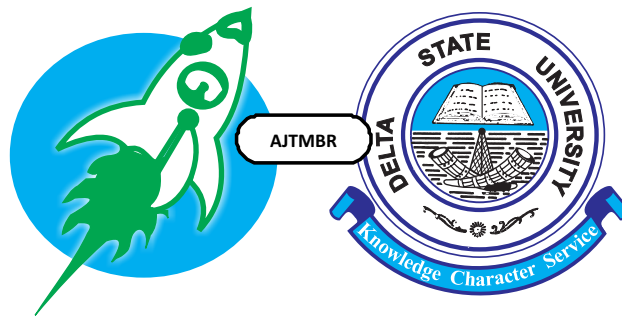


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Diagnostic Delay in Intestinal Malrotation Presenting During Adolescence: A Case Report and Review of Literature

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ABSTRACT

Introduction: Intestinal malrotation refers to any variation in the rotation and fixation of the gastrointestinal tract during development. While the majority of cases present in infancy, particularly with acute obstructive symptoms, a significant number of cases which remain undiagnosed until later in childhood or even adulthood often exhibit vague or nonspecific symptoms, complicating timely diagnosis and management.

Case Report: We report a 10 year old boy presenting with recurrent abdominal pains of 5 years duration associated with recent onset bilious vomiting and constipation. Requested barium meal and follow through showed markedly distended stomach and proximal duodenum that ended blindly. He had exploratory laparotomy with intraoperative finding of malrotation. Ladd's procedure was done and did well postoperatively.

Conclusion: This case report is intended to remind physicians to include intestinal malrotation as a differential diagnosis when an older child presents with recurrent abdominal pain. As early detection may prevent catastrophic bowel ischaemia, chronic midgut volvulus, malnutrition, short bowel and other complications.

Key words: Intestinal malrotation, adolescence, recurrent abdominal pain, Ladd's procedure

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INTRODUCTION

Intestinal malrotation (IM), which encompasses intestinal non-rotation or incomplete rotation, refers to any variation in the rotation and fixation of the gastrointestinal tract during development; this leads to various acute and chronic presentations of disease.¹ The incidence of intestinal malrotation is one in every 200-500 newborns, however symptomatic cases occur in 1 in 6,000 newborns.² While the majority of cases present in infancy, particularly with acute obstructive symptoms, a significant number of

cases remain undiagnosed until later in childhood or even adulthood. This diagnostic delay can have severe implications, as children presenting after infancy often exhibit vague or nonspecific symptoms, complicating timely diagnosis and management.³

In older children, the classic symptoms of bilious vomiting and abdominal distension are less frequently observed, resulting in a delay in diagnosis and poorer outcomes. Arthur *et al*⁴ reported an average time to diagnosis of three

months in patients older than one year, with one case experiencing a delay of up to two years due to atypical presentations such as retrosternal pain. The clinical presentation of IM in older children can range from chronic abdominal pain to intermittent symptoms that may last for months or years. Research by Nagdeve *et al*⁵ indicated that children over the age of two exhibited significantly increased chronic symptoms compared to their younger counterparts, emphasizing the importance of recognizing these atypical manifestations. Symptoms such as non-bilious vomiting, feeding intolerance, and failure to thrive are common, with many children experiencing recurrent abdominal pain and frequent hospitalizations.⁶ Thus, the aim of this case report is to remind physicians to include intestinal malrotation as a differential diagnosis when an older child presents with recurrent abdominal pain, especially when it is associated with vomiting and other gastrointestinal symptoms.

CASE REPORT

A 10 year-old-male admitted into the Children Emergency Unit with a two week history of abdominal pain, vomiting and constipation. The pain was mostly located at the epigastric and umbilical regions, colicky with no known relieving or aggravating factors. Vomiting was projectile, bilious and he vomited about 3 times per day. There was a history of recurrent epigastric pain for the last 5 years, the cause of which remained unidentifiable but received antacids with no relief of the pain. There was no abdominal distension. There were no previous abdominal surgeries or any comorbidity. He was managed for acute exacerbation of peptic ulcer disease at a private hospital for 5 days with no relief of symptoms before referral to our centre.

At presentation, he weighed 27 kg and was

dehydrated. His abdomen was scaphoid with no areas of tenderness. There was no organomegaly and his bowel sounds were normoactive. His rectum was collapsed but contained formed stool. Complete blood count showed Haemoglobin concentration of 12 g/dl with normal counts of white blood cells and platelets. Serum electrolytes showed hypokalemia. Abdominal X-Ray on presentation showed double bubble sign. He was resuscitated and hypokalemia was corrected. Requested barium meal and follow through showed markedly distended stomach and proximal duodenum that ended blindly. He was then worked up for exploratory laparotomy on suspicion of duodenal web.

The findings at surgery were: Markedly distended stomach and proximal duodenum with a 360° torsion of the midgut around the superior mesenteric vessels. There were also Ladd's bands from ascending colon to the right lateral wall of the peritoneal cavity. Ladd's procedure was then performed. He had an uneventful post operative recovery period and was discharged home after one week. He has been followed up for 3 months post operatively with no recurrence of the symptoms.

DISCUSSION

Cases of intestinal malrotation presenting during adolescence poses a significant challenge as it is not usually considered as the likely diagnosis based on clinical grounds. These patients often present with symptoms that are mistaken for irritable bowel syndrome, peptic ulcer disease, biliary and pancreatic disease, and psychiatric disorders.⁷ Symptoms of intestinal malrotation are linked to the anatomical anomalies associated with it and two distinct patterns of presentation in older children and adults have been reported in the literature: acute and chronic.⁸ The chronic presentation is more common. This is characterised by intermittent crampy abdominal

pain, bloating, nausea and vomiting over several months or years which underlines the need for a high index of suspicion of midgut malrotation in older children as the cause of these intermittent and varying abdominal symptomatology.⁸ Diagnostic delays are common in this group of patients because of the non-specific nature of the presentations. The pathophysiology of these chronic symptoms may relate to the compression effect of Ladd's bands running from the caecum and ascending colon to the right abdominal wall.⁸

Acute presentation is usually characterized by features of acute bowel obstruction with or without chronicity of symptoms. In cases of acute presentation in the older child or adult with malrotation, midgut volvulus is the most common cause of the bowel obstruction.⁹ This was the case in our patient who presented with acute symptoms of colicky abdominal pains and vomiting but had been having chronic symptoms of recurrent abdominal pains for 5 years prior to the acute symptoms.

The diagnosis of malrotation can be made by radiographic studies. In the absence of volvulus of the midgut, a plain x-ray of the abdomen is of little diagnostic value. The absence of cecal gas shadow or the localization of small intestinal loops predominately in the right side should arouse the suspicion of malrotation.¹⁰ The standard upper gastrointestinal series is considered the gold standard for diagnosis and may show a vertical duodenum that does not cross the midline. The entire small bowel is found in the right half of the abdomen. The accuracy of the upper gastrointestinal series is reported to be over 80%.¹⁰ However, in the index case, the findings on upper GI series done were not typical of malrotation syndrome, rather the features were in keeping with a duodenal obstruction. This is probably because the patient

had a complete obstruction from the 360° torsion of the midgut around the superior mesenteric vessels.

Malrotation is increasingly being diagnosed with Computed Tomography scan by the anatomic location of a right-sided small bowel, a left-sided colon, an abnormal relationship of the superior mesenteric vessels, and aplasia of the uncinate process.¹¹ Similarly, abdominal colour Doppler Ultrasound scan may reveal malposition of the SMA, raising the suspicion of gut malrotation with or without the abnormal location of the hollow viscus.¹²

The classic treatment for malrotation is the Ladd's procedure. This involves de-rotation of the bowel if torsion is present, division of the peritoneal attachments lying across the duodenum from cecum to right upper quadrant, widening of the base of the mesentery, appendectomy, taking down the ligament of Treitz, straightening the duodenum to the right and finally, returning the bowel to a position of non-rotation with the cecum placed into the left upper quadrant.¹³ Published reports for the laparoscopic Ladd procedure are now appearing in literature as well Laparoscopic Ladd's procedure in asymptomatic patients which confers a shorter time to tolerating feeds, shorter length of stay, and a decreased rate of bowel obstruction and other complications. It may, however, carry a higher risk of postoperative volvulus.¹⁴ Diagnostic laparoscopy is recommended in asymptomatic patients with high suspicion where the diagnosis is not certain after upper gastrointestinal imaging.¹⁵ Our patient had open Ladd's procedure with a good postoperative outcome and has remained asymptomatic after a 3 month follow-up period.

Malrotation of the intestine, though commonly seen in infancy, is a condition that should be

suspected in older children when the patient present with prolonged and recurrent abdominal pain and vomiting. There is a need to have a high index of suspicion for the condition and the use of appropriate imaging modality could help clinch the diagnosis. Early suspicion and detection by the paediatrician and paediatric surgeon may prevent catastrophic bowel ischaemia, chronic midgut volvulus, malnutrition, short bowel and other complications. Complete resolution of acute obstruction or chronic abdominal pain is the result of a high index of suspicion for malrotation, appropriate diagnostic studies, and aggressive definitive treatment.

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