

CASE REPORT

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Intestinal duplication in an adult presenting as acute small bowel obstruction with malrotation and volvulus: Case report and updated review of the literature

Maria Eugenia Gomez, Marie Nunez Duarte, Rebecca Silver, Alayna Dieter

ABSTRACT

Introduction: Intestinal malrotation and duplication are rare congenital anomalies that typically present in childhood, but can also occur in adults. Adult presentations are often associated with acute obstructive symptoms, and complications like volvulus and ischemia can rapidly lead to life-threatening outcomes. Early recognition and intervention are essential for ensuring favorable outcomes.

Case Report: We report an interesting case of intestinal duplication and malrotation in a 49-year-old male who presented with acute abdominal pain, bilious vomiting, and bloating. Imaging revealed a twisted intestinal segment, initially mimicking a large Meckel's diverticulum. Surgical exploration uncovered an ischemic segment of ileum with complex torsion and gangrene, necessitating resection and anastomosis.

Conclusion: This case highlights the rarity of intestinal malrotation presenting in adulthood, and the severe complications that can result. The rapid progression to ischemia and necrosis emphasizes the need for timely surgical intervention. While Ladd's procedure is often the treatment of choice, it is not feasible when bowel viability is compromised, as in this case. Adult presentations of intestinal duplication with volvulus are rare, but should be considered in patients with acute obstructive symptoms.

Keywords: Adult congenital anomalies, Intestinal duplication, Intestinal malrotation, Ischemic bowel, Small bowel obstruction, Surgical resection, Volvulus

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INTRODUCTION

Intestinal duplications are cystic or tubular structures that develop with a shared common blood supply found in 1 per 4500 live births [1]. Previously, duplications were known as giant diverticula, enteric cysts, intestinal duplex, or a variation of Meckel diverticulum until the term “intestinal duplication” was popularized in the 1930s [2]. The most common site where they may be found is the ileum and jejunum, then the esophagus, stomach, and colon. Intestinal malrotation in adults has been estimated to have an incidence of 0.2–1% [3].

Males make up 60–80% of cases of intestinal duplication and a third may have associated congenital anomalies. The majority of cases present before the age of 2 years old, but many remain asymptomatic [4]. Presentations include bleeding, chronic pain, or perforation if the duplication contains heterotopic mucosa.

While the cause of intestinal duplications remain unclear, theories include a failure in recanalization, a vascular insult, persistence of embryonic diverticula, and partial twinning. Currently, surgical resection is still the mainstay of treatment. The results form a systematic search of the literature is shown in Table 1. We aim to

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compare our patient's clinical data and associated previous studies to further elucidate the diagnosis and potential treatment options.

CASE REPORT

A 49-year-old male patient presented to urgent care 4 hours following the onset of abdominal pain severe enough to awaken him from sleep. He also reported bilious vomiting and bloating.

A computed tomography (CT) of the abdomen and pelvis was obtained, showed dilated thin loops of bowel in the meso- and hypogastrum, full of liquid, and with a thickened wall surrounding engorged mesenteric vessels, forming the swirl sign, compatible with small bowel obstruction and volvulus (Figures 1 and 2).

A supra/infraumbilical median laparotomy was performed. During surgical exploration, an ischemic segment of the ileum was appreciated arising adjacent



Figure 3: Gross pathology (above) notable for a torsioned, necrotic, and adherent bowel.



Figure 1: Abdominal free fluid, diffusely dilated and edematous small bowel loops in the meso and hypogastrum.

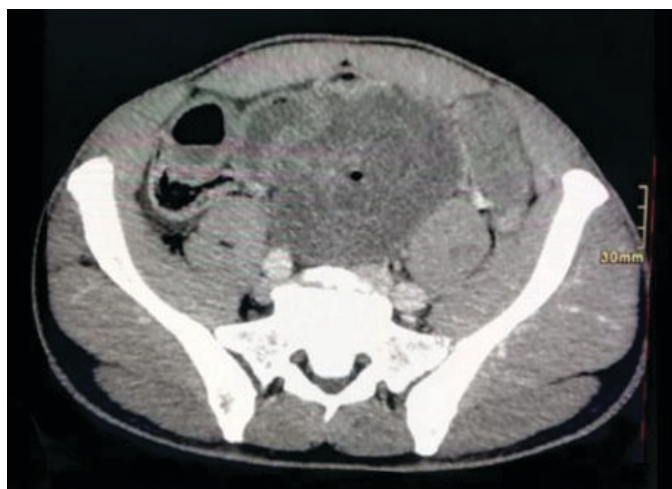


Figure 2: Dilated small bowel loops forming "swirl sign" surrounding mesenteric vessels suggesting small bowel obstruction and possible ileal volvulus.

to the ileocecal valve, initially giving the appearance of a large Meckel's diverticulum. This segment of small bowel included the appendix and was herniated through an internal hernia orifice in the ileal mesentery, with complex torsion and gangrene, but no perforation (Figure 3).

Devolvulation in a counterclockwise direction followed, then dissection and section with the Voyant sealing device. The ascending colon was exposed via the Toldt's fascia. The healthy bowel loops were connected with the proximal end 3 meters from the ligament of Treitz and the distal end at the cecal pole. A linear anastomosis with gastrointestinal anastomosis (GIA) was completed and reinforced with stitches on the anterior surface of the anastomosis. Endoluminal hemorrhage was appreciated in the specimen that was sent for frozen section. An intraoperative diagnosis of intestinal duplication, obstruction, and volvulus was made. In total, the operative time was 1 hour.

The specimen was found to be a 75 cm long intestinal segment with transmural, coagulative necrosis, without evidence of malignancy or epithelial dysplasia. The cecal appendix measured 1.5 cm and was without histological lesions. The perivisceral adipose tissue also showed mixed ischemic and hemorrhagic necrosis. Lymph nodes were without injury.

Postoperatively, the patient spent less than 24 hours in the intensive care unit (ICU). After transferring to the floor, he had normoactive bowel sounds and was started on early oral feeds. On postoperative day (POD) 2 he was found to have mild abdominal distention, bright red rectal hemorrhage mixed with melena. His vitals remained stable. On POD 3, a CT angiogram was ordered, which was negative for active bleeding, with an intact anastomosis. Hemoglobin trended upward without the need for transfusion. Postoperative ileus was also appreciated and resolved without intervention. The patient was discharged on POD 7.

Table 1: Summary of adult patients with small intestinal duplications as found in the United States National Institute of Health National Library of Medicine (PubMed) database as of May 13, 2024

Database	Search strategies				Papers found	Related papers	
MEDLINE (via PubMed)	("Adult"[Mesh] AND "Intestine, Small"[Mesh]) AND ("Congenital Abnormalities"[Mesh]) AND duplication [Title/Abstract]				40	20	
Study (year)	Age (years) /gender	Presenting features	Operative management	Form	Size (cm)	Malignancy	Outcomes
Ho (2012) [12]	25/male	Abdominal pain	Resection	Tubular	NR	No	Required stoma
Vivier et al. (2013) [13]	32/female	Routine control examination	Resection	Cystic	2	No	No complications
Handra-Luca (2013) [14]	65/male	Routine control examination	Resection	Tubular	8	Yes	No complications, found incidentally during adenoma resection
Barbosa et al. (2015) [5]	36/male	Abdominal pain, vomiting	Resection	Cystic	NR*	No	No complications
Yu et al. (2014) [22]	24/female	Abdominal pain	Resection	Cystic	28 × 20	No	No complications
Li et al. (2013) [15]	25/male	Abdominal pain, weight loss	Resection	Tubular	15.5 × 4	No	No complications
Nussbaum et al. (2014) [16]	51/male	Abdominal pain, bloody stools	Resection	Tubular	9.5 × 2	Yes	No surgical complications, adenocarcinoma
Kim et al. (2014) [17]	19/female	Abdominal pain	Resection	Cystic	2.5	No	No complications
Park et al. (2014) [18]	36/female	Abdominal pain	Resection	Cystic	12	No	No complications
Martínez-Alcalá et al. (2014) [19]	37/female	Abdominal pain, vomiting	Resection	Cystic	5	No	No complications
Furuya et al. (2012) [23]	70/female	Abdominal pain	Resection	Cystic	3	Yes	Wound infection, also GIST
Miloudi et al. (2012) [24]	26/male	Abdominal pain, vomiting	Resection	Cystic	4	No	No complications
Palacios et al. (2013) [25]	35/female	Routine control examination	Resection	Cystic	1.3	No	No complications
Antaki et al. (2013) [6]	21/male	Abdominal pain	Excision	Cystic	NR*	No	No complications
Kusnierz et al. (2014) [20]	31/female	Abdominal pain, nausea, lack of appetite	Resection	Cystic	2.9 × 2.6	No	No complications
Cheng et al. (2014) [10]	49/male	Gastrointestinal bleeding, melena passage	Endoscopic	Cystic	NR*	No	No complications
Kurien et al. (2014) [11]	NR/female	Abdominal pain	Endoscopic	Cystic	NR*	No	No complications
Kachi et al. (2016) [26]	64/female	Abdominal pain	Resection	Cystic	4	No	No complications
Gupta et al. (2016) [21]	20/male	Constipation, vomiting	Resection	Cystic	NR*	No	Required stoma due to significant bowel distension. Reversed after 3 months.

Table 1: (Continued)

Database	Search strategies				Papers found	Related papers	
Inoue et al. (2016) [27]	56/male	Appetite loss, palpitations, orthostatic syncope, hematochezia	Resection	Cystic	NR*	No	No complications
Huang et al. (2018) [8]	53/male	Abdominal pain, constipation	Resection	Cystic	4-5	No	No complications
D'Souza et al. (2018) [9]	56/male	Abdominal pain, vomiting	Resection	Cystic/Tubular	6 tubular, 1 cystic	No	No complications
Schils et al. (2018) [28]	36/male	Abdominal pain, vomiting	Resection	Tubular	NR*	No	No complications

NR: Not reported.

DISCUSSION

This is a rare presentation of intestinal malrotation in an adult with only 23 cases reported in a systematic review on the subject as of 2018. It is quite rare for intestinal malrotations to remain asymptomatic until adulthood; most present with obstruction or bleeding before the age of two [5, 6]. Our case was unique due to the size of the defect, rapid progression from the onset of symptoms to ischemia and necrosis of the small intestine.

Adults with congenital intestinal malrotation may present with an acute abdomen and even strangulation or necrosis [7]. Ladd's procedure allows for the broadening of the mesentery and lysis of Ladd's bands, positioning the bowel in a non-rotated position. A non-rotated position is one where the small bowel is on the right and the large bowel is on the left. However, this procedure can only be performed if the intestinal segment remains viable. Surgical resection and anastomosis, as in our case and as reported in 17 others, remains the cornerstone of management due to tissue necrosis and concerns for malignancy [5, 8, 9]. Two cases of duodenal duplication were managed endoscopically; one with hemoclippping and another with drainage [10, 11].

Surgical management leads to exceptional outcomes in the majority of cases. Seventeen of the patients described in the literature were discharged within days of their surgery without major complications, similarly to our case, and with relatively no impact on their daily functioning [6, 8, 10, 12–20]. Two patients required a stoma due to extensive tissue necrosis, although one was able to be reversed three months later [12, 21]. This is likely due to the fact that most patients were largely in good health prior to developing abdominal issues, and over half were under the age of 40.

CONCLUSION

A high index of suspicion must be maintained for intestinal duplication in the acute development of

obstructive symptoms for adult patients who fit the clinical picture and, in particular, patients with associated congenital conditions.

Our case was unique because of the length of torsioned intestine and rapid progression to ischemia and necrosis. While there is no current standard of practice regarding adult congenital intestinal duplication with volvulus, multiple cases across the literature report excellent outcomes with the Ladd's procedure. However, this procedure can only take place if the segment of bowel remains viable. Additional consensus is required regarding the approach to these types of cases to enable early identification of volvulus. The differential for small bowel obstruction in an adult should include congenital malrotation leading to volvulus, as demonstrated in the cases presented. One must maintain a high index of suspicion for malrotation and minimize time to operation in order to preserve the viability of the small intestine and nutritional status of the patient.

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Author Contributions

Maria Eugenia Gomez – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Marie Nunez Duarte – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Rebecca Silver – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Alayna Dieter – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

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Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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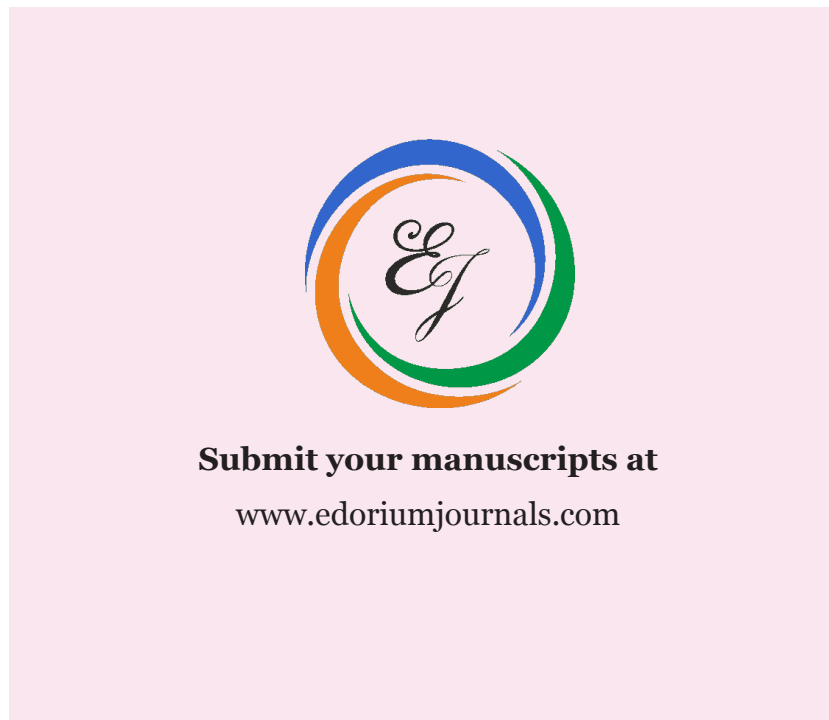
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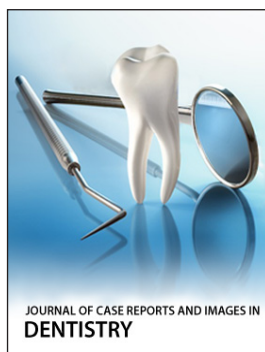
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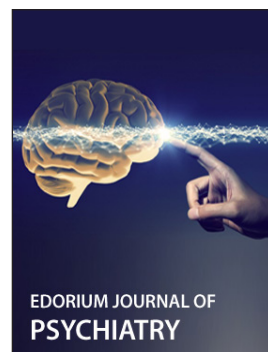
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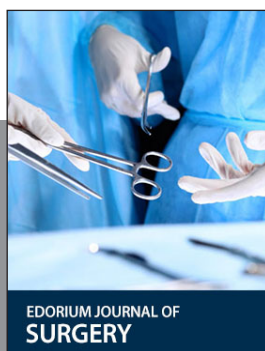
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