

INTESTINAL INFARCTION THROUGH SECONDARY VOLVULUS – TWO CASES FROM PEDIATRIC SURGERY CLINIC CLUJ-NAPOCA

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Abstract

This article presents two case reports of intestinal infarction which were successfully managed in the Department of Pediatric Surgery, Emergency Hospital for Children Cluj-Napoca. In the first case a volvulus secondary to a congenital malrotation in a 15 days old neonate is described. The second case is focused on a segmental volvulus caused by adhesions in a 16 years old female patient.

Key words: malrotation, volvulus, intestinal infarction

Introduction

Intestinal ischemia consists of the interruption of the blood flow in the irrigation area of the superior or inferior mesenteric artery, that results in intestinal infarction – the hemorrhagic necrosis of the intestines [1]. Intestinal ischemia is a rare condition in neonatology and paediatrics.

Volvulus is a special form of mechanical intestinal and vascular obstruction which results from abnormal twisting of a loop of bowel around the axis of its own mesentery in malrotation [2]. Mesenteric rotation causes vascular insufficiency, and ischemia; infarction occurs in approximately 50% of cases [3]. Surgical intervention is necessary to avoid intestinal infarction necrosis of the bowel [4].

Volvulus can be primary, without any predisposing anatomic abnormalities and risk factors, or secondary, caused by anatomical anomalies (midgut malrotation, congenital fibrous bands [3,5]) or acquired lesions (postsurgical adhesions) [6-7].

Cases presentation

Case 1. Malrotation with volvulus

B.M. a two days old neonate was hospitalised in the Pediatric Department for repeated nonbilious, nonfeeding

related vomiting. At this stage a clinical diagnosis of maternofetal infection was proposed, but the antibiotic treatment with Ampicilinum didn't lead to a favorable evolution. The patient kept on vomiting accompanied by mucosanguineous stools, so he was transferred to the Pediatric Surgery Department for further investigation and treatment.

At admission in the Surgical Department the clinical examination was suggestive for dehydration (dry skin and mucosa) with decreased blood pressure (70/40 mmHg), heart rate (40 beats/ minute) associated with distended abdomen and oliguria. Laboratory investigations revealed metabolic alkalosis, hypochloremia and hyponatremia. The radiological examination with contrast agent showed the opacification of the stomach until the distal part of the duodenum without a further passage (Fig. 1). The abdominal ultrasound showed a distended, fluid-filled jejunum with diminished or absent peristaltic.

The patient was rehydrated and was given a large spectrum antibiotic treatment with Meropenem and Teicoplanin.

After the initial fluid and electrolyte resuscitation the patient was taken to the operating room where jejunal atresia, a 360° intestinal volvulus on a common mesentery and secondary intestinal perforation with generalized peritonitis were discovered. The intestinal derotation, segmental resection of the necrotized jejuno-ileum part which included the atretic portion of 30 cm (Fig. 2) were performed followed by a lateral jejunostoma with the resection of adherences, the lavage and drainage of the peritoneal cavity. At a microscopic level the resection piece presented necrotic areas which involved all the width of the intestinal wall (Fig. 3).

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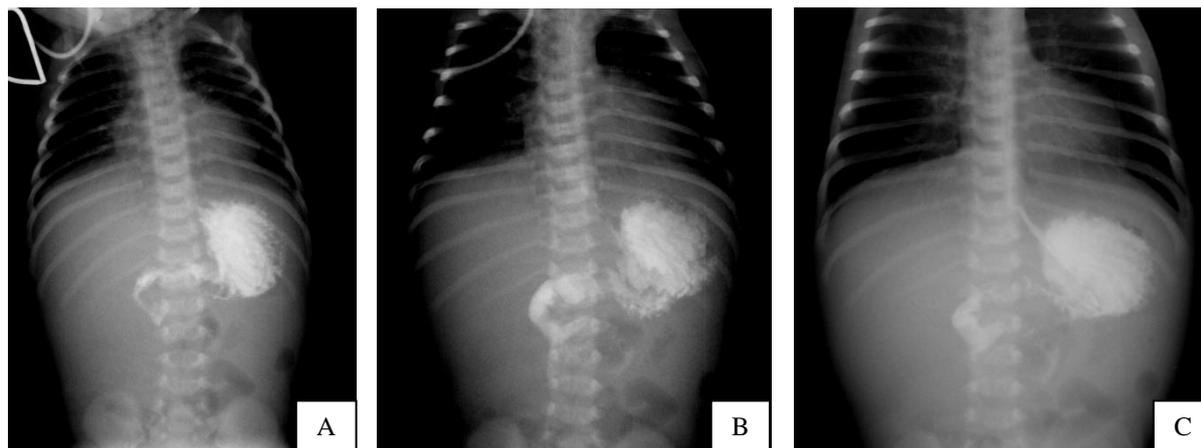


Fig. 1 Radiological examination with contrast agent: postduodenal digestive obstruction; A –30 seconds capture; B –10 minutes capture ; C – 20 minutes capture.



Fig. 2 Macroscopic aspect of the resection piece (at 24h after formal fixation): 3 jejunum-ileum segments of resection.

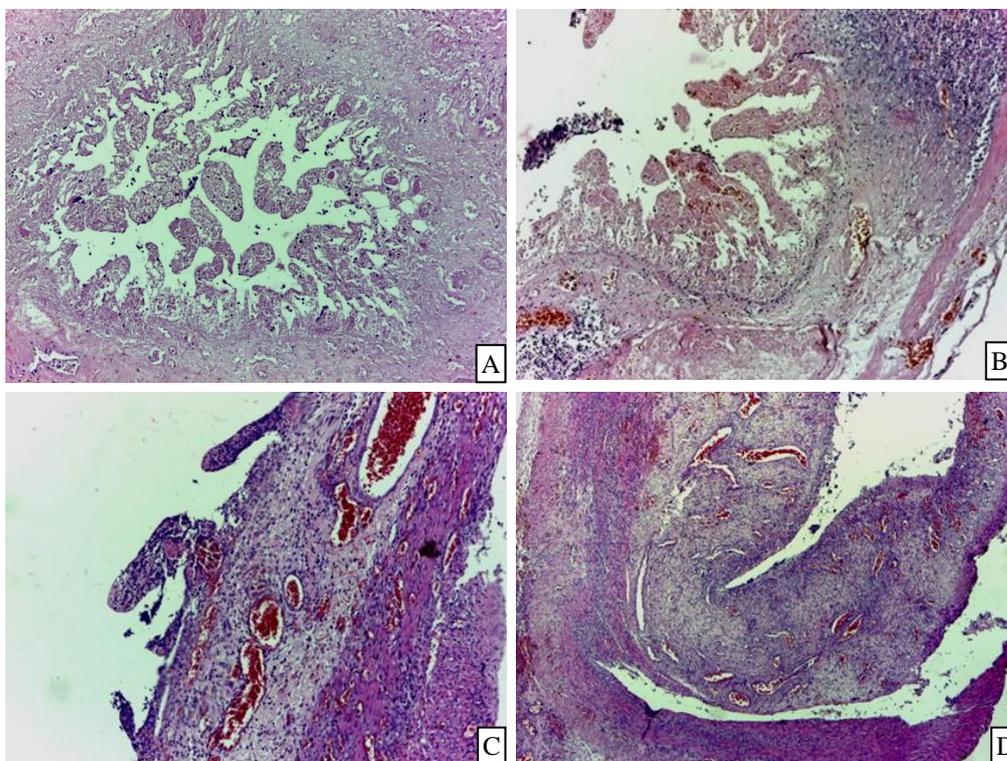


Fig. 3 Microscopic aspect (jejunum, HE, 40x HE, 40x): A – complete coagulation necrosis areas of mucosa (ischemia) of the intestinal wall: transmural infarction; B – necrotised mucosa and submucosa with a normal muscular layer; C – villus necrosis; D – necrotised area, ulcerated and replaced by a granulation tissue.

After the surgical intervention, the patient was transferred to the neonate intensive care unit in order to continue monitoring. The postoperative course was uneventful with a relatively good digestive tolerance for delactosed milk-powder.

Case 2. Segmentary volvulus through postsurgical adhesions (band)

J.A., a 16 years old girl patient arrived in the pediatric emergency department with epigastric pain that actually started within one hour prior the arrival. Laboratory investigations were within normal range and the transabdominal ultrasound didn't show any pathological aspects. At this stage the clinical diagnosis of a dyspeptic syndrome was proposed and a treatment with omeprazolom and sucralfate was prescribed. In the following two days, the abdominal pain persisted, being accompanied by repeated emetic episodes. The patient was hospitalized in the III Pediatric Clinic Cluj-Napoca on 11/04/2013.

The patient was appendicectomized 14 months prior to admission.

Her abdomen was asymmetrically distended with tenderness all over.

The transabdominal ultrasonography revealed at the right side of the abdomen, under the umbilical line, a

distended ileum, without peristaltic movements, with both liquid and solid content that could not be compressed with the transducer (Fig. 4). The Doppler mode examination didn't point out any arterial blood flow signal in the intestinal wall. The abdominal plain radiograph revealed multiple hydroaeric levels.

The clinical, laboratory and imagistic findings were highly suggestive for acute abdomen associated to intestinal necrosis so the patient was transferred to the Department of Pediatric Surgery Cluj Napoca.

The patient was immediately taken to the operating room. The abdomen was accessed through a midline laparotomy. Intra-operatively, a segmental intestinal volvulus determined by a cecum adherence and terminal ileum secondary necrosis were found (Fig. 5). A segmental resection of the affected portion and a termino-lateral ileocolic anastomosis were performed followed by the lavage and drainage of the abdominal cavity.

Microscopically, the resection sample presented a haemorrhagic necrosis affecting all the width of the intestinal wall (Fig. 6). At the surface of the adjacent mesentery fibrin deposits could be seen.

The patient was discharged on postoperative day 8 without further complications.



Fig. 4 Transabdominal ultrasonography: aperistaltic and distended ileum, with fluid and solid content.



Fig. 5 Intraoperative image: adherence; terminal ileum necrosis.

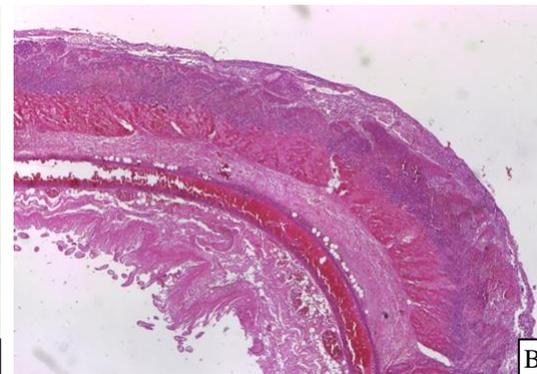
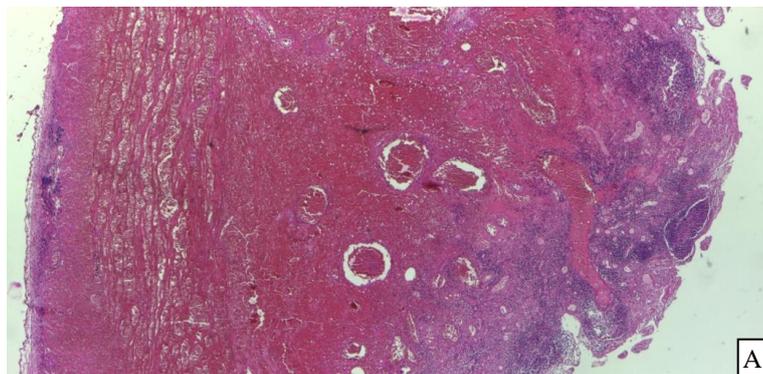


Fig. 6 Microscopic aspect (ileum, HE, 40x): A – intestinal infarction extending to the whole width of the intestinal wall; B – intestinal infarction with mucosal necrosis.

Discussions

Malrotation with volvulus is one of surgical emergencies of infancy and childhood [8].

The process of intestinal rotation begins during the fifth gestational week and involves a series of steps during which the bowel undergoes counter-clockwise rotation around the superior mesenteric artery. This physiologic fixation keeps the ascending and descending colons anchored in the right and left abdominal gutters: the ligament of Treitz in the upper left and the cecum in the lower right [9].

Disturbances in the normal rotational pattern of the midgut will result in a spectrum of malrotation possibilities. Intestinal malrotation is a developmental anomaly affecting the position and peritoneal attachments of the small and large bowels; it has been defined as absent or incomplete rotation and fixation of the embryonic gut around the superior mesenteric artery and predisposes to midgut volvulus. In malrotation, the right colon can be abnormally positioned, resulting in aberrant attempts at fixation; these attempts cause the colon to develop attachments to the right side of the retroperitoneum that have become known as Ladd's bands [9-11].

Symptomatic malrotation is a diagnosis usually made in the newborn and infant; traditional teaching says that up to 75% of cases occur within the newborn period and up to 90% of cases occur within the first year of life [8]. However, intestinal malrotation can occur in patients of any age [12]. Patients with anorectal malformations and two or more VACTERL anomalies should undergo screening for malrotation [13].

The classic clinical presentation of *midgut volvulus* on malrotation is of an infant discharged from the hospital after birth, only to return with bilious vomiting. However, the clinical picture is frequently ambiguous and the child might present with nonbilious vomiting, diarrhea, suspected sepsis, shock or gastrointestinal bleeding; symptoms can be long standing with signs of malabsorption and failure to thrive [14].

Proximal large bowel volvulus is considered as an extremely rare surgical emergency in children. A child with neurodevelopmental delay and a history of constipation presenting with an acute onset of colicky abdominal pain and progressive abdominal distension with vomiting should be suspected of having a cecal and proximal large bowel volvulus [15-16]. Also, in infants and children, *sigmoid volvulus* is exceedingly rare [17].

In acute obstruction through midgut volvulus, the simple radiographs most often show air in the stomach with little or no distal bowel gas. The radiographs may be useful in determining if there is a distal bowel obstruction or free intraperitoneal air [18].

The gold standard diagnostic investigation for intestinal malrotation is the upper gastrointestinal (UGI) contrast study. It may be performed via a nasogastric tube. Thus, barium can be injected into the stomach in a controlled fashion, avoiding overfilling of the stomach. The

normal position of the duodenal-jejunal junction (DJJ) is a critical anatomic landmark at UGI imaging. The DJJ should be located to the left of the left vertebral body pedicle at the level of the inferior margin of the duodenal bulb; it should also be located posteriorly on the true lateral view since it is a retroperitoneal structure [19]. There are variations that may cause the DJJ to be displaced either inferiorly or medially. Situations that may mimic malrotation with abnormal DJJ position include splenomegaly, liver transplant, gastric overdistention, small bowel obstruction, and spinal curvature. In children under the age of 4 years, the normal peritoneal ligaments are lax and so the DJJ may be manually displaced [20]. Therefore, UGI contrast study can occasionally be misleading. There is a significant rate of negative laparotomy following diagnosis of malrotation on UGI contrast study [19, 21].

The role of the enema therefore is a secondary one and may help to determine the position of the cecum and colon in indeterminate cases [18].

The surgical approach to malrotation with or without ("Ladd's procedure") consist of: (1) detorsion of the bowel when volvulus is present, (2) lysis of peritoneal bands, (3) broadening the mesentery to separate the duodenum and cecum as far away as possible, (4) placement of the small bowel to the right side of the abdomen, and (5) placement of the colon to the left side of the abdomen [22]. Historically, surgeons used to perform pexy of small bowel loops to the parietal peritoneum [23].

In case 1, the repeated emesis in a newborn should first of all suggest a congenital digestive malformation. The emesis in the first days could also point out a neonate with a maternofetal infection, the mother being the infected one or just a carrier.

Probably, at the arrival in the pediatric department the neonate was septic due to the peritonitis which appeared consequently to the intestinal perforation (less probably a congenital peritonitis). This case was interpreted as a maternofetal infection and was treated with antibiotics followed by fluid and electrolyte resuscitation.

The alternation of the symptoms with short periods of amelioration was due to the repeated episodes of volvulus (Fig. 3D) with subsequently intestinal ischemia and the excretion of necrotising mucosa as mucosanguineous stools. Consequently, the intestinal infarction lead to perforation and peritonitis. The septic exacerbation was hidden by the antibiotic treatment.

Clinical examination and laboratory investigations suggested medium to severe dehydration characterized by hypovolemic hypotonic hyponatremia with fluid loss in the third space, metabolic alkalosis due to the repeated loss of H⁺ during each emetic episode and hypochloremia given to the loss of Cl⁻ within the HCO₃⁻/Cl⁻ exchanger.

Taking into account the length of the jejuno-ileal portion, a short bowel syndrome should not be excluded.

Peritoneal adhesions may be classified as congenital (embryological anomaly in the development of the peritoneal cavity) or acquired (inflammatory or

postsurgical). Postsurgical adhesions, which constitute the majority of the peritoneal adhesions, develops as a result of the wound healing process (injured tissue surfaces following incision, cauterization, suturing or other means of mesothelial trauma) and can be associated with any kind of abdominal surgery [24-25]. Inflammatory response has a pivotal role in peritoneal adhesion formation through immune cells and mediators [26-28].

Intestinal obstructions, chronic pelvic pain and female infertility are associated postoperative problems with adhesion formation [29].

Adhesions are an important cause for long-term complications in both open and laparoscopic surgery; adhesiolysis during reoperations seems to impact adhesion-related morbidity most [30-31].

Complete adhesion prevention is an unsolved problem, and the search for an ideal antiadhesion agent is still ongoing [32].

In case 2, the adherences appeared consequently to the prior appendicitis surgery. The main complaint was the pain followed by emesis due to the occlusive syndrome. The presence of daily stools most probably originating from the subjacent portion of the volvulus area was misleading, but explanatory for the late diagnosis of the intestinal occlusion.

Conclusions

Midgut volvulus continue to represent a complex problem for surgeons and radiologists. Midgut volvulus can lead to necrosis of the midgut with significant mortality. An interdisciplinary collaboration is important, since patients are not initially evaluated by surgeons.

References

- Gheorghescu B, Rebedea D. Intestinul subțire. București: Editura Medicală; 1975: 356-60.
- Katis PG1, Dias SM. Volvulus: a rare twist on small-bowel obstruction. CMAJ. 2004;171(7):728.
- Akgür FM, Tanyel FC, Büyükpamukçu N, Hiçsönmez A. Anomalous congenital bands causing intestinal obstruction in children. J Pediatr Surg. 1992;27(4):471-473.
- Lepage-Saucier M, Tang A, Billiard JS, Murphy-Lavallée J, Lepanto L. Small and large bowel volvulus: clues to early recognition and complications. Eur J Radiol. 2010;74(1):60-66.
- Klein J, Baxstrom K, Donnelly S, Feasel P, Koles P. A Fatal Twist: Volvulus of the small intestine in a 46-Year-Old Woman. Case Rep Med. 2015;2015:391093.
- Roggo A, Ottinger LW. Acute small bowel volvulus in adults. A sporadic form of strangulating intestinal obstruction. Ann Surg 1992;216(2):135-141.
- Iwuagwu O, Deans GT. Small bowel volvulus: a review. J R Coll Surg Edinb 1999;44(3):150-155.
- Strouse PJ. Disorders of intestinal rotation and fixation ("malrotation"). Pediatr Radiol. 2004;34(11):837-851.
- Warner BW. Malrotation. In: Oldham KT, Colombani PM, Foglia RP (eds) Surgery of infants and children: scientific principles and practice. Lippincott-Raven, Philadelphia, 1997: 1229-1240.
- Marine MB, Karmazyn B. Imaging of malrotation in the neonate. Semin Ultrasound CT MR. 2014;35(6):555-570.
- Aslanabadi S, Ghalehgalab-Behbahan A, Jamshidi M, Veisi P, Zarrintan S. Intestinal malrotations: a review and report of thirty cases. Folia Morphol (Warsz). 2007;66(4):277-282.
- Nehra D, Goldstein AM. Intestinal malrotation: varied clinical presentation from infancy through adulthood. Surgery. 2011;149(3):386-393.
- Chesley PM, Melzer L, Bradford MC, Avansino JR. Association of anorectal malformation and intestinal malrotation. Am J Surg. 2015;209(5):907-912.
- Millar AJ, Rode H, Cywes S. Malrotation and volvulus in infancy and childhood. Semin Pediatr Surg. 2003;12(4):229-236.
- Folaranmi SE, Cho A, Tareen F, Morabito A, Rakoczy G, Cserni T. Proximal large bowel volvulus in children: 6 new cases and review of the literature. J Pediatr Surg. 2012;47(8):1572-1575.
- Takada K, Hamada Y, Sato M, Fujii Y, Teraguchi M, Kaneko K, Kamiyama Y. Cecal volvulus in children with mental disability. Pediatr Surg Int. 2007;23(10):1011-1014.
- Osiro SB, Cunningham D, Shoja MM, Tubbs RS, Gielecki J, Loukas M. The twisted colon: a review of sigmoid volvulus. Am Surg. 2012;78(3):271-279.
- Applegate KE. Evidence-based diagnosis of malrotation and volvulus. Pediatr Radiol. 2009;39 Suppl 2:S161-163.
- Applegate KE, Anderson JM, Klatter EC. Intestinal malrotation in children: a problem-solving approach to the upper gastrointestinal series. Radiographics. 2006;26(5):1485-1500.
- Katz ME, Siegel MJ, Shackelford GD, McAlister WH. The position and mobility of the duodenum in children. AJR Am J Roentgenol. 1987;148(5):947-951.
- Stephens LR, Donoghue V, Gillick J. Radiological versus clinical evidence of malrotation, a tortuous tale-10-year review. Eur J Pediatr Surg. 2012;22(3):238-242.
- Shew SB. Surgical concerns in malrotation and midgut volvulus. Pediatr Radiol. 2009;39 Suppl 2:S167-171.
- Vălean C, Bratu N, Fufezan O, Iacob D, Nanulescu M. Malrotația intestinală cu volvulus recurent. Pediatru.ro. 2007;3(6):66-68.
- Liakakos T, Thomakos N, Fine PM, Derveniz C, Young RL. Peritoneal adhesions: etiology, pathophysiology, and clinical significance. Recent advances in prevention and management. Dig Surg. 2001;18(4):260-273.
- diZerega GS, Campeau JD. Peritoneal repair and post-surgical adhesion formation. Hum Reprod Update. 2001;7(6):547-555.

26. Cheong YC, Laird SM, Li TC, Shelton JB, Ledger WL, Cooke ID. Peritoneal healing and adhesion formation/reformation. *Hum Reprod Update.* 2001;7(6):556-566.
27. Sulaiman H, Dawson L, Laurent GJ, Bellingan GJ, Herrick SE. Role of plasminogen activators in peritoneal adhesion formation. *Biochem Soc Trans.* 2002;30(2):126-131.
28. Chung DR1, Chitnis T, Panzo RJ, Kasper DL, Sayegh MH, Tzianabos AO. CD4+ T cells regulate surgical and postinfectious adhesion formation. *J Exp Med.* 2002;195(11):1471-1478.
29. Kossi JA, Salminen PT, Laato MK. Surgical workload and cost of postoperative adhesion-related intestinal obstruction: importance of previous surgery. *World J Surg.* 2004;28(7):666-670.
30. Kamel RM. Prevention of postoperative peritoneal adhesions. *Eur J Obstet Gynecol Reprod Biol.* 2010;150(2):111-118.
31. Ten Broek RP, Bakkum EA, Laarhoven CJ, van Goor H. Epidemiology and Prevention of Postsurgical Adhesions Revisited. *Ann Surg.* 2016;263(1):12-19.
32. Shin YC, Yang WJ, Lee JH, Oh JW, Kim TW, Park JC et al. PLGA nanofiber membranes loaded with epigallocatechin-3-O-gallate are beneficial to prevention of postsurgical adhesions. *Int J Nanomedicine.* 2014;9:4067-4078.

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