestinal duplication cyst attached to the ileum. All laboratory investigation was normal including kidney function tests.

Under general anaesthesia abdomen was painted and draped. A 5 mm umbilical incision taken and Pneumoperitoneum created by varies needle and 5 mm port was inserted. There was a cystic lesion 5x3 cm, arising in the ileum and confirms the diagnosis of a duplication cyst. An additional 3 mm port inserted at right upper side of the public symphysis and left lower abdomen Mc-Burning point and 3 mm trocar was placed in each incision. An intestinal clamp and dissecting forceps were placed. Pneumoperitoneum was established with the pressure of 10-12 mm of Hg. Laparoscopically we checked the mobility of

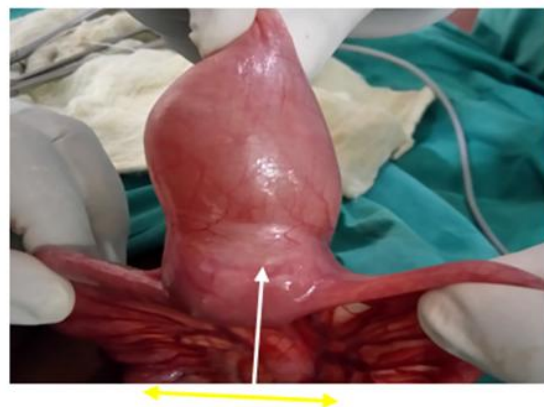
duplication cyst and it was freely mobile, no adhesions to the adjacent structure.

We decided to do TULA, which means Trans-umbilical laparoscopic assisted extracorporeal small bowel resection along with the duplication cyst and ileo-ileal anastomosis. We extended the umbilical incision and applied a self-retaining wound protector applied. We brought the cyst outside, in extracorporeal segmental resection of the cyst along with ileal segment were carried out. Stapler anastomosis done and haemostasis confirmed. The intestine was relocated in the abdominal cavity and the umbilical incision was closed with sutures, and all ports were closed.

Post-operative recovery was excellent with hospital stay 5-6 days. On gross examination of ileal cyst of size 5x3 cm and attached the ileum. Histopathological diagnosis was intestinal duplication cyst. Post-operative follows up after 2 months, with no scar of the abdomen and boy was healthy. (Case 2. Figs. 1, 2, 3, 4, 5).



Case 2 Fig-1 CT Abdomen showing enteric duplication cyst in the abdomen



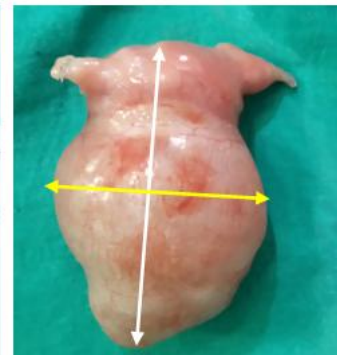
Case 2 Fig-2 Intraoperative photographs showing enteric duplication cyst of size 5x3 cm



Case 2. Fig. 3. Self-retaining wound protector Trans-umbilical laparoscopic assisted (TULA) Extracorporeal Excision



Case 2. Fig. 4. Stapler ileal anastomosis



Case 2. Fig. 5. Total excision of enteric duplication cyst of size 5x3 cm

3. DISCUSSION

A duplication cyst of gastrointestinal tract is a rare congenital anomaly found in about 0.2% of all children. Enteric duplication cyst must have three essential characteristics are 1. A well-developed smooth muscle coat. 2 mucosal linings found within the cyst. 3 Continuity in any segment of the alimentary tract [2,8].

Although the exact aetiology is unknown, several theories have been proposed like split notochord theory, partial twinning theory, persistent embryological diverticula theory or aberrant luminal recanalization theory. In infants or neonates present with abdominal pain, vomiting, bleeding, abdominal mass intestinal obstruction due to volvulus or intussusception. Some duplication cysts may remain asymptomatic till adulthood [1,2,8].

Ultrasonography in the imaging method of choice in the diagnosis of enteric duplication cysts. Classical findings, include the presence of a cyst in relation of the two the gut with double wall or muscular rim sign i.e. "gut signatory sign", which is caused by the inner hyperechoic mucosa and the outer hypoechoic smooth muscle layer. Ultrasonography sensitivity is only 20-30% CT Scans are more useful in demonstrating the precise anatomical relationship between the cysts and surrounding structures. Ultrasonography and CT Scan are able to identify the cystic masses in the abdomen. Histopathological examination played an important role in confirming the diagnosis of duplication cyst [2,3,4].

The definitive treatment for a duplication cyst is surgery. The surgical approach should be open laparotomy or laparoscopic surgery; it depends

on the expertise of the surgeon. Emergency surgery is recommended in all the symptomatic cases. Laparotomy with excision of cyst along with the ileal segment and end to end anastomosis [4,9,10].

4. CONCLUSION

Enteric duplication cysts are a rare congenital entity of the gastrointestinal system. Resection of the cyst along with involved segment of the bowel offers definitive treatment.

CONSENT

As per international standard or university standard, patient(s) and parental written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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