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A rare case of intestinal malrotation presenting as appendicitis and subacute intestinal obstruction in late adulthood

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Abstract

A case presentation of a patient having acute abdomen with findings of acute appendicitis, subacute intestinal obstruction and Malrotation.

Keywords: intestinal malrotation presenting, intestinal obstruction, late adulthood

Introduction

Midgut malrotation is a foetal anomaly resulting from the failure of rotation around the axis of the superior mesenteric artery and subsequent retroperitoneal fixation. It is a rare condition with an approximate incidence of one in 500 live births (1). 85% of cases have been estimated to present in the first two weeks of life (2). Presentation at adulthood is rare. Hence, it is difficult to establish an accurate incidence as this condition can be asymptomatic and undetected until it is discovered in investigations or surgery for other abdominal pathologies in adulthood (3). Clinical suspicion of intestinal malrotation declines with age because of the rarity of the disorder. We Present a case of 41 year old Male patient who presented with acute abdomen with preoperative ultrasound finding showing possibility of small bowel obstruction, inflamed appendix in right epigastric region, mild malrotation and minimal free fluid. The patient underwent an emergency laparotomy and LADD's operation was done.

Case-Report

A 41 year old male presented with complaints of Generalised abdominal pain for 3 days. Pain was sudden in onset, dull, generalised and radiating to back and was not relieved by taking painkillers. He had no significant past history of comorbidities or didn't underwent any Surgical procedure. He had history of Bidi smoking of 25/day for 10 years. On examination he had normal temperature and 78 pulse with Bp being 100/64mmhg. Respiratory and Cardiovascular examination was Normal. On abdominal examination he had Generalised Tenderness. Per rectal examination was normal with no ballooning. He was then Investigated. He had normal blood reports with Total WBC count being 10000 with Normal chest xray findings.



Fig 1: xray abdomen standing of the patient showing gas filled bowel loops

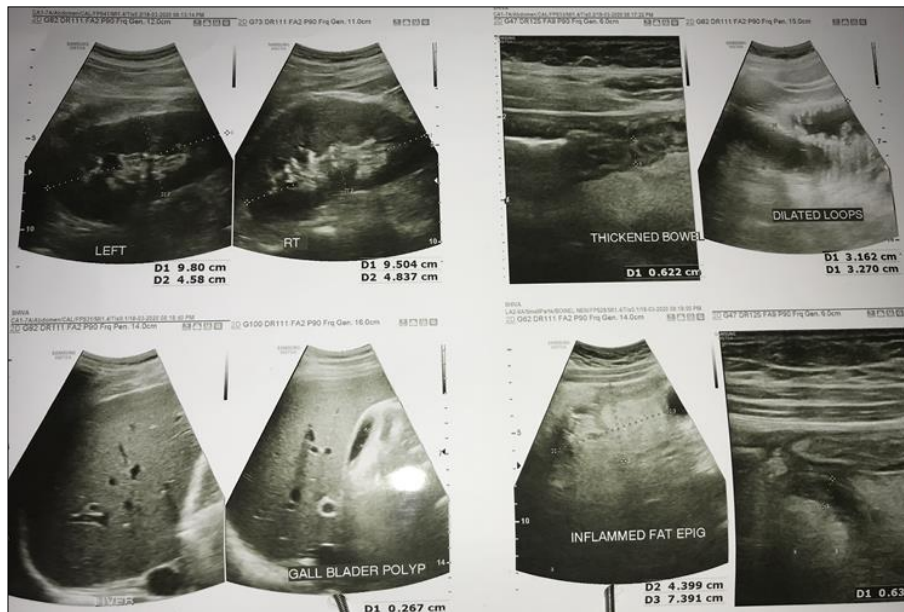


Fig 2: USG abdomen plates showing the findings described in manuscript

On Xray abdomen Standing Gas filled bowel loops were noted

On Abdominal Ultrasound following were found

Possibility of small bowel obstruction, inflamed appendix in right epigastric region, mild malrotation, inflamed fat and minimal free fluid.

He underwent Exploratory Laparotomy

Intra Operatively there was non-rotation of DJ with ceacal rotation.

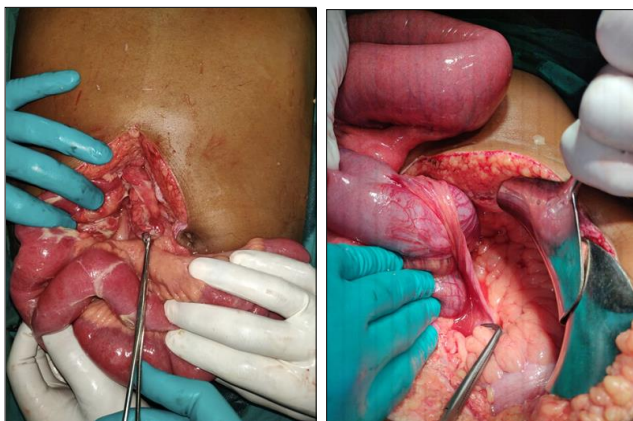


Fig 3: Intraoperative photographs showing a) appendix and caecum in epigastric region b) Ladd's band

Caecum was present in epigastrium with Ladd’s band above DJ flexure (on left side). Appendicular perforation was present at body with purulent peritoneal contamination. Exploratory Laparotomy was done with release of Ladd’s band and appendicectomy with thorough peritoneal lavage (Ladd’s Operation). Patient had Uneventful hospital stay with no major complications in post op follow ups.

Discussion

Embryology: During the sixth week of fetal development, the midgut grows too rapidly to be accommodated in the abdominal cavity and therefore herniates into the umbilical cord. Between the tenth and twelfth weeks, the midgut

returns to the abdominal cavity, undergoing a 270° counterclockwise rotation around the superior mesenteric artery. Because the duodenum also rotates caudal to the artery, it acquires a C-loop that traces this path. The cecum rotates cephalad to the artery, which determines the location of the transverse and ascending colon. Subsequently, the duodenum becomes fixed retroperitoneal in its third portion and at the ligament of Treitz, while the cecum becomes fixed to the lateral abdominal wall by peritoneal bands. The takeoff of the branches of the superior mesenteric artery elongates and becomes fixed along a line extending from its emergence from the aorta to the cecum in the right lower quadrant. Genetic mutations likely disrupt the signaling critical for normal intestinal rotation. For instance, mutations in the gene *BCL6* resulting in absence of left-sided expression of its transcript lead to reversed cardiac orientation, defective ocular development, and malrotation. The essential role of the dorsal gut mesentery in mediating normal intestinal rotation and the role of the forkhead box transcription factor *Foxf1* in formation of the dorsal mesentery in mice are consistent with the noted association of intestinal malrotation with alveolar capillary dysplasia, caused by mutations in *FOXF1*. If rotation is incomplete, the cecum remains in the epigastrium, but the bands fixing the duodenum to the retroperitoneum and cecum continue to form. This results in (Ladd’s) bands extending from the cecum to the lateral abdominal wall and crossing the duodenum, which creates the potential for obstruction. The mesenteric takeoff remains confined to the epigastrium, resulting in a narrow pedicle suspending all the branches of the superior mesenteric artery and the entire midgut. A volvulus may therefore occur around the mesentery. This twist not only obstructs the proximal jejunum, but also cuts off the blood supply to the midgut. Intestinal obstruction and complete infarction of the midgut occur unless the problem is promptly corrected surgically. Malrotation is interference in the process of normal rotation of midgut in fetus and its mesenteric fixation. Stages of Normal Rotation of midgut are; Stage 1: In 4th-8th week of intrauterine period, midgut supplied by superior mesenteric artery (SMA) grows rapidly. As coelomic cavity cannot accommodate growing midgut during that period, it

protrudes into the umbilical cord as physiological hernia. Stage 2: In 10th-12th week, midgut migrates into coelomic cavity. First, small bowel returns towards the left side of the abdomen. Then caecocolic loop returns to left lower abdomen. It rapidly rotates 270° counterclockwise to reach right iliac fossa. Then duodenojejunal segment rotates 270° counterclockwise to reach left of SMA and behind the colon. Stage 3: Fusion of different parts of mesentery and posterior peritoneum.

Different errors of Rotation: Stage 1: Exomphalos major/minor or gastroschisis. Stage 2: Errors of rotation in this stage is important and is usually considered under malrotation. Nonrotation: Causing small bowel in right side; colon in left side; caecum in midline as suspension. Incomplete rotation: It is the most common type of malrotation. Caecum is located in subhepatic right hypochondrium. Ladd's peritoneal band connects from caecocolic loop to posterior abdominal wall compressing the 2nd part of the duodenum. Entire midgut is hanging down along with SMA with a narrow based mesentery causing midgut volvulus. Reverse rotation: Final 180° rotation occurs clockwise bringing colon posterior to duodenum and SMA. Hyper-rotation: Rotation up to 360° or 450° causing caecum on left side of the abdomen. Encapsulated small bowel occurs while fetal midgut returns into coelomic cavity. Stage 3: Final defect in fixation causes mobile caecum and ascending colon leading into caecal volvulus.

Presentation: Acute/recurrent/subacute intestinal obstruction. Midgut volvulus (30%) (Usually clockwise rotation) with features of strangulation, perforation, peritonitis. Shock, septicaemia, passage of dark blood per rectum, oedema and erythema of anterior abdominal wall. In children—failure to thrive, recurrent abdominal pain, cyclical vomiting, constipation and diarrhoea.

Intestinal malrotation is one of the rarest causes of mechanical bowel obstruction. In adults, the incidence rate is 0.2%, and 15% of all patients with confirmed diagnosis remain asymptomatic throughout life. Surgery is generally required when the patient is symptomatic.

Management: Resuscitation, antibiotics, fluid and electrolytes, blood transfusion followed by Laparotomy through horizontal incision is done. Clockwise rotated midgut which is congested and cyanotic is identified. Untwisting of the midgut in counterclockwise direction is done. Viability of bowel is confirmed (colour, vessels in mesentery, peristalsis, on table Doppler). Ladd's band is divided. Large bowel is repositioned in left side. The entire duodenum is Kocherised and the ligament of Treitz is divided so that duodenum becomes straight towards right iliac fossa. This achieves wide root of the mesentery and places the small bowel in the right side of the abdomen thus preventing further volvulus. A complementary appendectomy is done—"Ladd's operation". In our case the appendix was inflamed so we had to remove it.

Conclusion

Appendicitis is a common surgical condition with various clinical presentations. In cases where peritonism is elicited elsewhere other than the right iliac fossa, clinicians could bear in mind the possibility of underlying intestinal malrotation, as this could be the first presentation of this rare congenital condition. Intestinal malrotation is

infrequently encountered in the adult population; however, it should be considered as a differential diagnosis whenever a case of acute intestinal obstruction in an adult presents without any significant past history.

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