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Case Report

Intestinal mal-rotation in an adolescent boy

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ABSTRACT

Intestinal malrotation is a developmental anomaly of the midgut in which the normal fetal rotation of intestines around the superior mesenteric artery and their fixation in the peritoneal cavity fail. Rotational anomalies of the midgut are rare in adolescent age. Operative intervention is required generally when they are symptomatic. While difficult to diagnose, prompt recognition and surgical treatment usually lead to a successful outcome. So here we have discussed this rare case of malrotation in adolosance, similar study has been published by Nakajima et al, in-world journal of emergency surgery 2013**.**

Keywords: Intestinal malrotation, Adults malrotation, Obstruction

INTRODUCTION

Congenital bowel malrotation resulting in midgut volvulus is traditionally regarded as a diagnosis of infancy.[1] Rarely, congenital bowel malrotation is diagnosed in adolescents or adults and requires a high index of suspicion. Presentations can be acute or chronic, and physical examination findings are nonspecific. Diagnosis is primarily achieved through abdominal computed tomography (CT) or during exploratory laparotomy. The pathophysiology in late-onset malrotation is similar to neonatal malrotation, with a division of Ladd's bands–peritoneal fibrous bands that connect the cecum to the right lower quadrant retroperitoneum-as the definitive treatment. We present a case of congenital bowel malrotation in an adolescent with persistent and worsening migratory abdominal pain.

CASE REPORT

A 14-year-old boy presented with generalized acute abdomen Pain which was non-radiating and colicky in nature with 5-6 episodes of bilious vomiting along with obstipation. On general physical examination his vital signs were temperature afebrile, pulse-90 bpm, Blood pressure-120/80 mmHg, SPO2- 99% on room atmosphere, respiratory rate 16 per minute. His abdomen was distended with generalized tenderness of abdomen with guarding in right hypochondrium and epigastrium, per rectal examination was normal. Hemoglobin, white blood cell count, basic biochemistry panel, and arterial blood gases were all within normal values. Plain radiographs suggested bowel obstruction with the localization of small intestinal loops predominantly on the right side. Chest radiography did not reveal any signs of perforation of a hollow viscus.MD-CT abdominal aortogram was done which showed- swirling of superior mesenteric vein around the superior mesenteric artery suggestive with significant compression of the SMV. With no free fluid/air in the abdomen or pelvis.

Operative details

Emergency exploratory laparotomy- LADDS Procedure done. Intra-operative findings small bowel volvulus with mesenteric venous collateralization. The duodenojejunal junction was in the right hemiabdomen with Ladd's bands coursing from the transverse colon to the right upper quadrant. The midgut was delivered into the operative field, and volvulus de-rotated through a counter-

clockwise rotation1800. No e/o bowel ischemia, however presence of engorged mesenteric vasculature was suggestive of a chronic or recurring midgut volvulus. Ladd's bands divided releasing transverse colon from its attachments to right lateral abdominal wall. Additional congenital bands divided; inversion appendectomy performed. bowel placed free of tension in the right hemiabdomen and the large bowel in the left hemiabdomen anatomically.

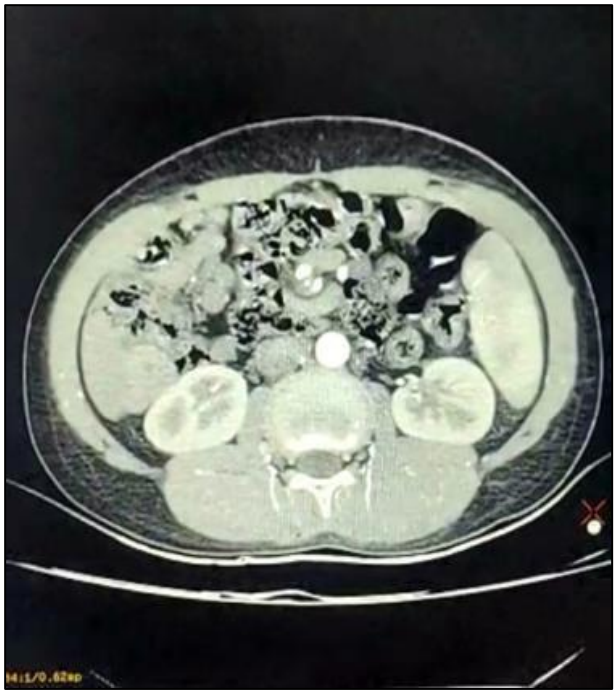

Figure 1: MT CT showing compression of SMV.

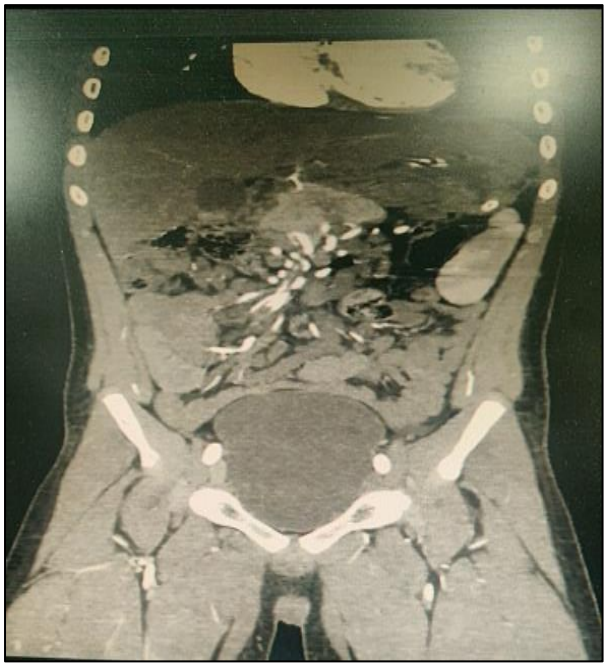

Figure 2: Abdominal aortogram showing swirling of SMV around SMA.

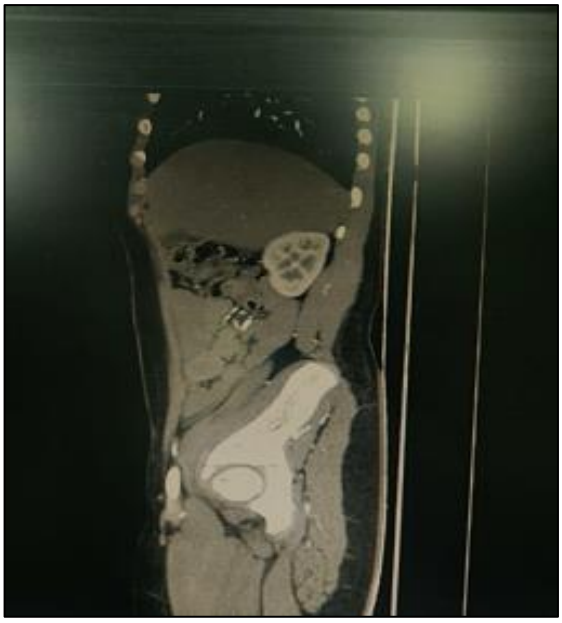

Figure 3: Localisation of bowl loops.

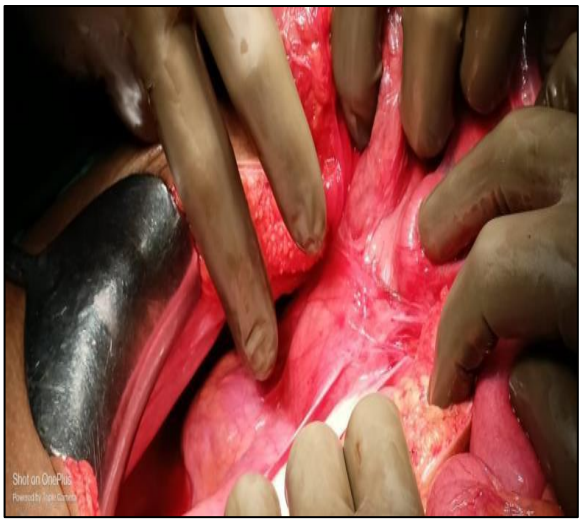

Figure 4: Congenital Ladds band.

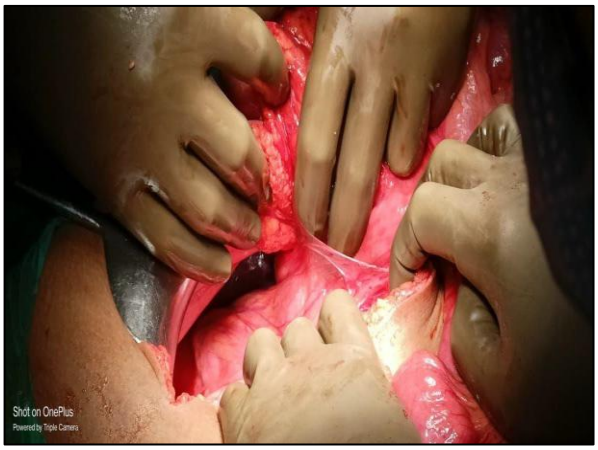

Figure 5: Separation of Ladds band.

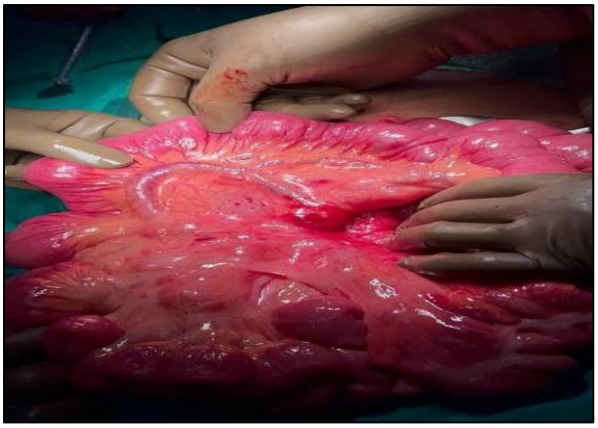

Figure 6: Malrotated venous collatralization.

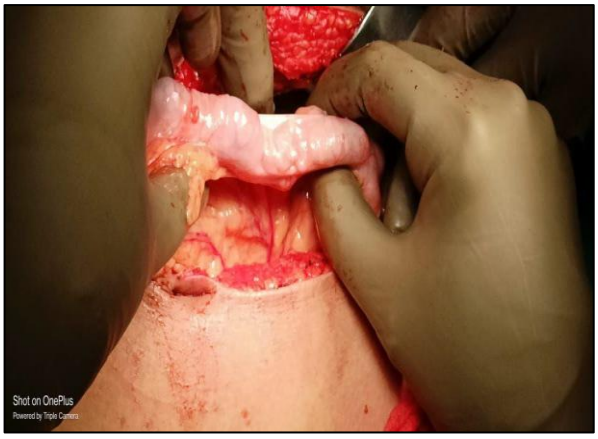

Figure 7: Non necrosed bowl loop.

Table 1: Reported cases of intestinal malrotation (13–19 years old).

Year	Author	Journal	Age	Gender	Symptoms	Surgery
1991	Ko et al	Jpn J Surg (in Japanese)	19	F	abdominal distention	Ladd procedure
1992	Lal et al	Indian J Gastroenterol	17	F	abdominal pain, vomiting	gastrojejunostomy, vagotomy
1994	Pelucio et al	Am J Emerg Med	15	M	abdominal pain	Ladd procedure
1997	Kimura et al	Jpn J Clin Surg (in Japanese)	16	M	vomiting	Ladd procedure
1997	Ishida et al	J Jpn Soc Pediatr Surg (in Japanese)	13	F	abdominal pain, vomiting	Ladd procedure
1997	Yahata et al	Surg Laparosc Endosc	17	F	abdominal pain	laparoscopic Ladd procedure
1998	Yokota et al	Kesennuma Hosp Medical J (in Japanese)	15	F	abdominal pain	Ladd procedure
1999	Kang et al	J Jpn Soc PediatrSurg (in Japanese)	16	M	abdominal pain, vomiting	Ladd procedure
1999	Yamashita et al	SurgEndosc	13	F	vomiting	laparoscopic Ladd procedure
2000	Walsh et al	J PediatrSurg	13	F	abdominal pain	laparoscopic Ladd procedure
2001	Horiba et al	J Jpn Clin Surg (in Japanese)	17	M	vomiting	Ladd procedure
2003	Tsumura et al	SurgEndosc	15	F	abdominal pain	laparoscopic Ladd procedure
2003	Singer et al	J Am Coll Surg	19	M	abdominal pain, vomiting	Ladd procedure
2004	Tseng et al	JBR-BTR	14	F	abdominal pain	Ladd procedure
2005	Sato et al	Hokkaido Surg J (in Japanese)	18	M	abdominal pain	release of ileus
2005	Kamiyama et al	Radiat Med	14	M	abdominal pain	Ladd procedure
2007	Vechvitvarakul et al	J PediatrSurg	13	M	abdominal pain, nausea, vomiting	Ladd procedure, appendectomy
2007	Kusuda et al	J Abdominal Emergency Medicine (in Japanese)	17	M	abdominal pain	Ladd procedure
2007	Draus et al	Am Surg	17	F	abdominal pain, nausea	laparoscopic Ladd procedure
2008	Duran et al	Turk J Gastroenterol	17	F	abdominal pain	division of adhesions

Continued.

Year	Author	Journal	Age	Gender	Symptoms	Surgery
2008	Uchida et al	J Pediatr Surg	13	F	vomiting	Bypass
2009	Fukushima et al	Jpn J Endosc Surg (in Japanese)	15	F	abdominal pain, distention	laparoscopic Ladd procedure
2009	Tazaki et al	J Abdominal Emergency Medicine (in Japanese)	14	M	abdominal pain, vomiting	release of ileus
2009	Shimodaira et al	J of Jpn Soc Psychosomatic Med (in Japanese)	17	M	vomiting	laparoscopic Ladd procedure
2009	Fujii et al	J Jpn Clin Surg (in Japanese)	14	M	vomiting	Ladd procedure
2009	Mano et al	J Jpn Soc Pediatr Surg (in Japanese)	18	M	abdominal pain	laparoscopic Ladd procedure
2010	Watanabe et al	J Jpn Soc Gastrointestinal Dis (in Japanese)	19	F	abdominal pain	release of ileus
2010	Takazawa et al	Jpn J Pediatr Surg Nutr (in Japanese)	14	M	vomiting, distention	resection of necrotic intestine
2011	Kokado et al	J Jpn Soc PediatrSurg (in Japanese)	13	F	abdominal pain, vomiting	fixation of colon
2011	Lam et al	J Pediatr Surg	14	M	abdominal pain, vomiting	resection of necrotic intestine
2012	Nath et al	Ann R Coll Engl	16	M	abdominal pain	laparoscopic Ladd procedure
2012	Jain et al	Case Rep Radiol	15	M	abdominal pain	Ladd procedure
2012	Wanjari et al	N Am J Med Sci	17	M	abdominal pain, vomiting	laparoscopic Ladd procedure
2012	Macedo et al	Einstein	13	F	abdominal pain	laparoscopic Ladd procedure
2012	Tran et al	J Pediatr Surg	18	M	abdominal pain	Ladd procedure
2012	Katsura et al	J Jpn Clin Surg (in Japanese)	19	F	abdominal pain	resection of necrotic intestine
2013	Nakajima et al	present case	17	M	abdominal pain, vomiting	laparoscopic Ladd procedure

DISCUSSION

Malrotation of the intestinal tract is a congenital anomaly referring to either lack of or incomplete rotation of the fetal intestines around the axis of the superior mesenteric artery during fetal development. The malrotation of the gut and abnormal location of the cecum produces a narrow superior mesenteric vascular pedicle, as opposed to the normally broad-based small bowel mesentery. This narrow superior mesenteric artery takeoff and lack of posterior peritoneal fusion predispose the patient to subsequent midgut volvulus and obstruction with potential vascular catastrophe. Approximately 85% of malrotation cases present in the first two weeks of life.[1,2] However, presentation of intestinal malrotation is very rare and its incidence has been reported to be between 0.2% and 0.5%.[3] True incidence of malrotation in older children or adults is unclear, because a number of patients may be asymptomatic. Not all patients with malrotation present with symptoms. Even once the anomaly is discovered, many live without complaint.

In adults or older children, the difficulty of diagnosis results from both the absence of specific physical findings and the low frequency in adults.[4,5] Midgut malrotation in adults presents in numerous ways and the symptoms are non-specific. There are no typical sets of symptoms that are diagnostic of clinical problems. Symptoms in the adult patient are often mistaken for irritable bowel syndrome, peptic ulcer disease, biliary and pancreatic disease, and psychiatric disorders.[4]

The location of the pain may vary from the epigastric region to the left upper abdominal quadrant, and the pain may be described as either intermittent cramping or persistent aching. It most often occurs postprandially and may last several minutes to an hour. Our patient had experienced abdominal distension, nausea, vomiting, and vague abdominal pain several times before, but the symptoms had always disappeared spontaneously. Frequently, the plain radiograph is normal or may show an incomplete bowel obstruction. Specific findings that are diagnostic of malrotation can be detected through the

use of both upper and lower gastrointestinal tract barium studies, angiography of the superior mesenteric artery, CT scan, and often emergency laparotomy. Occasionally, an abdominal radiograph will show dilated bowel loops with the orientation of a spiral nebula in the midabdomen.

Barium studies may reveal a dilated duodenal loop caused by bowel obstruction with a spiral configuration of the proximal jejunal loops. CT is also used to investigate small-bowel volvulus and various signs have been described. Characteristic findings include the positioning of the superior mesenteric vein lying to the left or anterior to the artery because of torsion of the mesentery around its attachment, the presence of a right-sided duodeno-jejunal junction, the absence of a cecal gas shadow on the patient's right side, or third and fourth duodenal junction that does not cross the patient's spine.[5,6]

Management of intestinal rotation without midgut volvulus is controversial. In general, symptomatic patients with malrotation should be treated with surgical intervention. The classic treatment for incomplete intestinal rotation is the Ladd procedure, which requires mobilization of the right colon and cecum by division of Ladd bands, mobilization of the duodenum, division of adhesions around the superior mesenteric artery to broaden the mesenteric base, and an appendectomy.[7,8]

Spigland et al, recommended that all patients with malrotation are candidates for laparotomy, even if they are asymptomatic.[9] Mozziotti et al, recently reported a series of malrotation patients managed successfully with laparoscopic intervention.[10] Laparoscopy can be used to determine the position of the Treitz ligament and whether the cecum is fixed in the right lower quadrant. If the patient is decided to be at risk for volvulus (i.e. a shortened mesenteric pedicle), a Ladd's procedure can be accomplished laparoscopically with good long-term results.[10,11] Due to the abnormal cecal position inflicted by malrotation, patients with associated appendicitis will demonstrate atypical symptoms with pain projected to the left of the middle line since the appendix will not be located in the normal area in the abdomen. This could lead to confusion and delay in diagnosing appendicitis in the future. Therefore, appendectomy is usually performed during surgical intervention.

Although most of the literature consists of occasional case reports or small case series, we searched for literature published between 1983 and 2012 using PubMed and Web Japan Medical Abstracts Society and found 37 reported cases of teenage patients (ages 13 through 19) with intestinal malrotation (Table 1). Twenty patients were male and seventeen were female. The diagnosis could be made by radiographic studies in all these patients. Patients presented with a variety of gastrointestinal disorders. Abdominal pain was the most frequent symptom (30/37). Other symptoms were nausea, feeding intolerance, reflux, and respiratory problems. The Ladd procedure was performed on 27 patients on 12 patients the procedure was conducted laparoscopically.

An important point is that since many patients with intestinal malrotation are asymptomatic, everyone in the medical community should be made aware of the problem. Also, patients with acute volvulus should be treated promptly. Some asymptomatic adults may not need surgery. Of note, there is always the possibility that laparoscopic surgery will not entirely rule out the chance of acute volvulus; it could introduce problems such as band adhesion and future adhesive small bowel obstruction

CONCLUSION

While symptomatic congenital bowel malrotation has been traditionally thought of as a disease of infancy, this case illustrates that it must also be considered as a part of the differential diagnosis of abdominal pain in older children and adults. Given the lower degree of suspicion for this diagnosis in these populations, delays in diagnosis may result in increased morbidity as intestinal necrosis can result from volvulus secondary to bowel malrotation, and the time to surgical intervention is crucial in preventing this complication. This case further illustrates that uncommon etiologies for a common chief complaint must be considered when a patient presents on multiple occasions despite an unremarkable initial evaluation.

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