

Various presentations, diagnosis, and management of enteric duplication cysts in high-volume tertiary healthcare center



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ABSTRACT

Enteric duplication cysts (EDCs), while having an incidence of 1 in 4500 live births, are not often diagnosed antenatally. Patients who are eventually diagnosed to have EDC have a varying presentation, demographically and clinically. EDCs are classified as cystic and tubular, with the latter being radiologically difficult to detect on ultrasound as compared to the cystic variant, and therefore requiring further investigations. With other differential diagnoses of mesenteric cysts and omental cysts, the clinical incidence of EDC is also reduced. This study was done over a 2-year period, where patients who were diagnosed with EDC at Sir Padampat Institute of Neonatology and Child Institute, Jaipur, were included in the study. They were monitored from the time of diagnosis, assessing their demographic data, pre-operative status, surgical management, post-operative treatment, and follow-up after discharge. A total of 14 patients were included in the study, of which a majority were tubular variants of ileal duplication cyst. Three patients had foregut duplication cysts (1 gastric and 2 esophageal). The age ranged from 3 days to 10 years, with the average age of 2.2 years. Each child had a varying presentation, some of them presenting with complications such as obstruction and perforation. The incidence was more in male children, as compared to females (3.6:1 – male: female). Two children had associated anomalies (one had a congenital diaphragmatic hernia and one had malrotation of the gut). In one child, a foregut duplication cyst was found to be communicating with a dilated segment of the jejunum, which was resected together. EDCs, while rare in occurrence, can have a myriad of presentations, and other associated congenital anomalies. Further studies are required to identify attributing risk factors and regional incidence along with better awareness for antenatal screening of EDCs.

Key words: Enteric duplication cysts; Obstruction; Perforation; Exploratory laparotomy; Resection and anastomosis; Marsupialization

INTRODUCTION

Enteric duplication cysts (EDCs) are congenital anomalies of the gastrointestinal tract, which can mimic other cysts of the abdominal cavity, such as mesenteric cysts and omental cysts. In literature, an incidence of 1 in 4500 live births¹ has

been mentioned. However, in a large population like that of India, this incidence may need to be reassessed. EDC, although a rare congenital anomaly, commonly presents as ileal duplication cysts. The other locations are mediastinal, colonic, gastric, duodenal, rectal, esophageal, and rarest, cervical. They are classified as tubular or cystic.

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EDCs also present with other anomalies or complications, which may be more emergent and severe. In our study, we demonstrate the varied presentations of duplication cysts, clinically and demographically. While EDCs may be detected antenatally, many of them may be detected in the first few years of life. They are believed to occur in the 4th–8th week of embryonic life. Although the exact etiology is still to be determined, several theories have been proposed to explain their pathophysiology. Theories such as split notochord theory, luminal recanalization, and incomplete twinning are popular hypotheses for the development of EDCs.

EDCs are rare congenital anomalies. However, we assessed the patients who were brought to our center and diagnosed with EDC, either preoperatively or intraoperatively. We wanted to demonstrate the various presentations of this congenital anomaly, and other associated anomalies that may mask a duplication cyst. This ambispective study, conducted over a 2-year period from September 2022 to October 2024, includes patients who presented to Sir Padampat Institute of Neonatology and Paediatric Health, Jaipur, and were diagnosed to have EDCs. These patients were assessed based on their demographic details, history and clinical presentation, associated anomalies, radiological findings, intraoperative findings, and management. They were also followed up in the post-operative period. The patients who were preoperatively suspected to have EDCs and were then intraoperatively found to have a different diagnosis were then excluded from the study. Few patients who were initially admitted or worked up for a different diagnosis or congenital anomaly and then intraoperatively detected to have an EDC were included in the study.

CASE 1

An 8-day-old female child presented with complaints of bilious vomiting and abdominal distension for 2 days, with no complaints of constipation. Antenatal scans were reported as normal. On examination, the abdomen was distended, with fecal staining on per rectal examination, and bilious aspirate on inserting NG tube. Ultrasound abdomen gave a differential diagnosis of the mesenteric cyst. Contrast-enhanced computed tomography (CECT) abdomen and pelvis detected a perforation with a cystic component as well. Intraoperatively, a tubular enteric duplication of the ileum was found, markedly inflamed bowel associated with perforation. Resection of the tubular duplication along with the perforated segment of the ileum was done. Mucosal stripping of the pathological segment bearing the common wall with ileostomy and distal mucous fistula was performed. Postoperatively, the child was started on oral feeds from the 2nd post-operative day (POD) once the stoma was functioning well, and supportive care in the form of probiotics and

nutritional supplements was given. Ileostomy closure was planned for the child but was then lost to follow-up (Figure 1).

CASE 2

A 5-year-old boy was brought to the outpatient department (OPD) with complaints of on-and-off abdominal pain for 6 months, with no significant medical or surgical history. Abdominal examination did not have any abnormal findings. Initial ultrasound revealed a possible enteric cyst, for which a CECT abdomen was done, which initially reported a tubular structure anterior to the hepatic flexure of the transverse colon, suspecting a colonic duplication cyst. Intraoperatively, a long tubular enteric cyst was found in the proximal ileum. Resection of the cyst with the adherent normal ileum was done with primary anastomosis of the bowel in a single-layer closure. The child was started orally after the 5th POD and was discharged on the 10th POD. The child has been on regular follow-up and has no post-operative complaints (Figure 2).

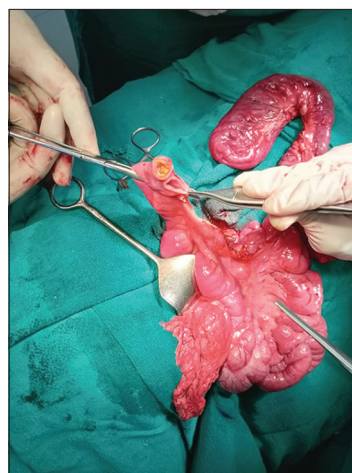


Figure 1: Double lumen with common wall seen of a tubular ileal duplication cyst

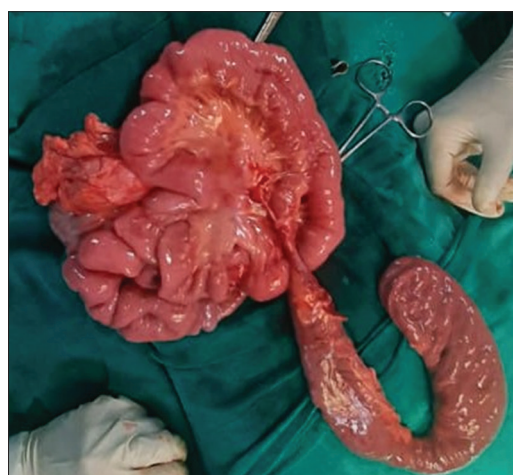


Figure 2: Long tubular ileal duplication cyst

CASE 3

A 1½-year-old male child was initially admitted under pediatrics with complaints of cough and respiratory distress. He was evaluated and found to have a left-sided congenital diaphragmatic hernia and associated foregut duplication cyst in the mediastinum, with right lung consolidation on HRCT thorax. Once the child was stabilized, he underwent a left diaphragmatic hernia repair. The foregut duplication cyst on the CT measures 31×28 mm and was on the medial aspect of the esophagus. It was not excised during the diaphragmatic hernia repair. The child had a prolonged intensive care unit (ICU) stay in view of the right lung consolidation and was on IV antibiotics for 14 days. He was then discharged on POD 15 and has had no complaints on follow-up.

CASE 4

A 2-year-old girl was brought to the OPD with complaints of on-and-off abdominal pain with intermittent episodes of vomiting. Per abdomen examination revealed no localized tenderness and no palpable mass. An ultrasound abdomen was done, which showed twisting of the mesentery along the mesenteric vessels and bowel loops at the level of umbilicus with a cystic structure of 7.4×3.4 cm seen in the right iliac fossa. The child underwent an exploratory laparotomy and was found to have a cystic type of ileal duplication cyst. Resection and anastomosis of the ileum were done. The child had a stable post-operative period and was discharged on the 7th POD tolerating full feeds. The child had no complaints on follow-up.

CASE 5

A 1-year-old boy was admitted with complaints of bilious vomiting for 7 days and abdominal distension for 3 days. The child was tachycardic and tachypneic on admission and was resuscitated and kept in the ICU. Erect X-ray abdomen showed multiple air fluid levels, suggestive of intestinal obstruction. An ultrasound of the abdomen was done and showed a midline volvulus with a sealed off collection of 38×27 mm below the umbilicus and another collection of 48×37 mm below the right iliac fossa. An exploratory laparotomy was performed, which revealed a terminal ileal duplication cyst causing obstruction and gangrenous changes to the native bowel. Resection of the ileum containing the cyst and the gangrenous bowel was done, with anastomosis of the ileum.

CASE 6

A 6-month-old female child was brought to the OPD with complaints of recurrent episodes of vomiting for 3–4 months, which was non-bilious. Per abdomen examination was unremarkable. A CECT of the abdomen was done which showed a multiloculated cystic lesion with peripherally enhancing wall in the subhepatic and perigastric region of 46×97×78 mm, possibly an EDC. An exploratory laparotomy was carried out, and a gastric duplication cyst at the greater curvature of the stomach with another smaller cyst at the first part of the duodenum were found. Deroofing of the enteric cysts with mucosal stripping was done, keeping the native duodenum and stomach intact. The child recovered well in the post-operative period and was discharged on full feeds on the 7th POD (Figure 3).

CASE 7

A 7-month-old boy was admitted with complaints of a mass per abdomen since birth and recently had complaints of abdominal distension and respiratory distress in the past 2 days. An ultrasound of the abdomen was done which showed a possible duplication cyst with echoes around the hepatic flexure or ascending colon. An emergency exploratory laparotomy was done, which revealed a jejunal duplication cyst with a perforation in the first part of the duodenum. The duodenal perforation was repaired primarily and the jejunal duplication cyst was resected. A Roux-en-Y gastrojejunostomy was also performed. The child recovered well in the post-operative period and was discharged on POD7.

CASE 8

A 1½-year-old boy was admitted with on-and-off abdominal pain and bilious vomiting for 6 months. On examination

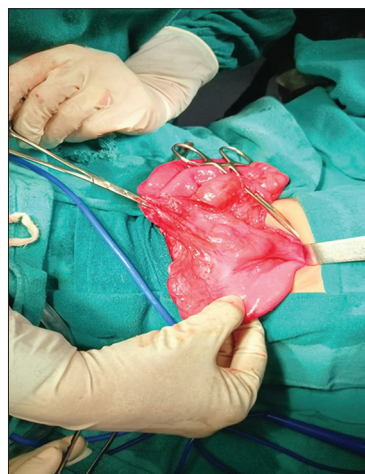


Figure 3: Gastric duplication cyst at the greater curvature of the stomach

of the abdomen, it was found to be unremarkable. An initial ultrasound of the abdomen was done which showed a transient whirlpooling of the mesenteric vessels with the jejunal loops on the right side and a cystic lesion of 24×26 mm in the right flank region. A CECT abdomen was then performed, which showed a well-defined non-enhancing mass of 24×20×29 mm noted in the right lumbar region adjacent to the ileal loops with twisting of the mesenteric vessels with the superior mesenteric vein anterior to the superior mesenteric artery. An exploratory laparotomy confirmed a malrotation of the gut with a 2 cm×2 cm size ileal duplication cyst; Ladd's procedure was done and a resection and anastomosis of the ileum bearing the cyst were done. He recovered well in the post-operative period and was then discharged on the 7th POD.

CASE 9

A 3-day-old male child was admitted with complaints of abdominal distension and bilious vomiting since birth. On examination, the abdomen was found to be distended. An erect X-ray abdomen showed multiple air fluid levels. An ultrasound of the abdomen done showed a 52×31 mm cystic lesion seen in the right hypochondrium without vascularity, with a differential diagnosis of a mesenteric cyst. An exploratory laparotomy was performed, which showed a large ileal duplication cyst in torsion arising <5 cm from the cecum, causing ileal obstruction. The cyst is excised with the ileum bearing it along with cecum, and an ileo-ascending anastomosis was done. In the post-operative period, the child did not pass stools and developed abdominal distension. On 5th POD, we go for chest X-ray and erect abdominal X-ray which shows free gas under the diaphragm [sign of bowel perforation]. Patients shifted for emergency exploratory laparotomy proximal ileal perforation was identified, and a resection of the perforated segment of the ileum was resected and an ileostomy with a distal mucous fistula was created. The patient then recovered well and was discharged 5 days later.

CASE 10

An 8-month-old male child presented with complaints of abdominal distension, which was on and off since birth and complaints of constipation. An initial ultrasound showed a midline cystic lesion with air fluid levels with no obvious peristalsis, with a possible redundant colon. CECT abdomen showed a single thin-walled cystic lesion of 15 × 23 × 21 mm with air fluid level noted on the prevertebral lesion anterior to T8-T10, right side of esophagus; multiple segmentation anomalies noted in thoracic spine T8-T10; SMV on the left side of SMA. Jejunal loops were present on the right side of abdomen with DJ on right side of

L3; dilated colon loops noted with 6 mm max diameter—probably transverse colon. Descending loops are seen in the right lower abdomen pushed by dilated colon. A differential diagnosis of a neuroenteric cyst was also given. On exploratory laparotomy, a dilated jejunal segment was seen, with a tubular structure connecting it and entering into the thorax on the right side. A right thoracotomy was also done and showed a foregut duplication cyst, which was excised in toto. The jejunal segment that was communicating with the foregut duplication cyst was also excised and a jejuno-ileal anastomosis was performed. The patient recovered well in the post-operative period, with ICD removed on POD 4, and the patient was discharged on POD 10.

CASE 11

A 3-year-old boy came with complaints of abdominal pain for 4 days and associated fever. Per abdominal examination was unremarkable. An ultrasound abdomen was done, which showed partial small bowel intussusception with lead point as cyst with gut signature of size 25×26 mm at the ileum. Intraoperatively, a cecal duplication cyst was found, and resection of the distal ileum with cecum with an ileo-ascending anastomosis was done. The patient recovered well in the post-operative period and was discharged on the 7th POD.

CASE 12

A 10-year-old boy came with complaints of vague abdominal pain, which was intermittent. On examination, a lump was palpable in the right iliac fossa. Ultrasound of the abdomen demonstrated a 43×23 mm cystic area adjacent to ileum and cecum. Intraoperatively, a cecal duplication cyst 5 cm from ileocecal junction was found. The distal ileum along with the cecum was resected, and an ileo-ascending anastomosis was done. He recovered well in the post-operative period and was discharged on the 7th POD (Figure 4).



Figure 4: Cecal duplication cyst

CASE 13

A 2-year-old male child came with complaints of pain abdomen and vomiting for 7 days. Per abdominal examination is unremarkable, and an initial ultrasound was done. It demonstrated a cystic lesion and reversal of SMA and SMV. A CECT abdomen was done, which showed whirlpooling of midgut vessels with reversal of SMA and SMV and a well-defined cystic lesion of 37×59×61 mm is noted in midline at L2 to L4 vertebrae. An exploratory laparotomy was done, which showed a jejunal duplication cyst of 10 cm, and a resection of the jejunum bearing the cyst was done with a jejunoileal anastomosis. The patient recovered well in the post-operative period and was discharged on POD 7.

CASE 14

A 3-year-old boy was admitted with complaints of chronic constipation, which was not relieving with laxatives. A barium enema was done which showed a loaded rectum, a redundant sigmoid colon, and a loaded descending colon. No any narrowing of colon was found on barium enema. A 24-h delayed X-ray showed retention of barium in rectum, sigmoid colon, and descending colon. Suspecting it to be a short segment Hirschsprung's disease, a laparotomy was done, which showed a jejunal duplication cyst 10 cm long, 20 cm from duodeno-jejunal flexure with malrotation. A Ladd's procedure was done, with resection of the jejunum bearing the cyst, and a jejuno-jejunal anastomosis was done. The patient recovered well in the post-operative period and was discharged on POD 7.

Special case

A 11-day-old male child was brought with complaints of an extra limb present on the back. The child was evaluated at another center, where an ultrasound of the abdomen was done, and showed a large cyst with gut signature with no obvious communication. A CECT was also done at another center which showed features of open spinal dysraphism in the dorsal spine with diastematomyelia and lipomyelomeningocele, with deformed upper-limb structures forming a joint with the widened spinal canal, with a bronchogenic cyst in the right chest, and large cystic lesion in the right hemiabdomen. An exploratory laparotomy was performed which revealed a large mesenteric cyst at the terminal ileum (a scenario that goes against the incomplete twinning theory).

DISCUSSION

Alimentary canal duplication cysts must have three main characteristics:²

1. An epithelial lining containing the mucosa of the alimentary tract
2. An envelope of smooth muscle
3. The cyst must be closely attached to the gastrointestinal tract by sharing a common wall.

On reviewing the existing literature about EDCs, it was noticed that many case reports and few case series were present. The case series that were present were usually a small group of cases or a moderate number that were collected over decades.³

The term EDCs were popularized by Ladd in the 1930's but were first used by Fitz before that.⁴ Duplication cysts, while they may also be asymptomatic and present late into adulthood, we found that they can present with complications during the neonatal period and in infancy. In children older than 3 years of age, they presented with vague symptoms.

We also found EDCs to be incidentally detected in patients with other congenital anomalies, such as congenital diaphragmatic hernia and malrotation.

The patients that were eventually diagnosed with EDCs presented in a wide age range, beginning from 3 days of age to 10 years of age, with an average age of 2.2 years. The male-to-female incidence ratio was 3.6:1, with a male preponderance (11 males-3 females).

The regional incidence was marked on a map of the state, with 3 patients hailing from the city of Jaipur. The incidence of ileal duplication cysts was the highest (35.7%) (Figure 5).

The most common presenting symptoms for children older than 2 years was found to be vague abdominal pain and

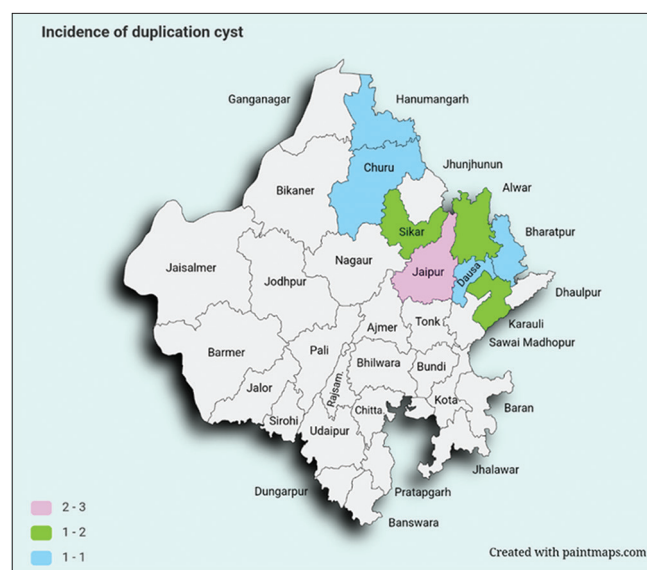


Figure 5: Incidence of duplication cysts in state Rajasthan

recurrent vomiting, while children <1 year of age presented with more severe symptoms. Three patients presented with complications such as perforation of bowel and intestinal obstruction. Two patients had associated malrotation of the gut, and one child had a congenital diaphragmatic hernia.

Most patients underwent resection and anastomosis of the bowel containing the duplication cyst; however, two patients underwent mucosal stripping – one for a jejunoileal duplication cyst and one for a gastric and duodenal duplication cyst.

Postoperatively, one child had a prolonged ICU stay due to right lung consolidation (CDH with foregut duplication cyst) and one child had a postoperative anastomotic leak and required re exploration. All patients were discharged, and there were no fatalities.

A case report of a 15 year old⁵ found to have a posterior mediastinal cyst and a large hiatal hernia was managed thoroscopically for the mediastinal cyst, which was found to be in the left chest cavity, with a type A Bochdalek hernia that was missed on the initial thoracoscopy. In our study, we had a child who presented with respiratory distress and was found to have a large left diaphragmatic hernia, with right lung consolidation and a foregut duplication cyst. As the need was to resolve the respiratory complaints, the diaphragmatic hernia was tackled first, with the foregut duplication cyst left *in situ*, to plan for excision at a later date.

Bui *et al.*,⁶ reported a 2-year-old boy with non-bilious vomiting and abdominal pain, with fusion and segmentation anomalies of the spine, from C5 to T4, along with 4 absent ribs on the left side. In addition, on the CT of the chest and abdomen, a possible anterolateral thoracic myelocoele was also seen, with a separate cystic structure also visualized. The cystic lesion extended into the right lower quadrant of the abdomen. Intraoperatively, an abnormal bowel-like cystic structure within the jejunal mesentery extending cephalad was present, crossing the SMA and coeliac axis. Malrotation of the intestine with the entire bowel on the right side was seen, with appendix and cecum on the left upper quadrant. The malrotation was corrected, and the cyst was separated from the adjacent bowel wall. On entering the thorax, the extension of the cyst was seen in the posterior mediastinum and was seen separate from an anterior myelocoele that was at the apex of the left chest. The cystic mass was resected, with repair of the myelocoele done as well as the repair of the diaphragmatic hernia.

In our study, a similar patient presented with abdominal distention and constipation. CECT thorax and abdomen was done, which showed a cystic lesion in the mediastinum with hemivertebral anomalies, suspecting a neuroenteric cyst. This child intraoperatively had a mediastinal

duplication cyst that extended into the abdominal cavity and ended in a dilated jejunal segment.

A case report by Pai *et al.*,⁷ demonstrated a rare case of duplication cyst with midgut volvulus, seen in a 4-day-old child. In our series, we had two such patients, who presented after 1 year of age with malrotation. One child was found to have a jejunal duplication cyst, and the other one had an ileal duplication cyst. A Ladd's procedure with appendectomy was done for both our patients, along with resection and anastomosis of the bowel containing the cyst. This was a similar management to that reported by Azzam *et al.*⁸

At our center, we encountered two patients with cecal duplication, one of whom presented with intussusception and the other with a palpable lump in the right iliac fossa. Cecal duplication cysts are a rarer entity of EDCs, accounting for 0.4% of all duplications.⁹

Patients could be misdiagnosed with an appendicular lump, appendicular abscess, or even necrotizing enterocolitis. Temiz *et al.*,⁹ had done cyst excision with the cecum in 6 of the seven patients, along with an ileo-colostomy, similar to what was performed at our center for the two cecal duplication cysts.

CONCLUSION

EDCs are a rare gastrointestinal anomaly that requires a high index of suspicion when it comes to a pre-operative diagnosis. It can present as benign vague abdominal pain that is intermittent or can present along with bowel perforation, leading to sepsis and shock. They can mimic mesenteric cysts or omental cysts based on their size and location. They can also be associated with other anomalies such as malrotation of gut and congenital diaphragmatic hernia. While the combination of EDC with other anomalies is extremely rare, it does prompt further enquiries and studies related to the causality of both occurring in the same patient.

This study has demonstrated a relatively large volume of patients who were diagnosed with EDC during a 2-year period, which also prompts more questions about the association of this rare entity and the environmental demography as well. Further studies, classified by region, can aid in determining the association between other anomalies and EDCs.

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AKK- Definition of intellectual content, prepared the first draft of the manuscript; **GR**- Literature research identification, data analysis; **AKC**- Manuscript preparation; **PG**- Editing and manuscript revision; **AP**- Submission of article and manuscript revision; **AJB**- Editing; **KS**- Data analysis; **VKS**- Funding support

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