



Unexpected Ileal Perforation in A Neonate with Anorectal Malformation: A Rare Case Report

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Abstract: Intestinal perforation in neonates with anorectal malformations (ARM) is rare but associated with high mortality. This report presents a rare case of ileal perforation in ARM of ileal perforation associated with ARM. A 4-day-old female neonate presented with abdominal distention, fever, and failure to pass meconium; babygram examination showed only meteorism without signs of pneumoperitoneum. Exploratory laparotomy revealed ileal perforation with meconium leakage, followed by resection and anastomosis, along with the creation of a sigmoid loop colostomy with a plan for subsequent anoplasty. Despite surgical intervention, the patient unfortunately died on the third postoperative day due to sepsis. Bowel perforation is a known complication of ARM and may arise from vascular or congenital causes; the ileum, being thinner than the colon, is less able to tolerate increased intraluminal pressure, and perforation can occur even in the absence of pneumoperitoneum, with anterior abdominal wall edema serving as an important clinical clue. Although ileal perforation in ARM is extremely uncommon, early recognition of this potential complication is essential to prevent deterioration and reduce mortality in affected neonates.

Keyword: Anorectal Malformation, Ileal Perforation, Bowel, Intestine.

INTRODUCTION

According to statistics from many major hospitals in different regions of Indonesia, ARM occurs in between 1 in 5,000 to 1 in 7,000 live births. (Chan et al., 2014) Intestinal perforation in newborns is an uncommon condition that can increase mortality from 3% to 23% for patients with anorectal malformations. Bowel perforation has been recorded in 2% of neonates with ARM, and it can rise to 9.5% with delayed presentation. (Gunadi et al., 2020;

Olatunji et al., 2015) To the best of our knowledge, this case is the second case report of ileum perforation in patient with ARM. We merely found a similar case from Nigeria in 2015. This is an uncommon case as there has only been one documented case of ileal perforation and most ARM-related intestinal perforations occur in the colon. (Gunadi et al., 2020) When ARM patients have an intestinal perforation, their condition might deteriorate and they may experience complications. Therefore, it's critical to understand the potential issues associated with ARM.

METHOD

A 4-day-old girl presents with abdominal distention and no anal opening. The infant hadn't passed meconium for four days. She also experienced fever. Heart rate was 157 bpm, Respiration rate was 57 breaths/min, temperature was 38.8°C and oxygen saturation was 93% with nasal cannula. There was no erythema or edema of the anterior abdominal wall, although the abdomen was painful and enlarged upon physical examination, and bowel sound was absent. There is no anal dimple and there was no visible meconium on the urine or perineum. Blood test showed hemoglobin 20, leukocytes 21,640 and platelets 258,000, serum electrolytes showed sodium 144, potassium 4.29 and chloride 109.71. Babygram revealed only meteorism.



Figure 1. Babygram showed meteorism



Figure 2. Ileal perforation was discovered intraoperatively

Laparotomy was done, and an ileal perforation was discovered with meconium leaking out. The ileum was debrided followed by resection and anastomosis. The perforation was closed using Vicryl - 4.0, interrupted sutures.

In this case, we did a sigmoid loop colostomy and intended to have an anoplasty later. The patient experienced peristomal infection and surgical site infection in the suture wound after colostomy. The infection was then treated. Anoplasty was performed after 6 months and anal dilatation was continued on a regular basis. There were no complaints or problems after six months of follow-up, although the patient was later lost to follow-up.

RESULTS AND DISCUSSION

Bowel perforation is a serious complication of ARM with high mortality from 3% to 23%. (Parelkar et al., 2016) There are two possible causes of bowel perforation: vascular and congenital. As a result of vascular etiology, blockage in the ARM raises intraluminal pressure and impairs with transmural perfusion, resulting in localized ischemia. This can also happen when there is distal blockage, and a congenital muscular insufficiency in the tail gut area causes the rectum at risk for perforation. (Gunadi et al., 2020; Parelkar et al., 2016; Singh et al., 2023) In our case, due to the incompetent ileocecal valve and the ileum wall being relatively thinner than the colon, the intraluminal pressure could not be maintained resulting ileal perforation. (Tong & Ludwig, 2013)

Boys experience about 85% of perforations, whereas girls are less likely. This is probably because low ARM with large rectoforchette fistulas is more common in girls. (Olatunji et al., 2015) Patients with a fistula often have a lower risk of perforation.

However, the presence of a fistula in patients with ARM does not always rule out the likelihood of a perforation, particularly in individuals with tiny fistulas. Fistula obstruction induced by inspissated meconium can produce a rise in intra-luminal pressure, leading to tension gangrene and perforation. (Olatunji et al., 2015; Parelkar et al., 2016)

We found no indications of perforation on physical examination or in babygram. This is consistent with the findings of Tijesuni et al., which reported that 88% of these individuals had no clinical signs of intestinal perforation. (Gunadi et al., 2020) Perforation should be suspected if there is oedema in the front abdominal wall. Nonetheless, intestinal perforation is not excluded in the absence of pneumoperitoneum.

In this case, we planned an anoplasty after performing a sigmoid loop colostomy as the primary closure of the perforation. This is consistent with other studies stating that anoplasty and low-lying ARM may be performed concurrently. Nevertheless, in high-lying MAR cases, it is performed later.

CONCLUSION

Although ileal perforation in ARM is extremely uncommon, it is crucial to be aware of potential issues related to ARM in order to prevent the patient's condition from getting worse.

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