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Dorsal Mesenteric Agenesis without Small Bowel Atresia: A Rare Pediatric Case Insight

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Abstract: Dorsal mesentery agenesis is often associated with jejunal atresia and these patients present with symptoms in early neonatal life. However, very few reports were found on the agenesis of the dorsal mesentery without small bowel atresia. Here we report an 8-month-old child with mesenteric agenesis without bowel atresia presented as intestinal obstruction due to twisting of the gut along with internal herniation where a single marginal vessel was the key supply to the whole small bowel.

Keywords: Dorsal mesenteric agenesis, Intestinal obstruction, rare cause, RMCH, Single marginal artery, Without jejunal atresia.

Special Case Report

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Article at a glance:

Study Purpose: *To report a rare case of dorsal mesenteric agenesis without small bowel atresia causing intestinal obstruction.*

Key findings: *Dorsal mesenteric agenesis without small bowel atresia is a rare condition that may present with intestinal obstruction in children. Only a few reports exist in the English literature with agenesis of the dorsal mesentery without small bowel atresia. In dorsal mesenteric agenesis without small bowel atresia, the small bowel is supplied by a single marginal artery, which may be the only blood supply. Careful preservation of this vessel is necessary to avoid catastrophic bowel death.*

Newer findings: *This case adds to the understanding of mesenteric agenesis without jejunal atresia, highlighting the role of a single marginal artery in blood supply.*

Abbreviations: *DMA – Dorsal Mesenteric Agenesis, SBA – Small Bowel Atresia, IA – Internal Herniation, SMV – Superior Mesenteric Vessel.*

INTRODUCTION

Intestinal obstruction in children is a prevalent surgical emergency that encompasses various underlying causes, ranging from congenital anomalies to acquired conditions. Among the congenital factors, small bowel atresia is a well-recognized etiology, often resulting from vascular events or developmental defects during gestation.[1,2] However, dorsal mesenteric agenesis, an infrequent congenital abnormality characterized by the absence of the supporting mesentery for the small intestines without the actual atresia of the bowel, remains an uncommon and less comprehended cause of intestinal obstruction in the pediatric population. Dorsal mesenteric agenesis gives rise to an abnormal fixation of the intestines, which can predispose to volvulus, internal hernias, and subsequent intestinal obstruction, thereby

presenting significant challenges in diagnosis and management1. In such cases, the absence of small bowel atresia further complicates the clinical presentation, as the typical radiographic and intraoperative findings associated with atresia are absent, often leading to delays in diagnosis and appropriate intervention. The rarity of dorsal mesenteric agenesis without small bowel atresia as a cause of intestinal obstruction highlights the importance of considering a broad range of possibilities in pediatric patients presenting with symptoms of intestinal obstruction. This case report aims to emphasize the clinical presentation, diagnostic complexities, and management strategies related to dorsal mesenteric agenesis without small bowel atresia, thus contributing to the limited literature available on this uncommon condition and emphasizing the necessity for clinicians to be aware to enhance outcomes in affected children. Through the presentation of this case, we aim to underscore the significance of thorough diagnostic evaluation in pediatric intestinal obstruction, the role of imaging in identifying uncommon causes of obstruction, and the surgical considerations required to manage such rare anomalies. This case contributes to the knowledge essential for pediatric surgeons, radiologists, and pediatricians to recognize and effectively manage this rare cause of intestinal obstruction, potentially resulting in earlier diagnosis and improved patient outcomes.

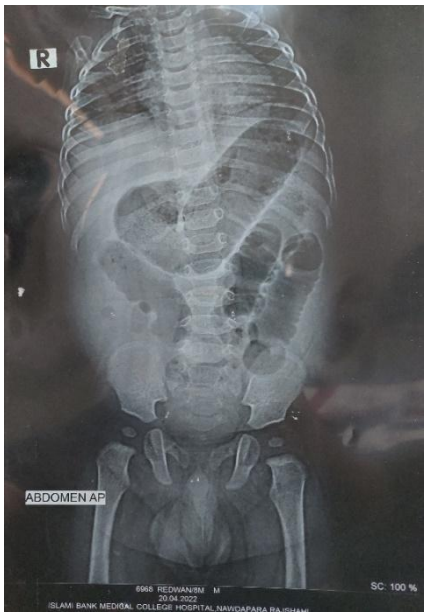

Figure 1: Plain X-Ray of The Abdomen Showing Multiple Dilated Bowel Loops Suggesting Intestinal Obstruction

CASE REPORT

An 8-month-old male Asian boy was admitted to the Department of Pediatric Surgery unit of RMCH via the emergency department with a 3-day history of vomiting, obstipation, abdominal pain, and distension. On examination, he was moderately dehydrated; his abdomen was distended and diffusely tender. There were no bowel sounds, and no intra-abdominal masses were palpable. A plain abdominal X-ray showed dilated loops of the small bowel with multiple air-fluid levels suggestive of intestinal obstruction (Fig. 1). Primary diagnosis was intussusception. Still, the ultrasonogram showed no evidence of intussusception. The patient was resuscitated and taken to the operation theatre for a diagnostic exploratory laparotomy. During the operation, the small bowel loops were found internally herniated and twisted around the superior mesenteric axis. Enterotomy was needed to decompress the gut and to reduce internal hernia. We also noticed that the entire small bowel from the DJ flexure to the terminal Ileum received its blood supply from a large single marginal vessel arising from the superior mesenteric artery, and there was a total absence of the small bowel mesentery. The ileocolic and right colic arteries were absent (Fig. 2). On inspection, the fourth part of the duodenum was

found to be normally fixed with the ligament of Treitz, and the ileocecal valve was in its normal anatomical position with a floating caecum. The edges of the Peritoneal fold along the marginal vessels were approximated to close the defect, and the small bowel was fixed to maintain its normal anatomy. The patient recovered expectantly postoperatively, a normal diet was started on the 5th postoperative day and discharged on the 7th postoperative day.

DISCUSSION

The development of the mesentery starts when the foregut, midgut, and hindgut are in broad contact with the mesenchyme of the posterior abdominal wall.[3] In the 8-mm embryo, the connecting tissue bridge becomes narrow, and the caudal part of the foregut, midgut, and major part of the hindgut is suspended from the abdominal wall by the so-called dorsal mesentery.[4] A ventral mesentery only exists in the terminal part of the esophagus, the stomach, and the upper part of the duodenum. The ventral mesentery gives rise to the lesser omentum, the falciform ligament, and the visceral peritoneum of the liver. The dorsal mesentery extends from the esophagus's lower end to the cloacal region. In the area of the stomach, it is known as the dorsal mesogastrium or greater omentum; in the area of the duodenum, as the dorsal mesoduodenum; and in the region of the colon, as the dorsal mesocolon. The dorsal mesentery of the jejunal and ileal loops is the mesentery proper. It is well known that mesenteric defects can lead to intestinal volvulus even when the midgut is normally rotated.[5] Not less than 3608 reported cases of volvulus exist because of an internal herniation through the mesenteric defect, which in turn, causes intestinal obstruction.[1] The mesenteric defects range from lesions involving a short, clearly defined area of the small intestine (segmental defects) to lesions involving the entire base of the mesentery (basilar defects). Agenesis of the dorsal mesentery is usually associated with a high jejunal atresia. In this condition, the primary superior mesenteric arterial branches to the jejunum and Ileum are absent, with an absent dorsal mesentery and the distal small bowel suspended from a vascular stalk.[2] The first description of an isolated agenesis of the dorsal mesentery was in 1960. A case of intestinal obstruction in a newborn caused by intestinal herniation through a mesenteric defect, with marginal vessels to the terminal Ileum around the free edge of the defect, was reported.[1,2]

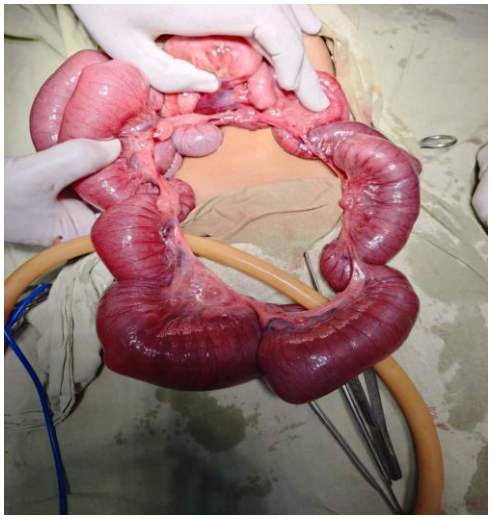

Figure 2: On Untwisting, It Reveals Complete Agenesis of The Dorsal Mesentery

Fifteen years later, a case report described a 2 1/2–year old child with agenesis of the dorsal mesentery and the apple peel deformity but without small bowel atresia.[2] The mesentery was absent from the superior mesenteric artery to the ileocolic vessels, and the blood supply to the small bowel was from a marginal vessel. Our patient may be similar to the second reported case of agenesis of the dorsal mesentery without jejunal atresia or apple peel deformity. Another similar case was reported In the Journal of Pediatric Surgery in 2006.[1] It is believed that isolated small bowel atresia, jejunal atresia with agenesis of the dorsal mesentery, and isolated mesenteric

defects/agenesis are all variants of an in-utero vascular accident involving various degrees of the superior mesenteric artery.[3, 6-8] In our case, the absence of intestinal atresia makes an intrauterine vascular accident an unlikely cause. Intestinal volvulus without malrotation is rare; it is not as easily recognizable as volvulus with malrotation and is associated with a high mortality rate. Knowledge of mesenteric defects and agenesis as a possible cause of midgut volvulus in infants and children is essential for early recognition and treatment of this potentially lethal condition.

Summary

Agenesis of the dorsal mesentery but without small-bowel atresia can present beyond the neonatal period. Here, the small bowel is supplied by a single marginal artery, which may be the only blood supply, so careful preservation of this vessel is necessary to avoid catastrophic bowel death.

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