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Case Report

Enteric Duplication Cyst –A Rare Congenital Anomaly in Adolescence Presenting as Recurrent Pain Abdomen

Dr. Harleen Bawa[1*], Dr. Parthasarathi Hota[2]

[1]Post Graduate Year 2, Pacific Institute of Medical Sciences, Udaipur 313015
[2]MS, FMAS, Assistant Professor, Department of General Surgery, Pacific Institute of Medical Sciences, Udaipur, Ambua Rd, Umarda, Rajasthan 313015, India

***Corresponding Author**
Dr. Harleen Bawa
Post Graduate Year 2, Pacific Institute of Medical Sciences, Udaipur 313015

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Abstract: ***Case Report*:** Presented by Dr. Harleen Bawa and Dr. Parthasarathi Hota, this case involves an 18-year-old male with recurrent abdominal pain, nausea, and fever. Initially suspected of congenital gastrointestinal malformation at 1 year, the patient underwent conservative management until symptoms reappeared. Imaging revealed blind-ended tubular structures attached to the small bowel, prompting further investigation for a definitive diagnosis. ***Management*:** An exploratory laparotomy revealed an enteric duplication cyst in the ileal region, 50 cm from the ileocecal junction. Resection of the involved ileum with end-to-end anastomosis was performed. Mesenteric lymphadenopathy and an inflamed appendix were also addressed. Biopsy results indicated changes consistent with chronic appendicitis. In Radiological findings CT images displayed well-defined blind-ended tubular structures arising from the distal ileum, suggesting Meckel's diverticulum. Fluid density cystic lesions near the duodenum and pancreas raised the possibility of an enteric duplication cyst. ***Discussion*:** Enteric duplication cysts, though rare, can manifest with varied symptoms, including recurrent abdominal pain and intestinal obstruction. Imaging techniques such as CT, USG, and technetium pertechnetate scanning aid in diagnosis. Resection of the cyst and adjacent intestine with end-to-end anastomosis is a common surgical approach. The prognosis is excellent when complete excision is possible without compromising intestinal length. ***Conclusion*:** Enteric duplication cysts, located along the gastrointestinal tract, may present challenges in diagnosis and management. Surgical intervention, guided by imaging and clinical findings, often leads to favorable outcomes. Awareness of potential complications, such as bowel obstruction and bleeding, is crucial for timely intervention.

Keywords: Enteric Duplication Cyst, Congenital, Meckels Diverticulum, Exploratory Laparotomy, Ileal Resection, Mesenteric Lymphadenopathy, Gastrointestinal Malformation, Intestinal lumen Duplication, Rare adolescence presentation, Resection Anastomosis.

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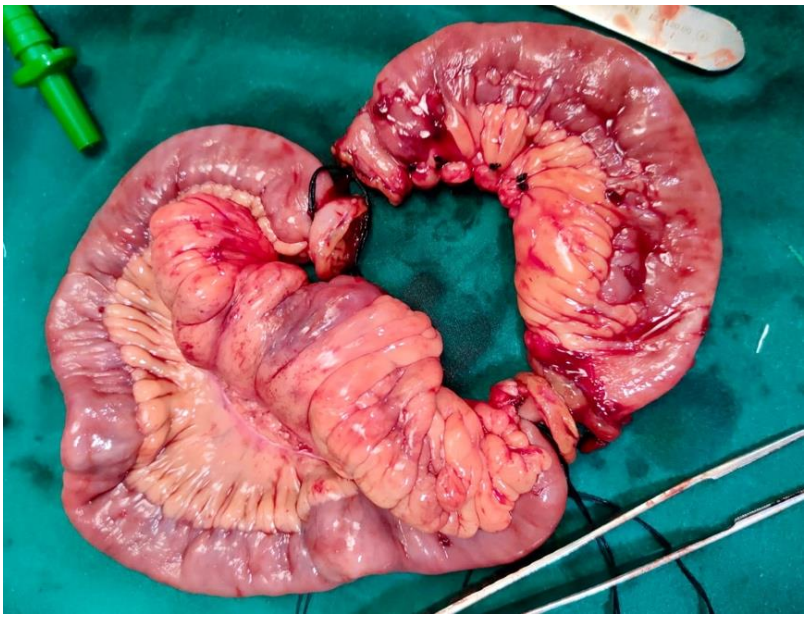
INTRODUCTION

Enteric duplication cysts (EDCs) are rare congenital malformations formed during the embryonic development of the digestive tract [1]. They are usually detected prenatally or in the first years of life. The size, location, type, mucosal pattern, and presence of complications produce a varied clinical presentation and different imaging findings [2]. The incidence is 1:4,500 births, found in 0.2% of all children, with a slight male predominance. They most commonly occur in the ileum, oesophagus, and colon [3]. They may be contained within the gastrointestinal tract wall or extrinsic to it. Duplication cysts can also be cystic (80%) or tubular (20%) [2]. These are believed to occur between the 4th and 8th weeks of embryonic development [1]. Their aetiology is still unknown; several theories have been proposed to explain their pathophysiology, but no single hypothesis can justify all duplications, locations, and associated anomalies. Split notochord theory is often postulated. The luminal recanalisation theory explains duplications in those portions of the GT that have a solid stage, including the oesophagus, small bowel, and colon; nevertheless, it does not explain duplications at other levels. The intrauterine vascular accident theory suggests that gastrointestinal duplications arise from an intrauterine vascular accident during early fetal development and may be a valid explanation for isolated duplication [5].

Associated anomalies such as spinal defects, cardiac, or urinary malformations, are reported with an incidence rate of 16–26%. Other digestive anomalies are present in about 10% of cases. Therefore, once an EDC is found, a search for other anomalies is needed [6]. An enteric duplication cyst must have three characteristics: an epithelial lining containing the mucosa of the alimentary tract, an envelope of smooth muscle, and the cyst must be closely attached to the GT by sharing a common wall [1].



CASE PRESENTATION

A 18-year young male presented with pain abdomen complaint episodically off and on since 1 year. These episodes were frequently accompanied with nausea and fever. The patients' family was informed of the possibility of congenital gastrointestinal malformation at 1 yr of age when the child was investigated for an illness. However, the patient was managed conservatively till symptoms reappeared. After which patient presented with complaints of recurrent episodes of pain abdomen and fever. On further examination, the patient had a mildly tender abdomen and a history of episodes of vomiting and bowel movement complaints. Further investigations were done to make a diagnosis, and USG W/A revealed two blind-ended tubular structures seen attached to the small bowel (Ileum) and appears inflamed noted in the umbilical region attached to the distal ileum and also noted in the left paramediam location. A differential diagnosis of Meckel's diverticulum was made after which a CECT whole abdomen was performed on which the possibility of enteric or duplication cyst was explained and the patient was managed as follows [7].

RADIOLOGY – CECT ABDOMEN

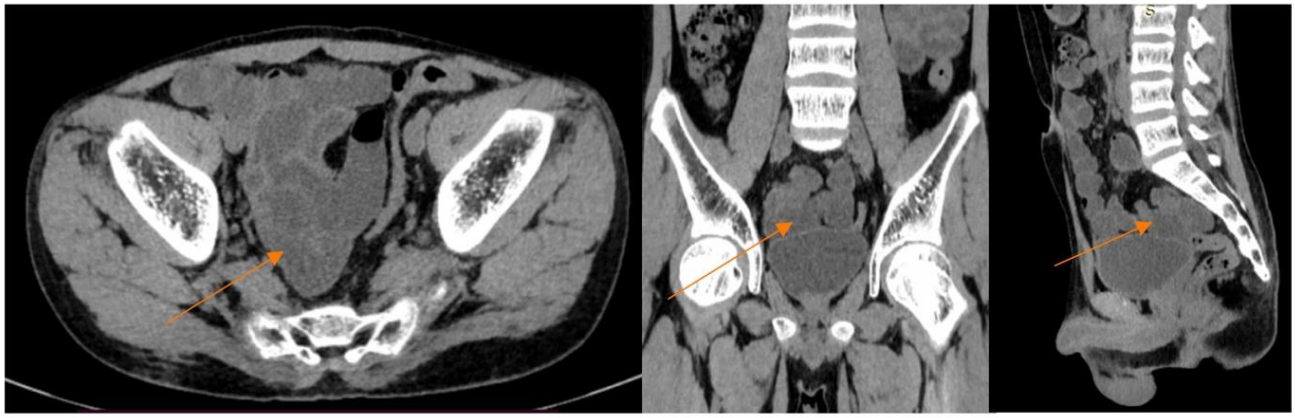


Figure 1: Well-defined blind-ended tubular structure (maximum AP diameter 25mm) with enhancing wall noted arising from the distal ileum and subcentric enhancing lymph nodes – possibility of Meckel's diverticulum. There are ill-defined irregular-shaped enhancements of the wall of this diverticulum at the tip and base – possibility of ectopic gastric mucosa or infective

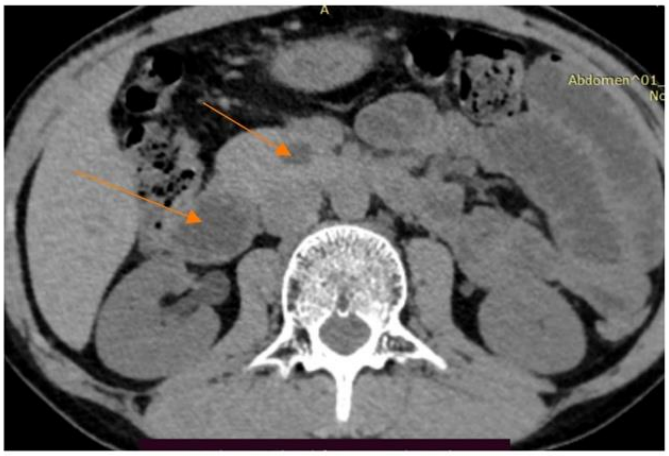


Figure 2: Fluid density cystic lesions measuring approximately 15 x 14 mm and 22 x 27 mm in close proximity to the 3rd part of the duodenum and the head and uncinate process of the pancreas, which are noted opacities by positive oral contrast – Possibility of enteric duplication cyst/ Pseudocyst

MANAGEMENT

The patient was operated with a midline incision, and exploratory laparotomy was done to find the intestinal duplication cyst in the ileal region, 50 cm from IC junction in the mesenteric border sharing a common blood supply, and hence resection of the involved part of ileum along with duplication dust was done with end-to-end anastomosis. Furthermore, duplication of approximately 15 cm of ileum containing two lumens as shown in Image 3 & 4 was also respected and anastomosed. On exploration, mesenteric lymphadenopathy was seen along with a long appendix of approximately 8 cm with an inflamed tip was seen, and Lymph node was excised for Biopsy, and Appendectomy was done.

The sample sent for biopsy contained a segment of ileum measuring 22.0 cm in length and 2 cm in diameter, an attached mass is seen at one end of the ileum measuring 11 x 4 x 3 cm. External surface smooth with some cystic area seen. On cut section - Though anti-Mesenteric border normal rougosity present, on cut section, the mass opening is seen in the ileum, few cynically dilated areas identified filled with clear fluid measuring 2 cm in diameter. The second segment of ileum measuring 12 cm in length and 2 cm in diameter – on cut section normal rougosity present similar to the ileum. Figure 5. The second sample was a single white, grey soft tissue piece measuring 1.5 x 1 x 0.5 cm on cut section of which grey homogenous area seen which was consistent with lymph nodes Figure 6. And the Third sample containing appendix measuring 7 cm in length and 0.3 cm in diameter, on cut section the lumen was patent, and no faecal material was seen. However, the findings were consistent with changes seen in Chronic appendicitis [7].

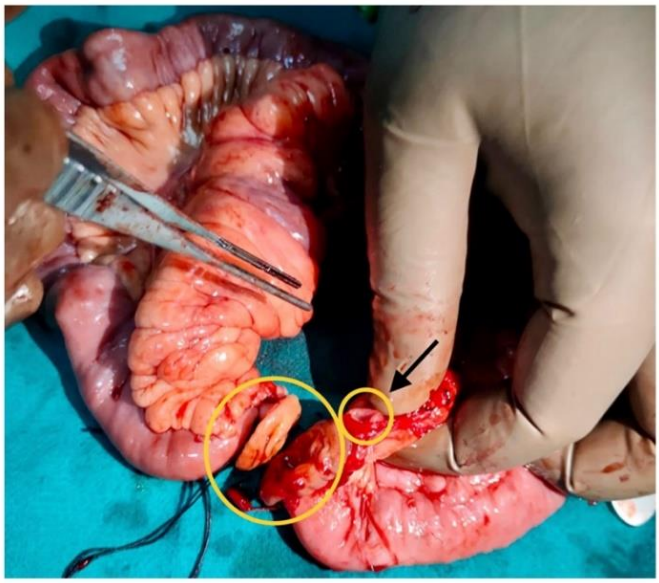
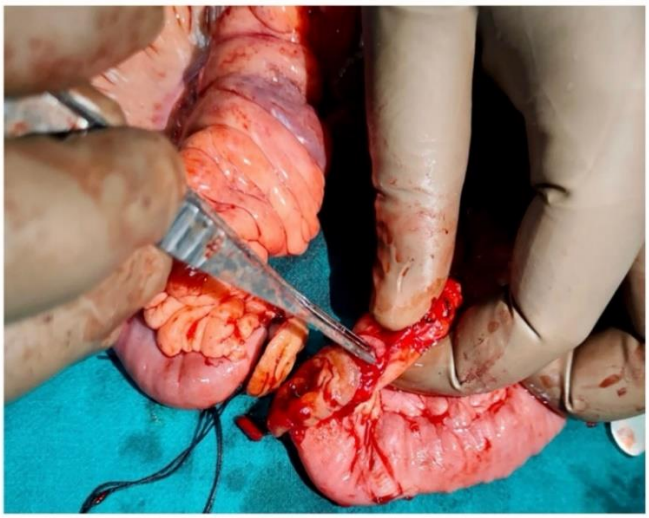


Figure 3:



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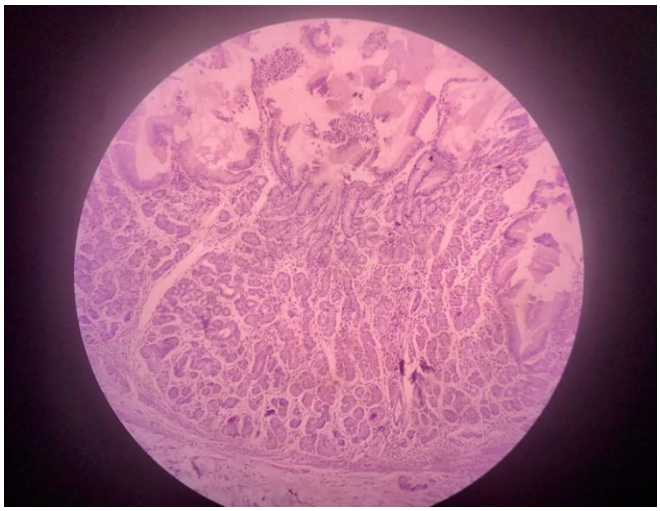

Figure 5:

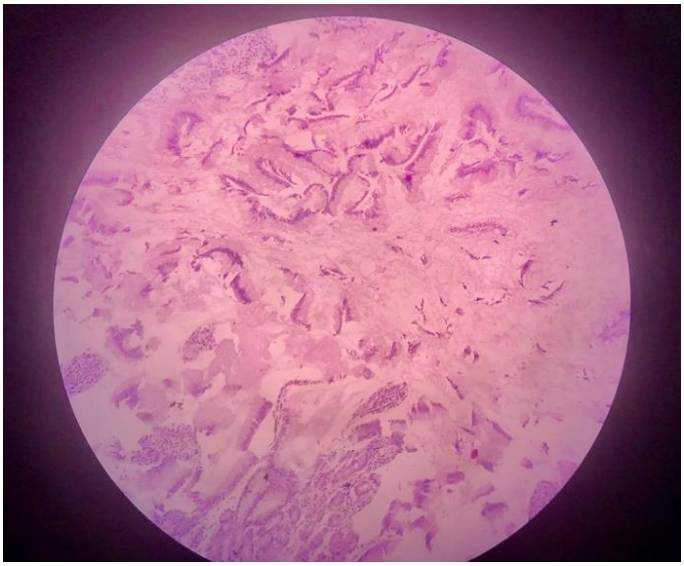

Figure 6:

DISCUSSION

Duplications represent mucosa-lined structures that are in continuity with the gastrointestinal tract [1]. Although they can occur at any level in the gastrointestinal tract, duplications are found most commonly in the ileum within the leaves of the mesentery. Duplications may be long and tubular but usually are cystic masses. In all cases, they share a common wall with the intestine. Symptoms associated with enteric duplication cysts include recurrent abdominal pain, emesis from intestinal obstruction, or hematochezia. Such bleeding typically results from ulceration in the duplication or in the adjacent intestine if the duplication contains ectopic gastric mucosa [9]. On examination, a palpable mass is often identified. Children may also develop intestinal obstruction. Torsion may produce gangrene and perforation.

The ability to make a preoperative diagnosis of enteric duplication cyst usually depends on the presentation. CT, USG, and technetium pertechnetate scanning can be very helpful [10]. Occasionally, a duplication can be seen on small bowel follow-through or barium enema. In the case of short duplications, resection of the cyst and adjacent intestine with end-to-end anastomosis can be performed. If resection of long duplications would compromise intestinal length, multiple enterotomies and mucosal stripping in the duplicated segment will allow the walls to collapse and become adherent. An alternative method is to divide the common wall using the GIA stapler, forming a common lumen. Patients with duplications who undergo complete excision without compromise of the length of the remaining intestine have an excellent prognosis.

Mesenteric cysts are similar to duplications in their location within the mesentery. However, they do not contain any mucosa or muscular wall. Chylous cysts may result from congenital lymphatic obstruction. Mesenteric cysts can cause intestinal obstruction or may present as an abdominal mass. The diagnosis may be made by abdominal US or CT. Treatment involves surgical excision. This may require resection of the adjacent intestine, particularly for extensive, multicystic lesions. In cases where complete excision is not possible due to the close proximity to vital structures, partial excision or marsupialization should be performed [9].

CONCLUSION

Enteric duplication cysts are benign cystic structures that are most commonly found at the mesenteric border of the jejunum or ileum, although they theoretically can form at any location along the gastrointestinal tract from the esophagus to the anus [1]. They may or may not communicate with the bowel segment with which they share a wall. Duplication cysts are often discovered incidentally but may occasionally cause complications such as bowel obstruction, volvulus, or intussusception. In a minority of cases, they contain gastric mucosa, which can lead to hemorrhage or perforation [9]. In the case of short duplications, resection of the cyst and adjacent intestine with end-to-end anastomosis can be performed. If resection of long duplications would compromise intestinal length, multiple enterotomies and mucosal stripping in the duplicated segment will allow the walls to collapse and become adherent. An alternative method is to divide the common wall using the GIA stapler, forming a common lumen. Patients with duplications who undergo complete excision without compromise of the length of the remaining intestine have an excellent prognosis [9].

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