Synchronous rectal prolapse and midgut volvulus presenting as complete small bowel gangrene

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ABSTRACT: Introduction: Adult midgut malrotation is very rare. The presentation in adults is mostly subtle; the diagnosis is often made on imaging.

Case report: A 32-year-old man presented with a 3-day history of an irreducible painful mass protruding per rectum, followed by abdominal pain and constipation. The patient was febrile and toxic, with marked signs of peritonitis and complete full-thickness irreducible rectal prolapse. Abdominal radiographs showed multiple air-fluid levels. A diagnosis of irreducible rectal prolapse with intestinal obstruction was made. Laparotomy revealed complete small bowel gangrene, abnormal rotation of the small bowel 180° around the mesenteric root, hypermobile duodenojejunal flexure and ileo-caecal junction and an abnormally mobile caecum lying in the central abdomen. Abnormal mobility of the sigmoid colon with a large mesocolon, and a large irreducible rectosigmoid intussusception, was also noted. Our patient is probably the first case in literature, which may prompt awareness of simultaneously-occurring disorders of fixation, and preventive steps.

KEYWORDS: acute intestinal obstruction, midgut malrotation, rectal prolapse, synchronous disorders of fixation

ABBREVIATIONS
CT – computed tomography

INTRODUCTION

Disorders of intestinal fixation with midgut malrotation and volvulus are common causes of intestinal obstruction in children. The incidence of midgut malrotation has been reported to be 1 in 500 live births, and the vast majority of cases present during infancy [1, 2]. Adult midgut malrotation, however, is exceedingly rare. The presentation in adults is mostly subtle, with colicky pain and other chronic atypical symptoms; the diagnosis is often made on imaging [1, 3]. Only a few cases of intestinal obstruction due to adult midgut malrotation and volvulus have been reported [1–4].

Other disorders of fixation include long redundant segments of intestinal mesentery, whether small or large bowel, which are the usual culprits in the causation of volvulus. Only a few case reports describe the synchronous or metachronous occurrence of volvulus involving multiple parts of the colon [5–9]. Other combinations of the simultaneous occurrence of two disorders of fixation have not been reported.

We report a unique case of a 32-year-old man, who presented with synchronous acute gangrenous midgut volvulus and an irreducible rectal prolapse. This combination, we believe, is hitherto unreported in literature. We present challenges faced during treatment along with a brief review of relevant literature.

CASE REPORT

A 32-year-old man was brought to the surgical emergency with a 3-day history of an irreducible painful mass protruding per rectum, followed a day later by the appearance of generalized abdominal pain, distension and constipation. The rectal protrusion had been recurrent and reducible over the previous 18 months, however, no treatment had been sought due to socioeconomic reasons. There was no history suggestive of bowel obstruction, and no other significant past medical or surgical history. On examination, the patient was febrile (temperature = 39.5°C) and toxic, with a GCS of 15/15. The pulse was 100/minute and feeble, BP = 82 systolic, and respiratory rate = 28/minute. Abdominal examination revealed marked distension with signs of peritonitis with generalized tenderness and guarding. Rectal examination revealed a complete full-thickness irreducible rectal prolapse, with congested and edematous rectal mucosa (Fig. 1.). Blood investigations revealed a total leucocyte count of 1,700/mm³ and blood urea = 64 mg/dL. Other laboratory tests were normal. Abdominal radiographs showed dilated small bowel loops with multiple air-fluid levels. A diagnosis of irreducible rectal prolapse with intestinal obstruction due to edema and fecalith impaction was made, and the patient was resuscitated with intravenous fluid and antibiotics. Exploratory laparotomy was performed after 2 hours of resuscitation. Operative findings were complete small bowel gangrene, abnormal rotation of the small bowel 180° around the mesenteric root, hypermobile duodenojejunal flexure and ileo-caecal junctions, and, an abnormally mobile caecum lying in the central abdomen (Fig. 2.). Abnormal mobility of the sigmoid colon with a large mesocolon, and a large irreducible rectosigmoid intussusception, was also noted. The small bowel volvulus was untwisted, the rectosigmoid prolapse was reduced with a combined abdominoperineal approach and 100% oxygen was administered through the endotracheal tube for 5 minutes. However, there was no improvement in the perfusion to the small bowel. The patient’s intraoperative condition was also poor, with refractory hypotension and negligible urine output. Resection of the complete small bowel from duodenojejunal flexure to ileo-caecal junction was performed, with suture rectopexy to the sacrum and bag laparostomy. Unfortunately, the patient’s condition worsened, and he died 6 hours after the surgery.
Midgut malrotation is a congenital anomaly where incomplete or absent rotation of the fetal intestines around the axis of the superior mesenteric artery predisposes the patient to variable and recurrent episodes of obstruction. Midgut malrotation is extremely rare in adults, i.e. less than 0.1% of cases. In most adults, the presentation is chronic, with a long-standing undiagnosed abdominal pain. Ultrasound or CT findings of ‘whorled-up’ bowel loops with a mobile cecum may be important clues to diagnosis [2, 3]. From another perspective, midgut malrotation is also a disorder of improper ‘fixation’ of the small bowel mesentery. Narrow vertical attachments in the form of peritoneal fibrous bands may persist (Ladd bands) and cause duodenal obstruction. Other complications like paraduodenal hernia or small bowel volvulus can also occur [3]. These complications of midgut volvulus are extremely rare in adults; we could find only about 10 patients in five articles on the subject [1–4, 10].

Rectal prolapse is a fairly common disorder of fixation of the rectum and sigmoid to the sacrum mainly seen in older patients. Resection of the intussusception and fixation by mesh or sutures remains the cornerstone of therapy [11]. This condition is also seen in younger patients. We treated a case with simultaneous acute gangrenous midgut volvulus in an adult patient, along with an irreducible rectal prolapse. To the best of our knowledge, this combination is hitherto unreported in literature. Our case, therefore, represents the simultaneous occurrence of two disorders of fixation. The first query that arose was whether it was possible for multiple segments of the bowel to be fixed inadequately, or with an abnormal mesentery. The second was whether a common etiopathogenesis could be attributed to this combination. On searching PubMed with MeSH keywords: rectal prolapse, small bowel gangrene, volvulus, midgut malrotation, simultaneous, we could not locate any co-existent disorder of fixation with either rectal prolapse or midgut malrotation. However, a few case reports of synchronous or metachronous volvulus of different parts of the colon were identified [5–9]. It has been shown that the sigmoid is the most likely part of the colon to undergo volvulus, while the transverse colon is the least likely one [6, 7]. Avgerinos et al. found metachronous volvulus of the sigmoid colon, cecum, and stomach. The authors attributed the cause to a very long mesentery, coupled with an increased mobility of the organs. They feel that prophylactic pexis of the cecum and the stomach during the first operation, could have prevented the subsequent episodes of volvulus [8]. Apart from a long mesentery, other hypothesized causes of simultaneous volvulus include ‘dolichocolon’ (abnormally long colon seen in the elderly patients) and neuropsychiatric disorders (Parkinson’s disease, Chagas’ disease etc.) [8, 9]. It remains to be seen whether these factors could be responsible for the simultaneous ‘mal-fixation’ of the small bowel and the rectum, as in our case.

In conclusion, our patient was probably the first reported case in literature. Due to the sudden onset of illness and its extreme rarity, a detailed pathogenesis is difficult to offer. However, a study of similar cases may prompt surgeons to be aware of simultaneously-occurring disorders of fixation, and take preventive steps if the need arises.
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