Case series of unusual causes intestinal obstruction in infants and children

Hisham A. Almetaher

Introduction **Many of the causes of intestinal obstruction arise from congenital anomalies that can present at any time from infancy to adulthood. Other frequent causes of intestinal obstruction in pediatric population results from complicated external hernias, intussusceptions, volvulus, or bands of adhesions.**

Patients and methods **We report seven unusual cases presented with symptoms and signs of acute intestinal obstruction in infants and children, which necessitated surgical intervention. Radiological investigation showed signs of intestinal obstruction.**

Results **On exploration, rare causes of intestinal obstruction were found. In the first patient, congenital mesenteric defect was the cause of obstruction. The second patient had unusual entrapment of a loop of bowel in the ileocecal recess. In the third patient, non-Hodgkin's lymphoma was the leading point for intussesception. In the fourth patient, a large multiloculated chylolymphatic cyst was found. In the fifth, sixth, and seventh patients, a rare presentation of remnants of ophalomesentric duct was identified. In the eighth patient, a large polyp of Peutz–Jeghers syndrome was found to obstruct the lumen of the jejunum, and it was the nidus for jejunojejunal intussesception. All of the previously mentioned causes of obstruction are rarely observed in children.**

Conclusion **Management of these conditions requires a high degree of suspicion, careful assessment, and awareness while the appropriate treatment needs to be tailored to the individual situation.** ***Ann Pediatr Surg*** **12:50–58 © 2016 Annals of Pediatric Surgery.**

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Keywords: childhood period, intestinal obstruction, unusual presentations

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Introduction

Acute intestinal obstruction is one of the emergencies that pediatric surgeons encounter in daily practice [1]. It should be suspected in any child with persistent vomiting, distention of the abdomen, and abdominal pain [2]. Many of the causes of intestinal obstruction arise from congenital anomalies that can present at any time from infancy to adulthood [3]. Other frequent causes of intestinal obstruction in the pediatric population are complicated external hernias, intussusceptions, volvulus, or bands of adhesions [2,4]. Sometimes, recognition of the underlying cause of obstruction may not be easy, and in spite of the marked increase in diagnostic capabilities it is still difficult to differentiate between simple obstruction, which may resolve with conservative measures, and bowel strangulation, in which delay in surgery can have catastrophic consequences [5,6].

In this work, we report eight unusual cases of intestinal obstruction in infants and children in the period between March 2013 and January 2016 at the Pediatric Surgery Unit, General Surgery Department, Tanta University Hospitals, Egypt. Although the precise cause of obstruction could not be deduced preoperatively in most cases, we performed surgical intervention without delay. The demographics, histories, presenting symptoms and signs, investigations, laparotomy findings, procedures performed, and outcome of the patients are discussed.

Case report

Case 1

A 3-year-old male child was admitted to the Emergency Department with abdominal pain and biliary vomiting since 2 days. Physical examination revealed a temperature of 37.5°C, pulse rate of 100 beats/min, and abdominal distension. Laboratory investigations were normal. Radiograph showed air–fluid levels indicative of intestinal obstruction. The ultrasound scan also showed dilatation of the small bowel and excluded intussusception. The patient underwent laparotomy after initial resuscitation with intravenous fluid. During laparotomy, the patient was seen to have obstruction of the small bowel due to a small mesenteric defect (Fig. 1). Simple reduction of the incarcerated bowel with repair of the defect was performed. The postoperative course was uneventful. The patient was discharged on the fifth postoperative day. Follow-up continued for 3 months, with no recurrent intestinal obstruction.

Case 2

A 4-year-old boy was admitted because of severe abdominal pain, nausea, and bilious vomiting since the previous 2 days. There was no history of abdominal procedures. His temperature was 37.2°C, pulse rate was 90 beats/min, and blood pressure was 100/60 mmHg. Normal routine laboratory investigations were encountered. Physical examination revealed bowels distention and generalized abdominal tenderness. Other than multiple air–fluid levels the abdominal radiograph was normal. These findings were interpreted as mechanical intestinal obstruction. Preoperative laboratory tests were all within normal ranges, except for elevated total leukocyte count. Adequate hydration and laparotomy were performed sequentially. During laparotomy, a short small-bowel loop (15 cm) was seen entrapped in the inferior ileocecal fold, leading to small-bowel obstruction. The intestine was

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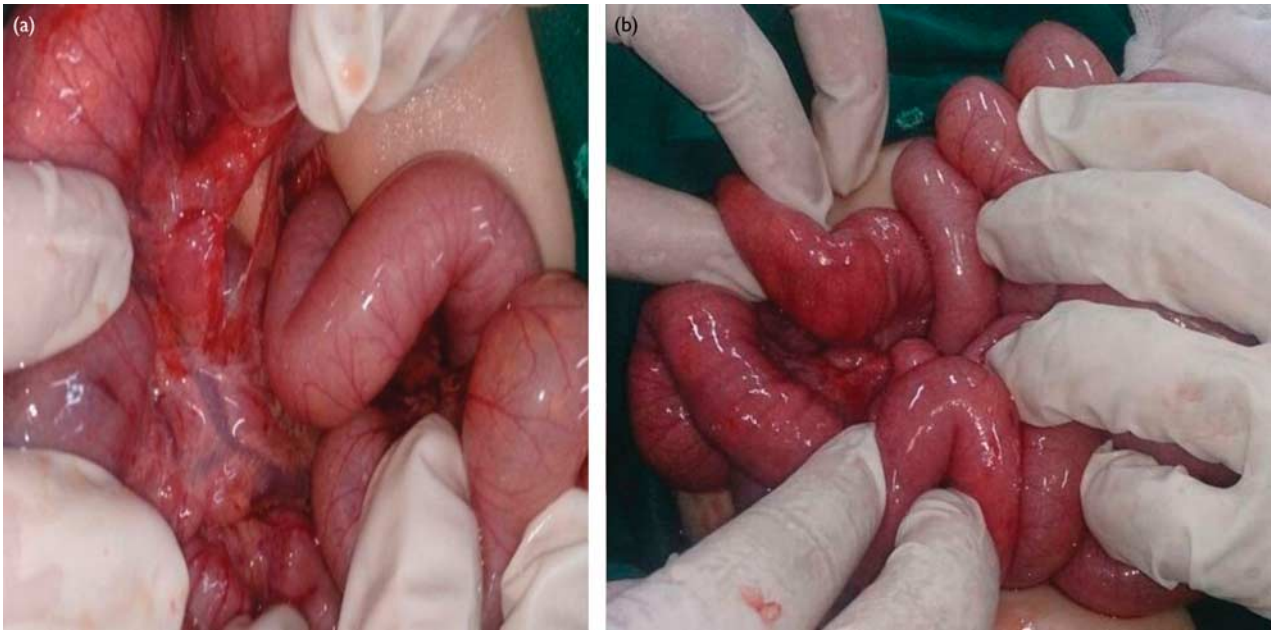
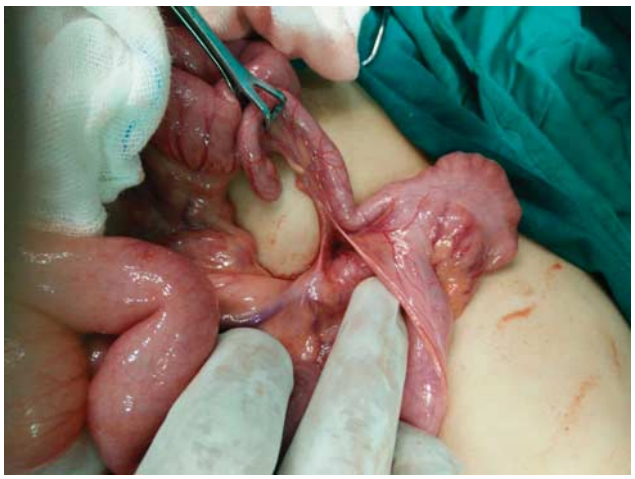
Fig. 1



(a, b) Obstruction of the small bowel through congenital mesenteric defect.

Fig. 2



The ileocecal recess after release of the obstructed bowel.

congested and regained its normal vascularity with hot fomentation; the small-bowel loop was released (Fig. 2) and the redundant peritoneum was resected. The patient had good postoperative recovery and was discharged from the hospital on the fourth day following the operation. The follow-up revealed no recurrence.

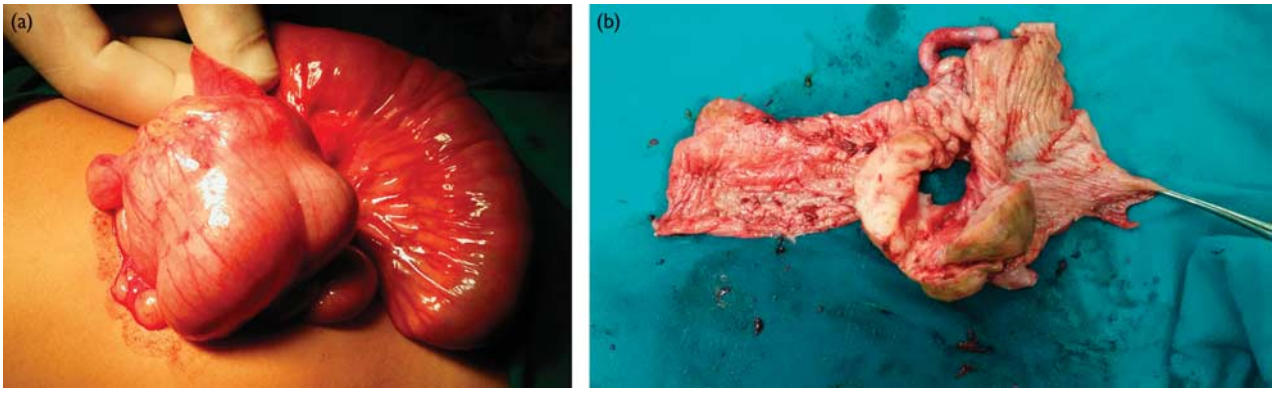
Case 3

A 3-year-old boy came with a history of intermittent progressive abdominal pain since 2 months, which was colicky, distributed throughout the umbilical region, and associated with repeated bilious vomiting. There had been a history of melena since day 1. His pulse rate was 110 beats/min, temperature was 38°C, and blood pressure was 90/60 mmHg. Laboratory investigations revealed hemoglobin 10 mg/dl. On examination, an oval mass was felt in the right hypochondrium having smooth borders and well-defined margins. Ultrasound revealed the typical 'target sign' that confirmed the diagnosis of intussusception. Water-soluble contrast enema was carried out, which revealed that the dye could not pass further than the level of the midtransverse colon, denoting complete obstruction of the colon. Hydrostatic reduction was tried but was not successful. Diagnostic laparoscopy revealed an ileocecal mass. Laporatomy revealed ileocecal intussesception with a palpable mass. Right partial hemicolectomy was performed (Fig. 3). The postoperative period was uneventful. The hemicolectomy specimen was sent for histopathological examination and revealed a partially multisected ileal mass measuring $4 \times 4 \times 3.5$ cm located proximal to the ileocecal valve with fleshy grayish pink and white cut section infiltrating down to the serosal fat and peritoneal covering. Dissection of the pericolic and mesenteric fat revealed seven nodules measuring up to 2×1.4 cm. Microscopic examination revealed diffuse infiltration of malignant lymphocytes with variable appearance from large transformed cells to blasmoblasts and plasma cells to atypical cleaved lymphocytes. The final diagnosis of the histopathological report revealed a large-cell non-Hodgkin's lymphoma of the ileum in addition to immunoproliferative small intestinal disease with free resection margins and reactive mesenteric and pericolic lymph nodes. The patient was started on chemotherapy and is still under treatment.

Case 4

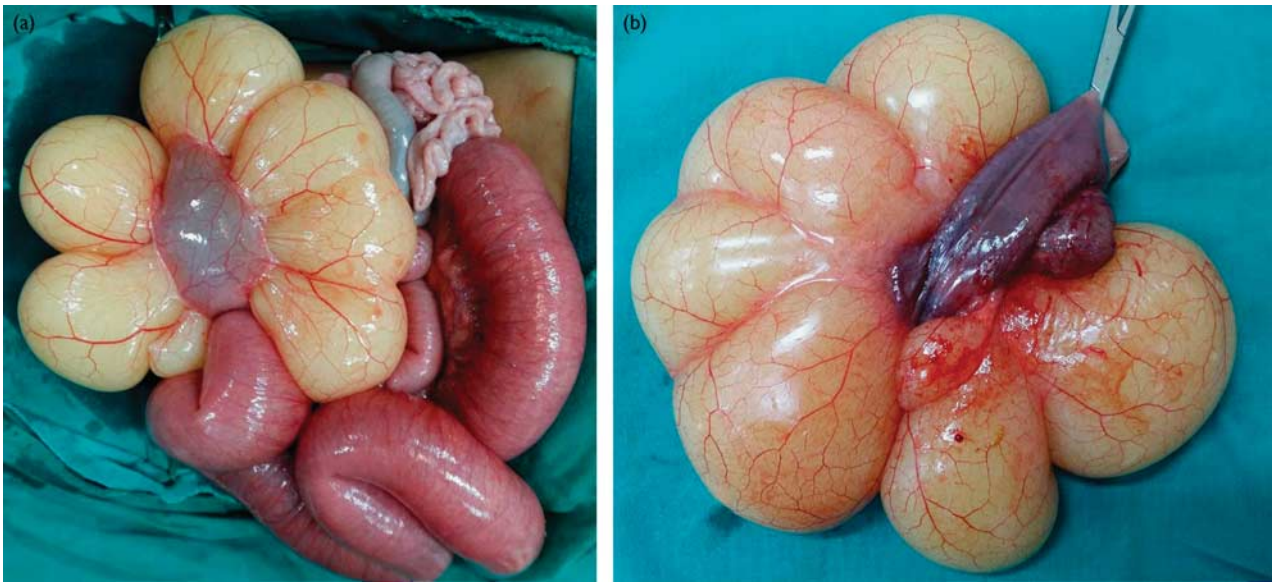
A 3-month-old girl presented with diffuse abdominal distention and bilious vomiting. Her temperature was 37.1°C and heart rate was 100 beats/min. Physical examination revealed severe abdominal distension. No abnormalities were detected in laboratory investigations.

Fig. 3



(a) Ileocecal intussesception. (b) Right hemicolectomy specimen showing multisected ileal mass located proximal to the ileocecal valve.

Fig. 4



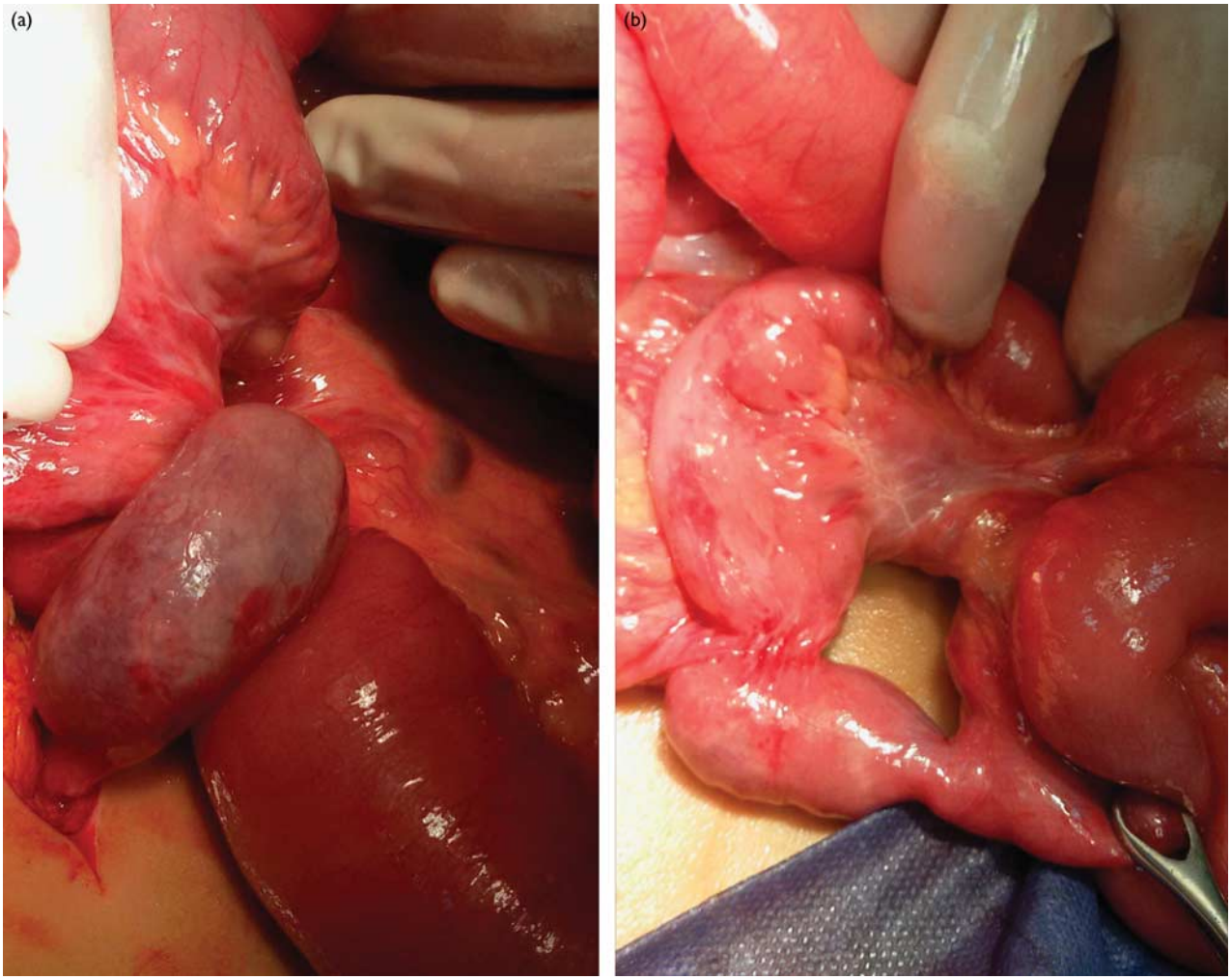
(a, b) Multiloculated chylolymphatic cyst involving the small-bowel mesentery.

Abdominal palpation was tympanic. Bowel sounds were hyperactive. Plain abdominal radiographs showed air–fluid levels and distended bowel loops, suggesting intestinal obstruction. Abdominal ultrasonography showed multiloculated cystic lesions in the gut. Computed tomography (CT) revealed a large well-defined thin wall creeping multilocular cystic lesion filling most of the pelvic cavity and extending superiorly into the right iliac fossa and lumbar region. It contained enhanced septae within and measured about $10 \times 8 \times 15$ cm at transverse section, anteroposterior, and craniocaudal dimensions, respectively. The small-bowel loops were dilated down to the proximal ileal level showing multiple air–fluid levels inside with collapsed ileal loops and colon distally. Laparotomy revealed a multiloculated cyst involving the small-bowel mesentery. The cysts were of varying sizes with the largest ~ 15 cm in diameter with acute stretching of the bowel loop leading to acute intestinal obstruction. Intestinal resection of the involved loops was necessary (Fig. 4). Postoperative recovery was excellent. No recurrence was noted during the follow-up period, which extended up to 1 year. The specimens were sent for histopathological examination, which revealed multiloculated cysts lined with endothelium and filled with chylous fluid and lymph. Thus, the case was confirmed to be chylolymphatic cysts by histopathology.

Case 5

A 7-year-old boy with no previous history of abdominal surgery presented with a history of abdominal pain and vomiting of 2 days' duration. A history of absolute constipation was present for 1 day. There was no history of a similar condition in the past. On examination, his vital signs were within normal levels, but signs of dehydration were present. The abdomen was distended, with muscle guarding and rigidity throughout. Peristaltic sounds were hyperaudible. Erect radiograph of the abdomen showed multiple air–fluid levels. Abdominal ultrasonography revealed hyperperistaltic dilated small-bowel loops with to and fro movements. Laboratory

Fig. 5



(a, b) Ileal loop entrapped by a loop formed by adhesion of the tip of the Meckel's diverticulum to the cecum.

investigation revealed anemia and hypokalemia. Initial resuscitation was done with intravenous fluid and a nasogastric tube that drained the bilious secretion. Exploration was done, which revealed a proximal ileal loop entrapped by a loop formed by adhesion of the tip of the Meckel's diverticulum to the cecum (Fig. 5). Proximal bowel loops were dilated, but fortunately not gangrenous. The small bowel was freed and decompressed. The Meckel's diverticulum was resected and continuity of the bowel was restored with end-to-end anastomosis. The patient made an uneventful recovery after the surgery. He was discharged on postoperative day 7 with no complications in the follow-up period, which extended for 2 months.

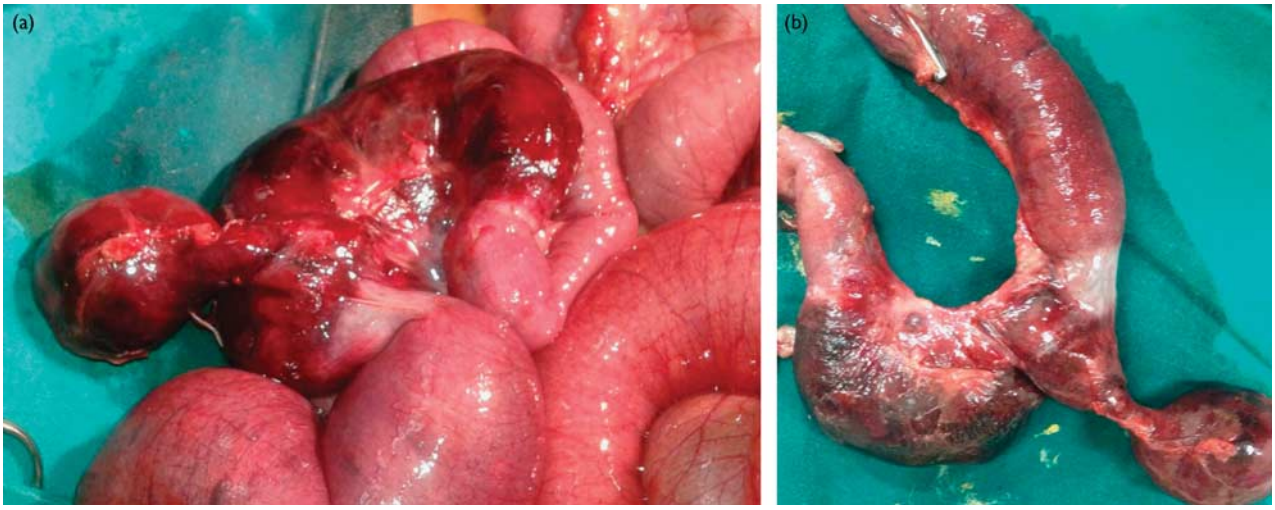
Case 6

A 2-year-old girl presented with abdominal pain and vomiting of 4 days' duration. There was no history of a similar condition or previous abdominal surgery. The patient looked ill, feverish (temperature 38°C), and her blood pressure was 90/50 mmHg and pulse rate was 110 beats/min. Physical examination revealed abdominal distention and increase in bowel sounds. Diffuse air–fluid levels were detected on plain abdominal radiograph. Laboratory examination showed increased total leukocytic count and C-reactive protein. The patient was hospitalized with a diagnosis of intestinal obstruction. She was resuscitated with intravenous fluid and nasogastric suction. Laparotomy was done, which revealed a fibrous cord originating 60–70 cm proximal to the ileocecal valve and ending with a cystic swelling with dimensions of 4 cm diameter and 7–8 cm length. The cyst was rotated, leading to ischemia and gangrene to a loop of ileum. The gangerenous loop with the cyst was resected (Fig. 6). The proximal and distal ends of the bowel were anastomosed with interrupted nonabsorbable sutures. Histopathological examination of extracted tissue was compatible with omphalomesenteric duct (OMD) cyst. The patient was discharged from the hospital on the seventh postoperative day.

Case 7

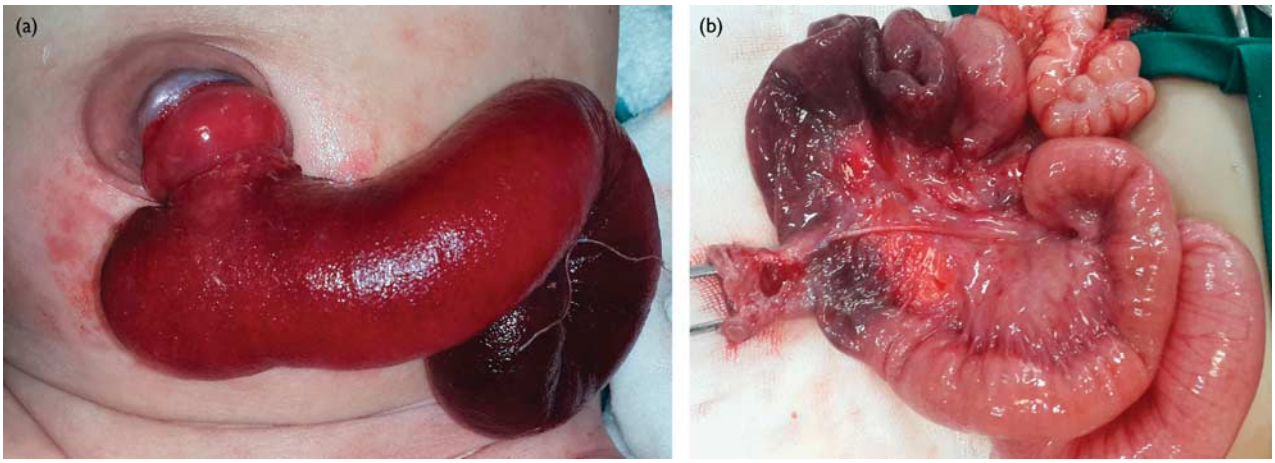
A 33-day-old female infant who had been born at term was brought to the emergency hospital with sudden acute small-bowel evisceration through the umbilicus. Her parents reported a history of umbilical discharge after the umbilical stump fell off, and vomiting 4 h before presentation. Her temp was 38.5°C; heart rate was

Fig. 6



(a, b) Torsion of omphalomesentric cyst with gangrene of small-bowel loop.

Fig. 7



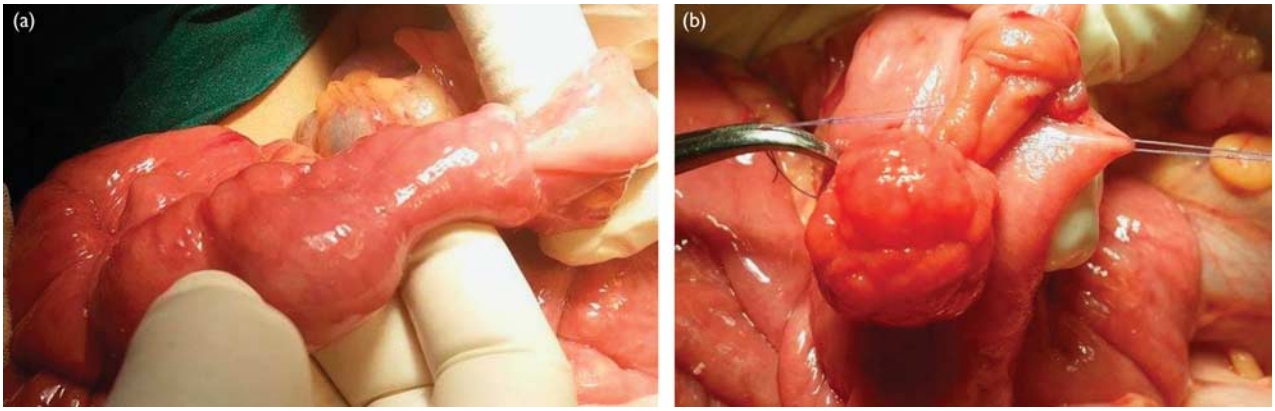
(a) Small-bowel prolapse through the umbilicus giving ram's horn appearance. (b) The patent omphalomesenteric duct completely dissected free from the umbilicus.

110 beats/min. On examination, there was prolapse of the small intestine through the umbilicus, giving a ram's horn appearance (Fig. 7a). Initial resuscitation was carried out with intravenous fluid and this was immediately followed by laparotomy. A supraumbilical incision was made, and the intussucepted small intestine was reduced manually. The duct was released from the umbilicus (Fig. 7b) and the nonviable loop of the small bowel was resected with ileoileal anastomosis (Fig. 7a,b). Primary closure of the abdominal wound and umbilical reconstruction were carried out. The infant was incubated and started feeding 4 days after surgery. The postoperative period was uneventful. She was discharged on the seventh day postoperatively after removing the sutures.

Case 8

An 8-year-old girl was admitted to our hospital with signs and symptoms of acute bowel obstruction. She had been complaining of intermittent severe colicky abdominal pain and bilious vomiting since the last 3 days. His pulse rate was 100 beats/min, temperature was 37°C, and blood pressure was 110/70 mmHg. Physical examination revealed severe abdominal distension and tenderness. Complete blood count revealed anemia (hemoglobin 9 mg/dl). Urine analysis and blood biochemistry were unremarkable. Her plain abdominal radiography was remarkable with sharp air–fluid levels. Ultrasonography of the abdomen revealed distended bowel loops but was not conclusive for intussesception. Subsequently, laparatomy was justified. Jejunojejunal intussesception was found. Palpation of the jejunum revealed a polypoid mass obstructing the lumen. Other small polypoidal masses were felt inside the lumen of the small intestine (Fig. 8a). The intussesception mass was reduced. Enterotomy was done with excision of the large pedunculated polypoidal mass that obstructed the lumen (Fig. 8b). The specimen was sent for histopathological examination. The postoperative course was uneventful. Histopathological

Fig. 8



(a, b) Jejunojejunal intussesception with a large pedunculated polypoidal mass of Peutz–Jeghers syndrome obstructing the lumen.

examination of the specimen revealed a 6×6 cm hamartomatous polyp compatible with Peutz–Jeghers syndrome (PJS). Physical examination could not recognize any mucocutaneous pigmentation; also the family history was unremarkable for PJS. Consequently the patient was referred for further endoscopic evaluation.

Discussion

Intestinal obstruction is a common pediatric surgical problem. It cuts across different age groups in children. Its occurrence in children may be acute or chronic [7,8].

Congenital mesenteric defect

Transmesenteric hernias are intraperitoneal hernias that have no sac and consist of the protrusion of a loop of bowel through a defect in the mesentery [9]. The subject of mesenteric hernias has been largely neglected in textbooks. Federschmidt [10] explained the defect by a partial regression of the dorsal mesentery in humans. Menegaux [11] explained the fenestration by the developmental enlargement of an inadequately vascularized area. Macklin [12] had a different opinion: according to him, a space or defect occurs when two epithelial layers are opposed with a deficient intervening supporting stroma of connective tissue; coalescence inevitably takes place. Ming and Luo [13] explained the rapid onset of gangrene in patients with mesenteric hernia by the small defect and absence of limiting sac with long portions of bowel passing through resulting in ischemia and gangrene. In case 1, the mesenteric defect was oval shaped and separated from the base of the mesentery by a thickened firm edge; this kind of defect from the anatomical point of view is considered congenital in origin. Open or laparoscopic explorations are the only way to establish a clear anatomic diagnosis of this type of internal hernia.

Hernia of the ileocecal recess

In human embryology, rotation of the midgut, followed by migration of the ileocecal portion of the intestine to the right iliac fossa, occurs in the fifth fetal month. Variant recesses and fossae may be formed during fusion and resorption of the peritoneal surfaces following arrival of the cecum in the right iliac fossa. The abdominal recesses may become hernia orifices; four types of peritoneal recesses are found in the pericecal region: superior ileocecal recess, inferior ileocecal recess, retrocecal recess, and paracolic sulci [14]. Waldeyer [15] explained the cause of pericecal hernia as that the lower end of the ascending colon is fixed by adhesion to the retroperitoneum, followed by space formation on the dorsal side of the ileocecal region due to the development and downward movement of the cecum. Nishi *et al.* stated that the cause is a gap formed by faulty union of the ascending colon or cecum with the retroperitoneum [16]. Pielacinski *et al.* [17] reported a case of incarcerated hernia of the inferior ileocecal recess after nephrectomy. Our case appeared to be congenital pericecal herniation of a segment of the ileum trapped in the peritoneal pocket of the inferior ileocecal recess. Although we did not proceed to CT study in our patient, CT has been known to show more precise abdominal anatomy than plain films and contrast studies. CT also can accurately demonstrate the site and cause of intestinal obstruction and show the pathologic processes of the bowel wall, mesentery, and peritoneal cavity [18,19]. Therefore, it is highly recommended to be performed if the patient is suspected to have intestinal obstruction when the clinical and initial radiographic findings remain indeterminate.

Intussusception with lymphoma

Approximately 75% of intussusceptions occur in children under 1 year of age [20]. Idiopathic cases account for more than 90% in children [21]. Malignant lymphoma, a neoplasm of the lymph nodes, can occur in any organ containing lymphoid tissue. It develops more commonly in the gastrointestinal tract, frequently in the stomach, and occasionally in the ileum or colon. About 80% of these tumors in the ileum or colon occur primarily in the ileocecal region, because Payer's patches develop in the terminal ileum [22]. Preoperative diagnosis of the cause of intussesception is difficult [23]. Imaging studies cannot disclose the etiology of intussesception, colonoscopic examination being more useful for this purpose, as a definite diagnosis can be made by the histological examination of biopsy specimens [24]. From 1970 to

1992, 56 cases of malignant lymphomas causing intussesception in adults were reported; the patient ages ranged from 15 to 79 years [25]. Wood classified malignant lymphomas macroscopically into four types: polypoid, ulcerative, aneurismal, and constrictive [26]. Intussesception occurs more commonly in the polypoid, perforation in the ulcerative, and ileus in the constrictive type [27]. The soft polypoid tumor easily forms intussesception because peristalsis is not impaired in malignant lymphoma with little infiltration to the muscle layer [28]. Similarly, our patient had the polypoidal type that was located in the terminal ileum but was reported at a younger age.

Chylolymphatic mesenteric cyst

Chylous cysts are rare variants of mesenteric lesions and constitute 7.3–9.5% of all abdominal cysts [29]. The chylolymphatic cyst, as indicated by its name, contains both chyle and lymph. The accumulation of chyle and lymph is considered to be the result of an imbalance between the inflow and outflow of fluid [29]. There are very few cases of pediatric chylolymphatic cysts reported in the literature [30–34]. The different surgical approaches used are marsupialization, sclerotherapy, drainage, enucleation, percutaneous aspiration, and excision of the cyst with or without resection of the involved gut [35–38]. Because of the high recurrence rates associated with marsupialization and drainage, complete excision of the cyst should be attempted whenever possible [36]. In adults, the cyst can often be enucleated or 'shelled out' from between the leaves of the mesentery; in children, however, bowel resection is frequently required [36,39,40]. Our patient presented with acute intestinal obstruction due to small-bowel volvulus. Exploratory laparotomy and complete excision of the cystic lesion with resection of the involved bowel was done. Multiloculated cysts filled with milky fluid were found. The cysts were of varying sizes, with the largest $\sim$15 cm in diameter. During the follow-up period, we did not observe any recurrences; thus complete excision of the chylolymphatic cyst is curative.

Omphalomesentric duct anomalies

During fetal life, midgut communicates with the yolk sac through the vitellointestinal duct (VID). Between the fifth and ninth week of gestation, communication between the yolk sac and the intestine becomes obliterated. Persistence of a part or all of the OMD results in a variety of abnormalities related to the intestine and abdominal wall [41–43]. Nerdrum [44] recognizes six varieties: patent OMD (umbilicointestinal fistula), Meckel's diverticulum, umbilical sinus, mucous polypus, fibrous band binding the bowel to the umbilicus with no open fistulas, and the omphalomesenteric cyst, which is a persistent lesion of the intermediate duct with closure at both ends, leaving a cyst that can be attached to the umbilicus, bowel, or both. We reported three cases of OMD anomalies (cases 6, 7, 8). Meckel's diverticulum remains the most common OMD anomaly [45]. The overall lifetime risk of development of complications from Meckel's diverticulum is said to be around 4% [46], with one-third of cases resulting in small-bowel obstruction [47]. Obstruction can be attributed to several reasons. The most common is compression of the small bowel due to band, closed loop obstruction, volvulus, and intussusceptions [48]. Loop formation due to adhesions between the distal end of the Meckel's diverticulum and the intestine or the mesentery is an infrequent phenomenon. In case 5, the obstruction was due to adhesion of the tip of Meckel's diverticulum to the ileocecal junction resulting in trapping of the ileal loops in it with loop formation. The literature shows that there are only very few case reports with a similar pattern of obstruction [49].

Intestinal obstruction secondary to OMD cyst is a rare cause of small-bowel obstruction, with very few cases reported in the literature [50–52]. The mortality in patients with these symptomatic cases was 17%, which was mostly related to the cases of intestinal obstruction. Similarly in case 6, the patient was presented late with definite clinical picture of obstruction. Immediate laparotomy was done, which revealed twisting of the cyst leading to intestinal obstruction and gangrene of a small loop of the ileum; fortunately, the patient had excellent recovery.

Nearly 20% of patent OMD cases are complicated by intussusception of the small bowel through the patent duct, leading to intestinal obstruction [52]. Another significant complication is progressive prolapse of the OMD, leading to a T-shaped bowel protrusion through the umbilicus and even ram's horn-type appearance [52]. A patent vitelline duct with intussusception of the small bowel is a rarely reported entity in the world [53]. Two reasons have been hypothesized to explain the mechanism of ileal intussusception into the patent VID; wide mouth of patent VID and shorter distance between the VID and ileocecal valve in infants leading to higher intraluminal pressure [54,55]. The principle of surgical management is reduction of the intussuscepted gut along with complete excision of the vitelline duct and restoring the ileal continuity as well as umbilical reconstruction [52,56]. Three approaches have been described: infraumbilical, supraumbilical, or through the umbilicus [54]. In case 7, the patient presented after the neonatal period with sudden prolapse of the OMD through the umbilicus leading to ram's horn appearance. We have used the supraumbilical approach for reduction of intussesception with resection anastmosis of the ileal loop. The patient showed excellent recovery.

Peutz–Jeghers syndrome

Ileocolic intussusception is one of the most common causes of intestinal obstruction in children aged 3–18 months. On the other hand, jejunal intussusception is an uncommon form of the disease. It has a tendency to occur in older children, has a more chronic course, and often has a demonstrable lesion as a lead point for the intussusceptions, and often presents with chronic abdominal pain and weight loss [57].

PJS is an inherited, autosomal-dominant disorder with variable inheritance, characterized by hamartomatous polyps in the gastrointestinal tract, mostly in the small bowel, and pigmented mucocutaneous lesions, and is one

of the uncommon causes of recurrent jejunal intussusceptions [57]. Those patients require multiple emergency laparotomies but preservation of the intestinal length is important to prevent short-bowel syndrome [58]. Consequently, minimally invasive approaches and double balloon enteroscopy are recommended to destroy the smaller polyps to achieve a polyp-free intestinal mucosa and reduce the need for further surgery [59]. In our case, we could not predict the diagnosis preoperatively because of the absence of mucocutaneous pigmentation and vague family history. On laparotomy, we found a large polyp obstructing the lumen. This polyp was the nidus for intussesception, which proved to be a hamartomatous polyp of PJS by histopathology.

Conclusion

There are common causes of intestinal obstruction in infants and children but the unusual causes described here should also be noted. Management of these conditions requires a high degree of suspicion, careful assessment, and awareness while the appropriate treatment needs to be tailored to the individual situation. Great caution should be paid; however, the incidence of bowel strangulation is quite high.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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