

Case report

Jejunioleal atresia with umbilical entrapment of distal bowel: a rare embryologic insight and surgical challenge

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Abstract

Jejunioleal atresia is a common cause of neonatal intestinal obstruction with various etiologies. We report a rare case of type IIIa jejunioleal atresia in which the distal bowel was entrapped within the umbilicus, suggesting failed midgut return during embryogenesis as a possible alternative pathogenic mechanism. A term male newborn presented with progressive abdominal distention and failure to pass meconium within the first 48 hours of life. Imaging studies were consistent with jejunioleal atresia. Intraoperatively, the distal small bowel was found entrapped within the umbilical cord, and a blind-ending proximal dilated bowel confirmed type IIIa atresia. Resection with end-to-end anastomosis was performed successfully. The patient remained well at the 4-year follow-up. This rare presentation suggests a possible embryologic mechanism of jejunioleal atresia. Caution is advised when evaluating abnormal umbilical cord features in neonates to avoid injury to underlying structures. Despite these unusual findings, bowel resection with primary anastomosis remains the standard treatment, while meticulous assessment of the umbilical cord is essential to prevent iatrogenic intestinal injury.

Keywords: Iatrogenic injury during umbilical cord clamping, jejunioleal atresia, umbilical entrapment

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Received: March 29, 2026

Revised: May 1, 2026

Accepted: June 6, 2026

Jejunioileal atresia is a relatively common cause of neonatal intestinal obstruction, typically presenting within the first days of life. The Grosfeld classification divides jejunioileal atresia into four types: type I, mucosal and submucosal atresia with intact serosa and mesentery; type II, complete discontinuity of the bowel with a fibrous cord connecting the proximal and distal ends, and an intact mesentery; type IIIa, complete discontinuity without a fibrous connection, accompanied by a mesenteric defect; type IIIb (“apple-peel” or “Christmas tree” atresia), a proximal atresia with absence of a major segment of mesentery, where the distal bowel coils around a single arterial vessel. Several hypotheses have been proposed to explain its etiology. Among these, intrauterine vascular insult is the most widely accepted mechanism. However, variations in anatomical presentation suggest that multiple mechanisms may contribute to its development.

We present a rare case of type IIIa jejunioileal atresia in which the distal small bowel was found entrapped within the umbilicus. This unique intraoperative finding may support the hypothesis of failed midgut return during embryonic development, with subsequent entrapment and ischemia occurring during abdominal wall closure. To our knowledge, this

presentation has rarely been reported in the literature. Relevant previously reported cases are also reviewed.

Case report

A term male infant was born at 38+4 weeks’ gestation via normal labour, with a birth weight of 2,730 g. APGAR scores were 9, 10, and 10 at 1, 5, and 10 min, respectively. He was born at a rural hospital and the pregnancy was considered low risk; therefore, no prenatal ultrasonography was performed.

The patient failed to pass meconium within the first 48 hours of life and presented with signs of early neonatal sepsis and progressive abdominal distention. Physical examination revealed abdominal distension, no signs of ascites, a patent anus, and whitish meconium on rectal irrigation. The umbilical cord measured 1.2 cm in diameter, and an umbilical rubber band had been applied 2 cm above the umbilical ring. Plain abdominal radiography (**Figure 1**) demonstrated bowel dilatation with an absence of rectal gas. He was referred to our hospital at 3 days of age. Initially, the umbilical lesion was not recognized. Jejunioileal atresia and total colonic aganglionosis were suspected. Abdominal distension did not improve after 48 hours of daily rectal irrigation.



Figure 1. Plain abdominal radiograph demonstrating bowel dilatation with absence of rectal gas.



Figure 2. Lower GI contrast study showing a small-caliber colon extending proximally to the distal small bowel and ending in the mid-abdomen.

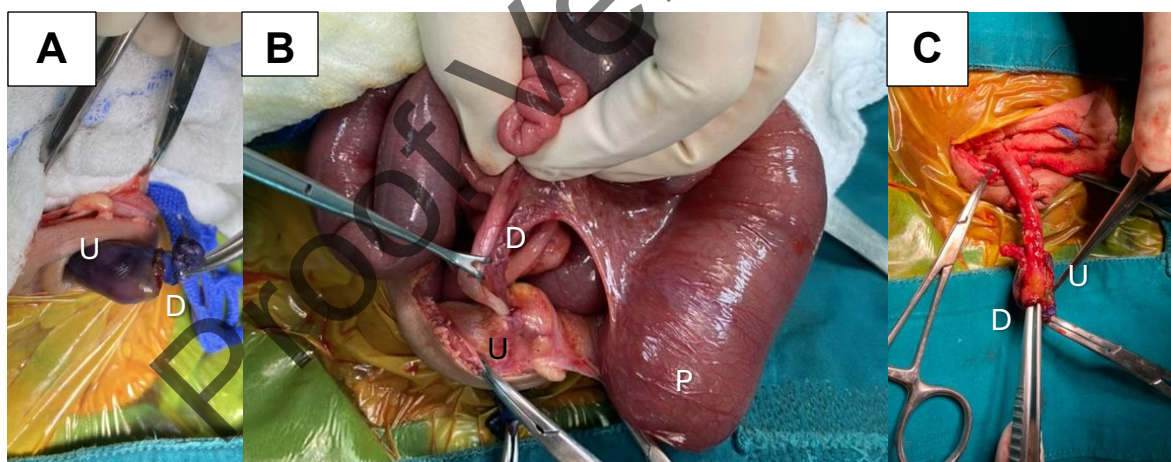


Figure 3. (A) Distal bowel injury with entrapment in the umbilicus; (B) blind-ending proximal jejunum and distal small bowel entrapped within the umbilical cord; (C) confirmation of distal bowel injury caused by the umbilical clamp. U, umbilicus, (D), distal small bowel, P, blind-ending proximal jejunum.

Investigations: A lower gastrointestinal (GI) contrast study was performed (**Figure 2**), showing a small-caliber colon extending proximally to the distal small bowel and terminating at the mid-abdominal level. These findings were suggestive of jejunoileal atresia, while total colonic aganglionosis remained in the differential diagnosis.

Surgery was performed on day 6 of life after referral, initial stabilization, serial rectal irrigations, and radiologic evaluation to differentiate jejunoileal atresia

from total colonic aganglionosis. However, worsening sepsis and persistent abdominal distension prompted exploratory laparotomy, which revealed 140 cm of severely dilated proximal small bowel, with a maximum diameter of 3 cm, ending in a blind pouch. In addition, a 25-cm segment of narrow-caliber, unused terminal ileum was entrapped within the umbilical area, and no portion of the colon was involved, as shown in **Figure 3**. The blind end of the distal ileum was injured by the rubber band. No remaining umbilical cord was

available for examination because the cord had been cut early at the rural hospital. The mesenteric defect measured 5 cm. The duodenojejunal (DJ) junction was located in the left upper quadrant and the ileocecal (IC) valve in the right upper quadrant, with a narrow mesenteric base measuring approximately 2 cm. These findings were consistent with atypical malrotation associated with type IIIa jejunoileal atresia.

The distal small bowel was released from the umbilical entrapment by incising around the umbilical ring. Normal saline was then used to irrigate the distal bowel to confirm the absence of additional distal atresia. To minimize the size discrepancy for anastomosis, the distal end was incised obliquely, and approximately 5 cm of the dilated proximal segment was resected. A primary end-to-end anastomosis was performed. The remaining small bowel length was 160 cm, with a patent colon and preserved ileocecal valve. Postoperative course: A minor anastomotic leak was noted on postoperative day 5 and managed conservatively. Enteral feeding was initiated on postoperative day 8 and advanced to full feeds by day 15.

At 4 years of age, the patient was clinically well, with weight at the 3rd percentile and height at the 25th percentile for age.

Discussion

Jejunoileal atresia is a common cause of neonatal intestinal obstruction, with an incidence of 1 in 1,000 to 1 in 5,000 live births. An intrauterine vascular insult remains the most widely accepted etiologic mechanism, resulting in ischemic necrosis and resorption of the affected bowel segment. However, not all cases can be explained by this theory, particularly those associated with atypical fixation or malrotation, suggesting that embryologic factors may also contribute.

In our patient, the distal small bowel was entrapped within the umbilical cord and was accidentally injured by umbilical cord clamping. The unusual finding may suggest that part of the midgut loop failed to return completely to the abdominal cavity during the physiological herniation phase (6th – 10th weeks of gestation),⁽¹⁾ resulting in persistent extra-abdominal entrapment, compression, or ischemia of the involved small bowel, which may provide embryologic support for a non-vascular, mechanical origin of jejunoileal atresia.

Comparable cases have rarely been reported in the literature.^(2–4) In these cases, the distal bowel was

found attached to the umbilical ring, with a blind-ending proximal intestine compatible with type IIIa jejunoileal atresia. They proposed that the bowel entrapment at the umbilical ring resulted from failure of the elongated bowel to return to the abdominal cavity, possibly in association with remnants of the omphalomesenteric duct. From a diagnostic perspective, the dilated intestine and absence of distal bowel gas on plain abdominal radiography supported a provisional diagnosis of jejunoileal atresia. A lower GI contrast study was also performed because total colonic aganglionosis was considered in the differential diagnosis.

Our case demonstrated viable distal bowel entrapped within the umbilical cord, together with iatrogenic bowel injury caused by umbilical cord clamping. Although the jejunoileal atresia itself may have resulted from failed midgut return and entrapment, the clamp-related bowel injury likely contributed to the patient's sepsis. Although rare, small-bowel injury due to umbilical cord clamping has been reported,^(5–7) most often in cases of hernia of the umbilical cord. Nonetheless, our case highlights the risk even in the absence of obvious herniation. The umbilical cord clamp should therefore be applied with caution, especially when the cord appears abnormal, such as when it is bulging or discolored.

Differentiating between true herniation into the umbilical cord and failure of midgut return can be challenging. In our case, distal bowel entrapment in the absence of a typical hernia sac, together with associated malrotation findings, may favor incomplete midgut return rather than post-return herniation; however, definitive distinction cannot be made.

Despite the unexpected anatomy, standard treatment remained resection of the atretic segment with primary end-to-end anastomosis.⁽⁸⁾ Our case contributes to the understanding that distal bowel attachment to the umbilical cord may occur even in presumed jejunoileal atresia.

Prenatal findings such as polyhydramnios and dilated bowel loops were unavailable in this case because no prenatal ultrasonography had been performed. This represented a limitation and may have contributed to delayed diagnosis.

Because this is a single case report, the proposed embryologic mechanism should be interpreted cautiously and regarded as hypothesis-generating rather than definitive.

Conclusion

This case adds to the understanding of jejunioleal atresia pathogenesis by suggesting a possible alternative embryologic mechanism. It also highlights the importance of meticulous umbilical assessment at birth to avoid preventable iatrogenic injury to the bowel. Surgeons and neonatologists should maintain a high level of suspicion for atypical anatomic variants in neonates presenting with intestinal obstruction and abnormal umbilical morphology.

Acknowledgements

The authors would like to express their gratitude to all participants who were involved in this study.

Conflicts of interest statement

The authors have each completed the International Committee of Medical Journal Editors Form for uniform Disclosure of Potential Conflicts of Interest. No authors have any potential conflict of interest to disclose/None of the authors disclose any potential conflict of interest.

Data sharing statement

Data generated or analyzed for the present report are included in this published article. Further details are available from the corresponding author on reasonable request after deidentification of the patient whose data are included in the report.

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