Case Report

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Neonatal appendicitis causing intramural ileocecal stricture and intestinal obstruction

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Abstract:

Neonatal appendicitis (NA) is a rare entity, and resultant ileocecal (I-C) segment stricture is never reported before. It often poses a diagnostic dilemma. Diagnostic delays could lead to increased morbidity, such as perforation, peritonitis, sepsis, and intestinal obstruction. We report a case of NA causing intramural I-C stricture and resultant intestinal obstruction in a preterm infant. Progressive abdominal distension, feeding intolerance, and constipation were the triad of clinical presentations purporting an evolving obstructing-inflammatory response to a recent episode of appendicitis. Early laparotomy, resection of the I-C segment along with the diseased appendix, and ileocolic anastomosis resolved the child's obstruction symptoms.

Keywords:

Case report, ileocecal stricture, neonatal appendicitis, neonatal intestinal obstruction

Introduction

Neonatal appendicitis (NA) is an extremely rare condition, and its true incidence is not known. Due to the lack of specific signs and low index of suspicion, the diagnosis of acute appendicitis in neonates is often delayed with a higher morbidity and mortality rate.[1-3] High index of suspicion in neonates with abdominal obstructive symptoms and radiological assessment could aid in early diagnosis and management. Intestinal obstruction in neonates is caused by a variety of conditions, including intestinal atresia, malrotation, volvulus, meconium ileus, meconium plug syndrome, and necrotizing enterocolitis. Only a few reports of NA associated with intestinal obstruction in neonates have been reported.[4] NA, causing intraluminal fibro-obliterative stricture and intestinal obstruction, has never been reported so far. Diagnosis can be delayed with serious consequences, including intestinal perforation, sepsis, bowel ischemia, and gangrene.

Case Report

Preterm (26 weeks gestation) male baby with a birth weight of 900 g with no significant antenatal events or maternal illness, was noted to develop feeding intolerance, abdominal distension, and progressive constipation for 3 days at 38 weeks of corrected age in the Neonatal Intensive Care Unit. Upon evaluation, the baby was found to have distended abdomen with prominent visible bowel loops, no abdominal wall edema/erythema, scanty stool on rectal stimulation associated with bilateral scrotal swelling [Figure 1]. The baby had associated retinopathy of prematurity and static left-sided hydrocephalus. Radiology revealed an air-fluid level all over the abdomen, ileal cutoff line, and paucity of air in the pelvic region, suggesting distal bowel obstruction [Figure 2]. Ultrasound of the abdomen revealed dilated small bowel loops with sluggish peristalsis, suspected bowel

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ischemia, thickened terminal ileum and ileocecal (I-C) region, with multiple lymphadenopathies. Urgent exploratory laparotomy was done which revealed dilated ileum with tight stricture at the I-C region and a fibrotic appendix associated with vascular fibrous adhesive bands and lymphadenopathy [Figures 3 and 4]. The rest of the intestine and peritoneal cavity was unremarkable. Enterotomy was done in the terminal ileum, 6 Fr feeding tube was introduced through the ileotomy, and was attempted to negotiate through the I-C junction but it was impassable. Only a small volume of saline could be passed with pressure. Segmental resection of the terminal ileum and I-C junction with the appendix was done [Figure 4], and end-to-side I-C anastomosis was performed to restore intestinal continuity. The child had a smooth recovery with rapid resumption of feeding on the first postoperative day and normal stooling in 3 days' time. Histopathology revealed I-C junction stricture with proliferation of cells with areas of ulceration and acute inflammatory exudate, nonspecific inflammation, and fibrosis of the appendix and reactive lymph nodes. The terminal ileum was dilated with a normal wall and mucosa. At 6 months postoperative follow-up, the child was thriving well with positive weight gain and normal stooling.

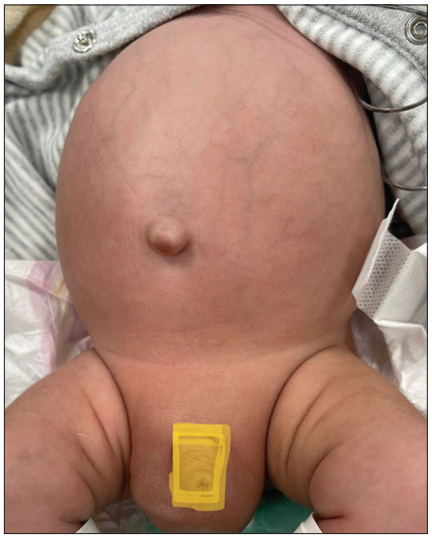

Figure 1: Abdominal distension with scrotal edema

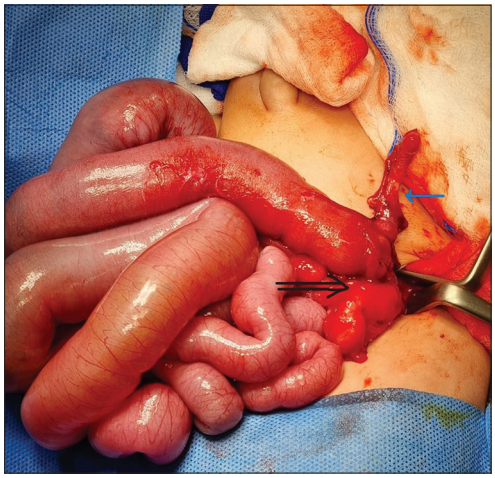

Figure 3: Intra-operative picture showing fibrotic appendix (blue arrow) and tight ileocecal junction stricture (black arrow)

Discussion

NA is extremely rare with a reported incidence of 0.04%–0.2%, more frequent in premature males, and is associated with high morbidity and mortality.[1,2] This low incidence is due to several factors. The appendix is still in its fetal form, i.e., funnel shaped having a wide opening into the cecum, and thus less prone to obstruction than the mature finger-like shape in older children.[2] Intraluminal obstruction is unlikely in

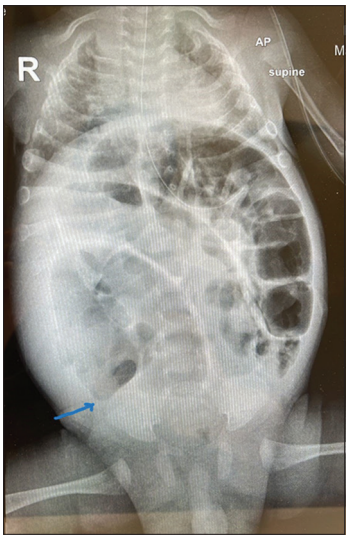

Figure 2: X-ray abdomen showing distended bowel loops, ileal cutoff line (blue arrow), and paucity of pelvic gas

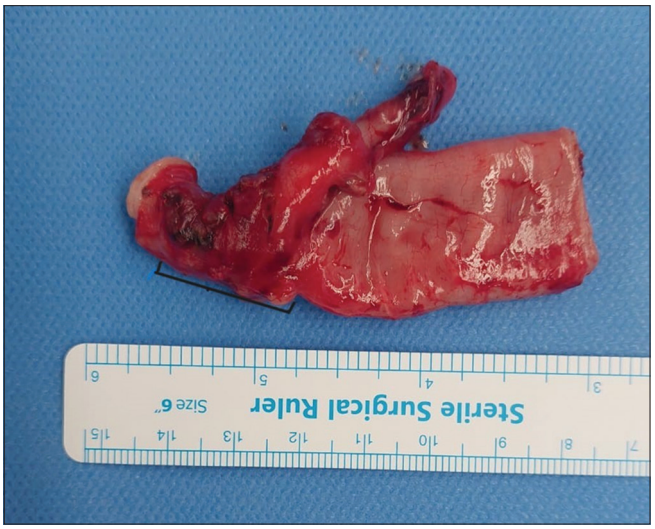

Figure 4: Resected ileocecal segment with appendix and stricture segment (black marking)

neonates due to recumbent posture and a liquid diet.[3] NA is highly fatal, especially among preterm infants, and needs early intervention to avoid complications such as perforation, intraperitoneal sepsis, delayed intervention with resultant systemic sepsis, and intestinal obstruction leading to bowel ischemia and gangrene.[4-7] The symptoms of NA are usually nonspecific, and a high index of suspicion is warranted for the diagnosis. Some features are indicative, such as irritability, distressed breathing, and wriggling, indicating peritoneal inflammation as seen in our patient for 1-week period. There may be abdominal distention, bilious vomiting with induration, edema, and erythematous rash over the abdominal wall. This may be associated with swelling of the scrotum and a right lower quadrant mass. Other, less consistent findings are anorexia, fever, and leukocytosis. Abdominal radiograph may show abnormal gas pattern, free peritoneal fluid, and air, thickened abdominal wall, a right scoliosis, and obliteration of the psoas margin. The presence of calcified appendicolith, a radiographic presence seen in a good number of patients in the older age group, has never been reported in newborns.[5-7] On abdominal ultrasonography, the presence of intra-abdominal abscess, the absence of gas in appendiceal lumen, or evidence of collection in the right iliac fossa strongly suggests acute appendicitis. Spiral computed tomography can also be a very useful diagnostic tool but rarely practiced in neonatal presentation.[8-11] As the signs and symptoms are not characteristic, the incidence of perforation is high in NA. The other reasons are a thin appendiceal wall and an indistensible cecum.[4,5] A relatively small, undeveloped, and functionally nonexistent omentum, small size of the peritoneal cavity allowing a more rapid and diffuse contamination, and little physiological reserve, are important factors contributing to the high morbidity and mortality associated with perforation peritonitis in infants.[8,11,12] However, contrary to late childhood, perforation of the appendix in this age group may be due to Hirschsprung's disease, meconium plug syndrome, cystic fibrosis, necrotizing enterocolitis, and gastroenteritis.[8,12]

The concept that green vomiting is indicative of mechanical obstruction until proved otherwise is a foundation of the neonatal practice. Few cases, however, there may not have a surgical cause, and many cases of distal bowel obstruction may not demonstrate bilious vomiting, especially the partial and the developing ones. Babies with definitive features of intestinal obstruction shall have an early and prompt evaluation by the neonatal medical and surgical teams.

In our case, the baby is presented with suspected obstructive features such as feed intolerance, distended and loopy abdomen, and progressive constipation. Following a period of irritability, hypoactivity for 1-week duration, radiological investigations were in favor of intestinal obstruction, and blood investigations were also within normal limits without signs of severe sepsis. Differential diagnosis included malrotation and congenital band obstruction. Intestinal obstruction in acute appendicitis is often due to mass formation, local abscess formation, generalized peritonitis, or late postoperative adhesions. There are a few reports of perforated NA causing intestinal obstruction in neonates.[7,9-12] Perforated infantile appendicitis cases complicated with intestinal obstruction due to adhesions have also been reported.[10-12] A systematic review was done by Makama *et al*. in 2017 about intestinal obstruction caused by appendicitis and found that it occurs predominantly among male babies and had a wide age range from 3 years to over 80 years.[10] In 84.4% cases the cause was mechanical obstruction resulting from one or a combination of the following: Adhesions, herniation through a ring or gap formed by the appendix tip being attached to its base, appendix tip attached to the bowel causing a torsion, kinking of the bowel and complex knotting Appendicular band syndrome has been reported as a rare sequel of NA where in adhesive band following an episode of NA tend to cause extraluminal obstruction of small/large bowel.[6] However, intramural fibro-inflammatory reaction to NA, causing I-C junction stricture and progressive intestinal obstruction, has never been reported hitherto. Most of these cases of postappendicitis intestinal obstruction had only intraoperative diagnosis, atypical progression of symptoms, and increased morbidity due to delayed presentation.

Conclusion

NA remains a challenging disease, with intestinal obstruction due to I-C junction stricture representing a rare complication. Their nonspecific symptoms, especially in the rare occurrence of fibro-inflammatory stricturization of the bowel lumen, pose a diagnostic dilemma in neonates. Early intervention is helpful for the diagnosis and treatment of both NA and intestinal obstruction when symptoms are persisting.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal patient identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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