# **Intestinal Obstruction in Pediatrics: Case Series of Unusual Causes**

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Abstract Introduction: Congenital anomalies are main cause of intestinal obstruction that occur from neonates to adults. Frequently, obstruction is due to either incarcerated hernia, adhesive bands, volvulus and intussusception. we illustrate all demographic & clinical data, imaging, surgical approach and outcome of unusual causes of pediatric intestinal obstruction.

**Materials and Methods:** We document a retrospective review of ten pediatric cases of acute intestinal obstruction for whom surgery was indicated and showed rare causes and pathology between 2020 and 2023.

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**Results:** On exploration, first and second patient had non-Hodgkin intestinal lymphoma. Third case had a large polyp of Peutz– Jeghers syndrome as a nidus for jejunojejunal intussusception. Fourth patient had jeujunogastric intussusception whereas fifth case had chylous cyst, in the sixth patient, cecal duplication cyst was a cause of intussusception. Seventh case had volvulus of small gut due to entrapment through a mesenteric defect was detected whereas eighth case had ileal gel ball, ninth case had volvulus on top of mesenteric lymphangioma and tenth case had huge mesenteric chylous cyst. All causes of obstruction are rare either as a pathology or its age or presentation.

# **Keywords**

- Pediatrics
- intestinal
- obstruction
- unusual causes

**Conclusion:** Suspicion, careful evaluation, and tailoring of appropriate treatment are a corner stone for the precise management of these unusual cases.

## Introduction

Intestinal obstruction is common in pediatrics who presents with recurrent episodes of vomiting, abdominal distention and pain.<sup>1</sup> The detected causes may be acquired or congenital from the neonatal to adult period.<sup>2</sup> The definite cause of obstruction may not be easily recognized, despite of the advanced diagnostic capabilities, we should not delay the surgery to avoid a catastrophe.<sup>3</sup>

In older children versus neonates, the symptoms are not specific; diagnosis of the bowel obstruction requires imaging for precise detection of its origin, site, and extension. Consequently, abdominal x ray is the initial workup for their assessment but it is usually followed by another one for instance; contrast of upper gastrointestinal tract (GIT) or enema, ultrasound, CT, or MRI.<sup>4</sup>

## **Materials and Methods**

We illustrate 10 rare causes of pediatric gut obstruction between January 2020 and January 2023 at our institute. All patients' data were retrospectively reviewed and contemporaneously collected. Demographic, operative, and follow-up data were thoroughly summarized. Written informed consent which was accepted from local Institutional Review Board of ethical committee affiliated to faculty of medicine, was obtained from parents for both operation and proposed publications.

#### Result

All demographic, clinical, management steps, postoperative findings are illustrated in **Table 1.** Among our patients, history

was significant in case 3 in form of her father who had pancreatic carcinoma while case 4 underwent surgery for gastric outlet obstruction in form of gastrojejunostomy. A palpable abdominal mass was detected in cases 3,6,7, 9 and 10, and peritonitis was detected in case 3 and 8 whereas the associated pathology and comorbidities showed adenoid lymphoma in cases 1. Imaging showed multiple air fluid levels in all cases except the last case but other patients needed further imaging. All postoperative course was uneventful except for surgical wound infection in case 8 which was managed by dressing. Postoperatively, follow up mandated medical oncology for case 1 & 2, annual colonoscopy for case 3 which showed a benign polyp for which excision was done one year postoperatively.

Item	Case 1	Case 2	Case 3
Sex	Male	Male	Female
Age(years)	7	8	12
C/O	2 days of abdominal pain &bilious emesis	3 days of abdominal pain &bilious emesis	3 days of abdominal pain bilious emesis
Fever	Yes	no	no
G. Examination	Signs of sepsis	dehydrated	facial pigmentation of lip mucocutaneou s junction Figures 3 A
Lab. Results	WBCs=40600	WBCs=35000	HB=9 gram
Ultrasound	Bowel dilatation, minimal free fluid, intussusception	Bowel dilatation, minimal free fluid, Multiple mesenteric lymph nodes	Bowel dilatation
CT Abdomen	No	No	Jejunojejunal intussusceptio n as in Figures 3 B, C
Operative Pathology	Jeujenojeujenal intussusception 60 cm from DJ & gangrene of the invaginated loop which contained polyp, a palpable intaluminal midileal polyp with serosal infiltration 50 cm from the proximal pathology, another palpable ileal polyp just proximal to ileacecal valve and multiple lymph	Amalgamated ileal loop with mural infiltration and luminal obstruction in addition to amalgamated enlarged lymph nodes <b>figures 2 A, B, C</b>	Jejunojejunal intussusceptio n after reduction of the mass; we found a polypoid mass obstructing the lumen <b>figure 3 D</b>

 Table 1: All collected data of illustrated cases:

	nodes adjacent to the pathological segments as		
Treatment	Laparotomy, resection & anastomosis	Laparotomy, resection & anastomosis	Laparotomy, resection & anastomosis
Size of pathological lesion	5×4×3 cm	12×6×3 cm	5 cm
Cause of obstruction	Secondary intussusception due to Intestinal Burkitt's lymphoma	Intestinal Burkitt's lymphoma	Secondary intussusceptio n due to hamartomatou s polyp
Histopatholog y	non-Hodgkin of one lymph node with positive immunophenotyping for CD20, BCL-6 and TdT	non-Hodgkin of one lymph node with positive immunophenotyping for CD20, BCL-6 and TdT	a hamartomatou s polyp compatible with Peutz– Jeghers syndrome
Item	Case 4	Case 5	Case 6
Sex	Male	Male	Female
Age(years)	16 years	11 months	6 years
C/O	2 days of abd pain, bilious emesis followed by three episodes of hematemesis	a two-day history of diffuse distention & bilious vomiting	a two-day history of bilious vomiting, distension & absolute constipation
Fever	no	no	no
G. Examination	Hypotensive, pale	stable	dehydrated with abdominal distension,

Lab. Results	HB= 8 g	normal	normal
Ultrasound		a mobile well-defined cystic lesion (6×5×4 cm) of the gut filling the right iliac fossa and lumbar region	Target sign with Cystic lesion about 5×5 cm In right iliac fossa
CT Abdomen	dilated stomach with herniated jeujonal loop through widened gastrojejunostomy ostium making mass like lesion with whorly appearance (intussusception) with preserved postcontrast enhancement, notable dilatation of duodenum and jeujenum proximal to the intussusception so the picture was suggestive of retrograde jeujonogastric intussusception ( <b>Figure 4</b> <b>A</b> , <b>B</b> )	no	no
Operative Pathology	retrograde jeujonogastric intussusception	a unilocular chylous cyst involving the ileal mesentery Figure 5	intraluminal cecal cystic mass about 5×5 cm obstructing ileocecal junction Figure 6 A, B
Treatment	Laparotomy manually reduced and revision to roux EN Y gastrojejunostomy (retro gastric, retro colic) was done fixing efferent loop to the mesocolon as <b>in</b> <b>Figure 4 C, D, E.</b>	Segmental intestinal resection of the involved loop	ileocecal resection and primary anastomosis

Size of pathological lesion Cause of obstruction	Jejunogastric intussusception	L	Comp	$7 \times 6 \times 5$ cm	5×5 cm Cecal duplication compression
Histopatholog y	Mucosal ulceration of the resected jejunum		unilocular cyst lined with endothelium and filled with chylous fluid and lymph confirming of chylolymphatic cyst		cecum containing cyst 4×4 cm with serous fluid content and smooth thin wall
Item	Case 7	Cas	se 8	Case 9	Case 10
Sex	Female	Ma	ale	Female	Female
Age(years)	7 years	4 ye	ears	8 years	10 years
C/O	a one- day history of abdominal pain & bilious vomiting	a th day h o abdor pa bili vomi dister & abs const	ree- istory of minal in, ous iting, nsion solute ipatio	a three- day history of agonizing abdominal pain, bilious vomiting, distension & absolute constipation	A one-month history of abdominal pain, distension, discomfort with feeding exacerbated in the last 2 days with persistent bilious vomiting and constipation.
Fever	Yes, 38 C	Yes,	38 C	no	no
G. Examination	dehydrated with abdominal distension, tachycardia	Dehyo wi abdor dister	drated ith minal nsion	Dehydrated with epigastric distension, guard, tenderness in upper and midabdominal region	Dehydrated with epigastric distension and palpable midabdominal mass (Figure 10 A)

Lab. Results	WBCs =15000	WBCs =16000	Low K	Normal
Ultrasound	Dilatation of the small bowel	Dilatation of the small bowel with suspected intralumina l cystic lesion	Dilatation of bowel suggestive of obstruction	Large cystic lesion 10×9 cm in center of abdomen with dilatation of small bowel
CT Abdomen	no	no	Small bowel obstruction, coiling of jejunoileal junction with transition zone midabdominal picture of volvulus, large pelvic cystic lesion mostly mesenteric lymphangioma 10×9 cm (Figure 9 A)	Large cystic lesion 15×6 cm in center of abdomen with dilatation of small bowel due to displacement of the gut suggestive of mesenteric cyst.
Operative Pathology	obstruction of the small bowel due to a large mesenteric defect through which twisted ischemic small bowel pass Figure 7 A, B, C	Ileal intralumina l mass with mural necrosis showed jelly ball with opening the lumen <b>Figure 8</b> <b>A, B, C</b>	Laparoscopy was done revealing large mesenteric cystic lesion causing abnormal twisting of the bowel loops causing lumenal obstruction without vascular insufficiency (Figure 9 B, C, D)	Large mesenteric chylous cyst related to small bowel (Figure 10 B)
Treatment	Resection and anastomosis of the incarcerated bowel with repair of the defect	Segmental intestinal resection of the	Resection & anastomosis was done after aspiration of the cyst &	Excision with preservation mesenteric blood vessels and gut

#### **Pediatric Intestinal Obstruction**

		involved loop	exteriorization of the affected loop through the umbilical port (Figure 9 E, F, G)	(Figure10 C, D, E)
Size of pathological lesion		5×5 cm	10×9 cm	15×6 cm
Cause of obstruction	Internal hernia through mesenteric defect	Jelly ball	large mesenteric cystic lesion causing abnormal twisting of the bowel loops causing lumenal obstruction	15×6 cm chylous cyst compressing small bowel
Histopatholog y	Resected bowel is necrotic	Resected bowel with necrotic wall	10×9 cm mesenteric lymphangioma attached to small bowel segment jejunum, no malignancy	15×6 cm chylous cyst



Figure 1: A, B Intraoperative findings of case 1.



Figure 2: A, B C Intraoperative findings of case 2



- Figure 3: A. Mucocutaneous junction pigmentation
- B. CT axial section of intussusception
- C. CT coronal section of intussusception
- **D.** Specimen of jeujunal polyp

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Figure 4: A. CT axial section of jejeunogastric intussusception

- B. CT coronal section of jejeunogastric intussusception
- C. Operative finding of jejeunogastric intussusception
- D. Operative photo after reduction and division of loop gastrojejeunostomy.
- E. Operative photo of Roux en Y gastrojejeunostomy after revision.



## **Figure 5: Operative finding**



Figure 6: A, B. Operative findings



Figure 7: A, B, C Operative findings



Figure 8: A, B, C Operative findings



## Figure 9: A CT finding

- B, C, D laparoscopic Operative findings
- E, F, G Operative steps

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**Figure 10: (A)** palpable mass under general anaesthesia B, C, D, E Operative steps of cyst removal

## Discussion

Intestinal obstruction is common in pediatrics which may occur either in acute or chronic form.<sup>5</sup>

#### Gastrointestinal lymphoma

Lymphoma is a neoplasm that can occur either in the nodal or extra nodal lymphoid tissue which is mostly developed in the (GIT); commonly in stomach, ileum or colon. 80% of these tumors involve the ileocecal region because the terminal ileum has Payer's patches.<sup>6</sup> Macroscopically, malignant lymphoma was classified into four types: polypoid, ulcerative. constrictive.<sup>7</sup> aneurismal. and The polypoid type is commonly associated with intussusception, whereas the ulcerative and constrictive types are associated with perforation and ileus respectively. The soft polypoid tumor easily forms intussusception because peristalsis is not impaired as in malignant lymphoma due to little infiltration to the muscle layer. Similarly, case 1 had the polypoidal type which was found in the ileum and jejunum but was detected at a younger age whereas case 2 was of the same age but had both the constrictive and amalgamated nature. Two cases have been reported as gastrointestinal

non-Hodgkin lymphoma in form of ileocolonic intussusception.<sup>8</sup>

#### **Peutz–Jeghers syndrome (PJS)**

Jejunal intussusception is not common as ileocolic type. It is commonly having a leading point, and usually shows weight loss and chronic abdominal pain.<sup>9</sup> PJS is an inherited, autosomal-dominant disorder, characterized by hamartomata's polyps small mostly in the gut, and mucocutaneous pigmented lesions, and is uncommon cause of recurrent jejunal intussusception.<sup>9</sup> Multiple laparotomies are usually required with a possible risk of short-bowel syndrome. In our case, both mucocutaneous pigmentation and palpable abdominal mass were indicators of the preoperative diagnosis. On laparotomy, we jejunojejunal intussusception found secondary to a large polyp as a nidus which was also proved to be hamartomata by histopathology. On annual endoscopic follow up, our patient showed colonic polyp was detected and removed showing same pathology. In literature, PJS was illustrated in form of case series and case reports.<sup>10</sup>

The cumulative intussusception risk in PJS was reported to be 15% by 10 years whereas 50% by 20 years.[11] PJ polyps include small gut  $\geq$  95% of cases

commonly in the jejunum, which may be a nidus for the intussusception especially if it is  $\geq 15$  mm in diameter.<sup>11</sup> Additionally, it was also illustrated that hamartomata's polyps most commonly occur in the small gut (78%), colon (42%), stomach (38%), and rectum (28%).<sup>12</sup>

Laparotomy is recommended as the safest option of intussusception secondary to PJS in form of the manual reduction followed by resection of the polyp at its base. Moreover, the entire formal gut check is mandatory to find and remove  $\geq 15 \text{ mm}$ size polyps. Laparoscopy in combination with endoscopy could be possible.<sup>13</sup> The recent guidelines recommend initial GIT surveillance after 8 years in an asymptomatic individual, and before if symptomatic in form of GIT endoscopy, and MRI studies or video capsule every 3 years. endoscopy Elective polypectomy should be performed for small polyps up to 15-20 mm to prevent intussusception. Lifelong cancer surveillance for adults having PJS is mandatory for early detection of pancreatic, testes, breast, and gynecological tumors by full blood cell count with physical examination every year.14

#### Chylous & lymphatic mesenteric cyst

Chylous cysts involve 7.3–9.5% of all abdominal cystic pathology which are little bit rare mesenteric lesions.<sup>15</sup> It contains chyle which is disequilibrium between its inflow and outflow.<sup>15</sup> Over the literature, few pediatric cases with chylous cysts are reported.<sup>16-17</sup>

As regard its different surgical management possibilities include marsupialization, sclerotherapy, drainage, enucleation, percutaneous aspiration, and excision of the cyst with or without resection of the involved gut.<sup>18</sup> As much as possible, it should be tried to excise the cyst completely due to its high potential recurrence rates.<sup>19</sup> Although enucleation from the mesentery leaves can be done in adults, gut resection is usually required in children.<sup>20</sup> Our cases presented with smallgut volvulus, compression of small gut and their operative findings and postoperative follow up are illustrated in table 1. In literature, there are four pediatric cases of cystic lymphangioma with only two cases of the chylous variety. Moreover, one chylolymphatic cyst out of five cases of cystic lymphangioma diagnosed on histopathology.<sup>15</sup> Additionally, a chylous mesenteric cyst was found the cause of neonatal intestinal obstruction and an isolated of chylolymphatic case a

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mesenteric cyst in a 10-day-old neonate whereas the mesenteric cyst was reported to be as a trigger for caecal volvulus.<sup>17</sup>

## **Enteric duplication**

According to the type and site of the duplication cysts, about 70% of enteric duplication cysts EDCs present with symptoms during the infancy whereas 85% in the second year.<sup>21</sup> Gastric and intestinal duplications have vulnerable symptoms as nausea, vomiting, distention, palpable mass and a recurrent pain as the commonest complaint. It is postulated due to the accumulation of secretions which increases the pressure inside the cyst. Intussusception is another presentation of the cyst acts as a leading nidus; besides, to obstruction due pain, external compression or bleeding are possible of presentation. patterns Bleeding, ulceration and perforation could occur due to ectopic gastric mucosa within the cyst but EDCs can be detected incidentally.<sup>22</sup>

The treatment of EDCs depends on the location, structure and communication. Cyst excision only could be an option, but a resection of the adjacent gut is mandatory in case of a communication. As regard the proposed recurrence or malignancy, it is crucial to respect the cyst completely. In literature, thoracoscopic and laparoscopic cystectomy were reported for only 20 cases.<sup>23</sup>

#### Jeujonogastric intussuception

Among adults, jejunogastric intussusception was reported after gastrojejunostomy either Billroth II gastrectomy or Roux-en-Y gastrojejunostomy.<sup>24</sup>

There are four types: afferent limb, efferent limb, a combination of afferent and efferent. and а rare side-to-side intussusception through а Braun anastomosis.<sup>24</sup> The common type is the efferent limb, about 75% of the adult reported cases. It has been suggested because of increased exposure to acid, short mesentery and or mesocolon, negative pressure in the stomach, a large stoma, jejunal stenosis, a long efferent loop, and gastrostomy tubes.<sup>24</sup>

The most clinical scenario has been reported; acute incarceration of intussuscipien in the stomach, presents with pain, vomiting, hematemesis, a palpable abdominal mass, and occasionally bilious vomiting.<sup>24</sup> Many radiological investigations can detect the pathology, as CT, ultrasound, and contrast studies. The pathognomonic sign on a barium meal study is the striated filling defect in the stomach. Although there is a success of

endoscopic reduction of acute on chronic jejunogastric intussusceptions, these emergent cases require exploration for urgent reduction and excision of non-viable bowel.<sup>25</sup> The ideal surgical procedure is unclear due to rarity of cases and the low recurrence rate. Isolated reduction of the intussusception as well as revision of the anastomosis to а Roux-en-Y gastrojejunostomy, a Billroth I, and/or suturing the reduced limb to the colon or opposite limb mesentery have all been reported. The Roux-En-Y reconstruction recurrence.<sup>24</sup> lowest showed the Jejunogastric intussusception is not common but lethal. The suspicion should be considered in any child that has had gastrojejunostomy. Urgent radiography and intervention to decrease the possibility of non-viable gut in addition to revision to a Roux-en-Y gastrojejunostomy, with or without jejunopexy is the standard procedure. А jeujenogastric intussusception was reported in a ten - year old child having a history of duodenal duplication surgery.<sup>26</sup>

#### **Congenital mesenteric defect**

Hernia through a mesenteric defect is intraperitoneal that has entrapment of an enteric loop within the mesentery without sac.<sup>27</sup> Ming and Luo explained the rapid onset of gangrene in patients with mesenteric hernia by the small defect and absence of limiting sac with long segment of gut having ischemia and gangrene. In case 7, the mesenteric defect was wide circular in shape. In literature review, 12 patients have internal hernias through mesenteric defect but resection of the gangrenous gut was done in seven cases without any mortality.<sup>27</sup>

#### Gel ball

There are 43 cases of ingested gel ball in form of 16 case studies and 2 case series (involving 6 and 21 cases each) but it was firstly reported in 2011.<sup>28</sup> Fourteen of 43 patients (33%) were  $\leq 2$  years old. Forty four percent (19/43 cases) of the reported cases had signs of GIT obstruction for whom laparotomy for retrieval of the foreign body was done. The youngest patient (6 months old) died due to sepsis of gut gangrene.<sup>29</sup> The decision of observation was taken for management of 22/43 cases (51%) patient inside or outside the hospital without complications. The whole bowel irrigation with oral Polyethylene Glycol 3350 with electrolytes was done for one asymptomatic patient and the gel beads were evacuated successfully without complication. One case showed successful mechanical breakage (ultrasound guided)

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of the gel balls in the gut.<sup>30</sup> As regard imaging, 42% of the symptomatic patients underwent an abdominal x-ray part showing intestinal obstruction whereas an ultrasound (US) was done for 37/43 (86%) of the cases showing an intraluminal cystic lesion. Three obstructed cases needed a CT scan illustrating a cystic lesion or a filling defect.<sup>30</sup> Our case presented with intestinal obstruction mandating the surgical

Conclusion

exploration.

Unusual rare causes of intestinal obstruction should also be noted in infants and children away from the common ones. A high degree of suspicion must be taken in diagnosis of these conditions in form of meticulous evaluation to preserve the gut whereas the ideal management should be specified case by case.

List of abbreviations GIT: Gastrointestinal tract CT: Computerized tomography MRI: magnetic resonance imaging PJS: Peutz–Jeghers syndrome CM: centimeter EDCs: enteric duplication cysts US: ultrasound CD20: cluster of differentiation 20
BCL-6: B cell lymphoma -6
TdT: terminal deoxynucleotidyl transferase
WBCs: white blood cells
OMD: Omphalomesentric duct

## **Ethical Consideration**

Informed written consent which was accepted from local Institutional Review Board affiliated to faculty of medicine, Tanta University, obtained from parents for operation in addition to obtaining appropriate institutional ethics committee approval Local Institutional Review Board approval code: 36262/12/22.

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## **Conflict of interests**

There is no conflict of interest

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