Rare Gastrointestinal Malrotation in an Adult Coexisting with an Incarcerated Incisional Hernia: A Case Report

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Abstract

Background: Intestinal malrotation in adults, a previously overlooked entity, coexisting with an incisional hernia is rare. The occurrence of these two acute abdominal conditions can increase the morbidity and/or mortality even with prompt surgical intervention. The study aimed to report a rare case of adult intestinal malrotation co-existing with an incarcerated incisional hernia, emphasizing the diagnostic and surgical challenges involved.

Case Summary: We present a 22-year-old who had an incarcerated incisional hernia following omphalocele repair who presented with chronic abdominal pain and subsequent partial intestinal obstruction. Following adequate perioperative resuscitation, urgent exploratory laparotomy was done, and an incarcerated bowel within a 2 cm umbilical fascia defect, a wide mesentery and a non-rotated bowel were observed. An appendectomy and a tissue-based repair of the defect were performed under general anaesthesia with endotracheal intubation. Immediate post-operative period and subsequent recovery profile were uneventful and she was discharged postoperative day seven.

Conclusion and Recommendation: This case highlights the clinical complexity of coexisting intestinal malrotation and incisional hernia following omphalocele repair. Adults presenting with atypical abdominal symptoms, especially in the context of previous abdominal wall defects, should be suspected. Multidisciplinary evaluation and individualized surgical planning are essential for a good outcome.

Keywords: Incisional hernia, Omphalocele, Intestinal malrotation, Ladd's procedure

Introduction

Intestinal malrotation is very rare in adult [1] and can be difficult to diagnose due to its non-specific symptoms.[2] Hence, its diagnosis in adults requires a high index of suspicion as delay can lead to significant morbidity and mortality. Therefore, awareness of its subtle presentations by general surgeons and its radiological features by radiologists will aid its prompt diagnosis in adults.

We report an incidental finding of an intestinal malrotation during exploratory laparotomy following a history of chronic abdominal pain in a patient who had a repair of an omphalocele major at infancy. This case is being reported according to the SCARE guidelines.[3]

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Case Presentation

A.T is a 22-year-old woman who presented to our surgical outpatient clinic with a 12 years history of intermittent colicky central abdominal pain that became severe in intensity 3 years prior, with no previous history of trauma to her abdomen. Over the first 9 years of her chronic abdominal pain history, the pain was initially said to be aggravated by ingestion of solid meals and relieved by Buscopan, but it was neither associated with abdominal distension, fever, vomiting, reduced stool calibre, melena, nor change in bowel habits. She is a known peptic ulcer disease patient with no other comorbidity, but had a repair of omphalocele major 22 years ago at infancy in our hospital. The abdominal pain was '*quite bad*' that she became addicted to pentazocine which she was procuring without prescription from an unregulated source. She was however successfully rehabilitated in a formal program.

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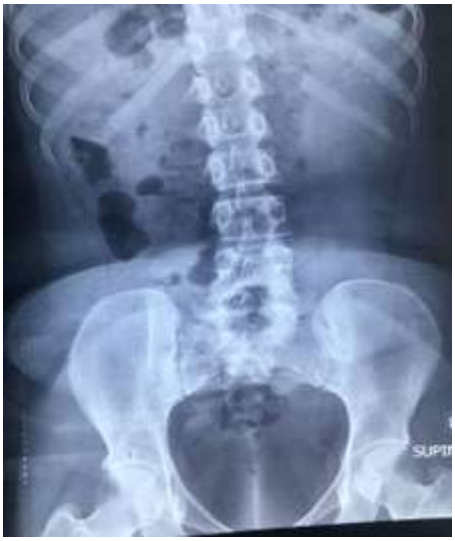

Figure 1: Supine Abdominal X-ray without features of intestinal obstruction

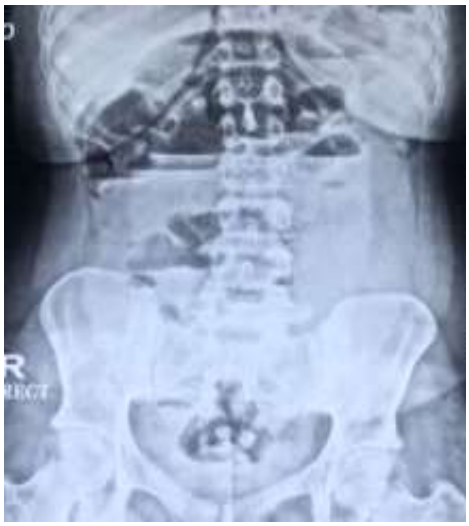

Figure 2: Plain X-ray Erect Showing Multiple Air-Fluid Levels

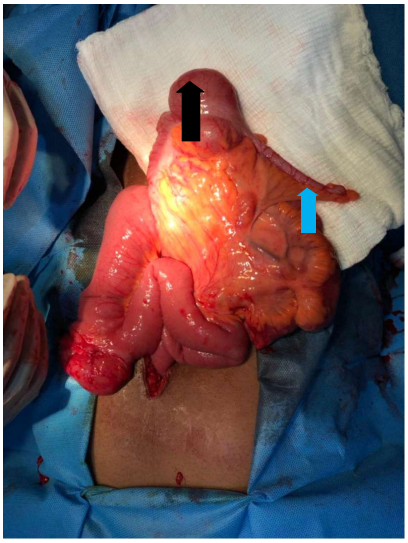

Figure 3: Blue arrow pointing to the appendix. Black arrow pointing to the cecum

On examination, she was in moderate painful distress with no pallor and jaundice. There was a transverse central abdominal scar with a peri-umbilical tenderness and rebound tenderness but no guarding. Her bowel sound was normo-active and her rectum was filled with faeces.

A clinical impression of symptomatic umbilical hernia was made with an acute exacerbation of peptic ulcer disease as a strong differential diagnosis.

Upper GI Endoscopy (esophago-gastro-duodenoscopy) revealed an antral polyp (gastric adenomyoma) and evidence of gastritis. Plain abdominal x-ray (Figure 1) and a computerized tomography (CT) scan of her abdominopelvic region showed an umbilical hernia with a loop of bowel. Her basic metabolic panel - serum electrolytes, urea and creatinine and packed cell volume (34%) were essentially normal.

Pharmacologic interventions with triple therapy with omeprazole, clarithromycin and amoxicillin to eradicate H. pylori, analgesics (dihydroxycodone and paracetamol) and occasional intramuscular injection of Buscopan were instituted but they failed to provide a significant resolution of her symptoms. She had used both orthodox and unorthodox interventions at various times during the duration of the symptoms.

Days prior to her presentation to our emergency department, she started experiencing frequent and severe colicky abdominal pain, bilious vomiting, and constipation. She was in painful distress with tachycardia and tachypnoea. Abdominal auscultation revealed a hyperactive bowel sound with scanty faeces in her rectum, and a tentative clinical diagnosis of an incarcerated incisional hernia with partial bowel obstruction was made. The plain abdominal x-ray showed multiple air-fluid levels with the presence of gas in the rectum (Figure 2).

She was immediately optimized, prepared, and an urgent exploratory laparotomy was performed under general anaesthesia. The intra-operative findings included a 2 cm defect beneath the scar, an incarcerated loop of ileum 35 cm from the ileocecal junction (ICJ), and an intestinal malrotation with a wide base (peritonealized caecum, appendix and the ascending colon, which were in the left upper quadrant of the abdomen) [Figure 3]. No Ladd's band nor ischemia of the bowels were noted. She had a tissue-based repair of the abdominal wall defect and an appendectomy, and the abdomen was closed in layers.

She was placed on *nil per oris*, intravenous fluids, parenteral antibiotics, and analgesics to maintain fluid balance, prevent infections and treat surgical pain. The post-operative period was uneventful, and she was discharged home 7 days after laparotomy. She was followed up at the Surgical Out-Patient Department at two and eight weeks post-operatively. The chronic abdominal pain had since resolved, and she was satisfied with the outcome of surgery.

Discussion

Intestinal malrotation is a spectrum of disorders. It is defined as the digression from normal 270 degrees anti-clockwise rotation of the midgut at embryologic stage.[4] Most of the cases of intestinal malrotation present within the first 12 months of life.[5] The prevalence of the condition in adulthood is unknown but a rate of 0.17

percent was suggested from screenings with a CT colonography. [6] The unfamiliarity of general surgeons with this rare condition can cause diagnostic errors and delays in treatment with consequent fatalities.[7] Although the reports of plain abdominal X-ray did not suggest the presence of an underlying intestinal malrotation, barium meal with follow-through might have assisted in the detection of the malrotation.

Intestinal malrotation can be classified into nonrotation and incomplete rotation.[8] The most common type seen in adults is the nonrotation, which is associated with exomphalos. In intestinal nonrotation, the small bowel is located on the right lower abdomen while the cecum is on the left upper abdomen. There is a lower incidence of volvulus compared to other types of malrotation.

Some studies reported that about 40 to 50 percent of patients with midgut nonrotation are symptomatic,[8,9] which could be either acute or chronic.[10] The chronic intermittent symptoms are more common in adults, and this makes their diagnosis challenging. In most instances, investigations would have been carried out, and several differentials would have been considered before it is diagnosed in adults.[11] The presentation of the index patient in this case report fits into the chronic intermittent symptomatic type (intermittent colicky abdominal pain which usually worsens after eating) that was eventually diagnosed at exploratory laparotomy after a long period of preoperative misdiagnosis.

Incomplete intestinal rotations are definitively treated by Ladd's procedure which involves; division of the Ladd's band and the widening of the narrow mesentery (the duodenum was mobilized, and the adhesions around the Superior mesenteric artery were divided), placing the cecum in the left lower quadrant within the peritoneal cavity and a prophylactic appendectomy is done.[12]

Incisional hernia following the repair of congenital abdominal wall defects, such as gastroschisis and omphalocele major, is about 3%.[13] However, its persistence into adulthood is not known. Omphalocele is the most common abdominal wall defect in neonates.[13] Various surgical techniques, such as tissue expanders,[13] component separation technique,[14] and the use of mesh,[15]have been used to reduce the incidence of incisional hernia after the repair of abdominal wall defects. This index case illustrated the challenges of closure of omphalocele major in paediatric patients, which eventually presented as intestinal malrotation with incisional hernia at adulthood.

Conclusion

Intestinal malrotation in adults remains a diagnostic challenge due to its rarity and non-specific presentation. Adult presentation of intestinal malrotation, though rare, should be considered in patients with chronic abdominal pain and prior abdominal wall surgeries. Intestinal malrotation in this case was diagnosed intraoperatively, emphasizing the need for a high index of suspicion, especially in patients with history of congenital anomalies. Multidisciplinary evaluation and individualized surgical planning are essential for a good outcome.

For closure of defects in the paediatric patients with omphalocele major, the use of tissue expanders, mesh and components separation techniques will go a long way in preventing the later occurrence of an incisional hernia.

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