Scientific Foundation SPIROSKI, Skopje, Republic of Macedonia
Open Access Macedonian Journal of Medical Sciences. 2024 Apr 30; 12(2):Ahead of print
https://doi.org/10.3889/oamjms.2024.11891
eISSN: 1857-9655
Category: *C - Case Reports*
Section: *Case Report in Surgery*

Spontaneous Caecal Perforation in a Male Neonate: A Case Report

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Edited by: Igor Spiroski
Citation: Banu S. Spontaneous Caecal Perforation in a Male Neonate: A Case Report. Open Access Maced J Med Sci. 2024 Apr 30; 12(2):Ahead of print. https://doi.org/10.3889/oamjms.2023.11891
Keywords: spontaneous perforation, cecum, neonates, surgical
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Received: 01-Mar-2024
Revised: 27-Mar-2024
Accepted: 30-Mar-2024
Ahead of print: 30-Apr-2024

Funding: This research did not receive any financial support
Competing Interests: The authors have declared that no competing interests exist

Abstract

BACKGROUND: Spontaneous perforation of the colon in full-term neonates is a rare case, with perforation of the cecum being rarely reported.

CASE PRESENTATION: We present a rare case of spontaneous perforation of the cecum in a term neonate who presented with vomiting and flatulence on the second day of life. Surgical intervention revealed full-thickness perforation in the caecum. Histopathologically, there was no necrotizing enterocolitis and Hirschsprung's disease.

CONCLUSION: Clinical awareness in this rare case can help prevent delays in diagnosis and hopefully prompt surgical management.

Introduction

Gastrointestinal perforation is a surgical emergency condition in neonates that is commonly observed in the small intestine. Meanwhile, perforation of the colon is a rare condition [1], [2], and [3]. In premature neonates, colon perforation is most commonly caused by necrotizing enterocolitis (NEC), generally associated with Hirschsprung disease, meconium plug syndrome, small left colon syndrome, idiopathic perforation, anorectal malformation, and cystic fibrosis [1], [4]. In the term neonates, Hirschsprung disease is the most common cause, followed by NEC and spontaneous idiopathic perforation [5], [6]. Spontaneous idiopathic gastrointestinal perforation is common in neonates, and it decreases with age in children. The cause of spontaneous idiopathic intestinal perforation is still unknown. Patient clinical presentation can be variable, with the majority of cases presented with a feature of diffuse peritonitis. Being an infrequent pathology, spontaneous idiopathic intestinal perforation is mostly an intraoperative diagnosis, which is associated with high mortality rates. The treatment for spontaneous idiopathic intestinal perforation is also seldom discussed. In neonates, idiopathic caecal perforation is very rare. We presented a case report of a term male baby with idiopathic caecal perforation to highlight this surgical emergency in terms of clinical presentations, surgical management, and outcomes [1], [2], [7], [8].

Case Presentation

A 3-day-old baby was brought to the emergency room with a complaint of vomiting and abdominal distention for 2 days. He was born with a weight of 2.5 kg from a 28-year-old woman vaginally. The baby cries immediately after birth, and meconium is present for more than 24 h. The mother's milk is hard to collect due to low output, and the baby was given formula milk. There were no abnormalities during pregnancy. There was no family history of the same illness. On physical examination, the baby tends to be inactive, with signs of dehydration, a distended abdomen, visible abdominal superficial veins, no erythema of the skin, and no bowel sounds. On rectal examination, anal sphincter tone was absent, and no stool was found on the gloves. The pneumoperitoneum is clearly visible on two positions of abdominal X-ray, namely, the Anteroposterior and left lateral decubitus, as shown in Figure 1. An orogastric tube was performed, and fluid therapy and antibiotics were administered. On examination, at the time before surgery, the baby had a pulse of 138×/min and a temperature of 38.5°C. Then, an exploratory laparotomy was performed by making a transverse supraumbilical incision. When opening the peritoneal cavity, fecal contamination was seen. One large 1 × 1 cm full-thickness perforation was found in the caecum, and then an edge biopsy was performed surrounding the perforation. The caecal perforation was repaired, and an ileostomy was

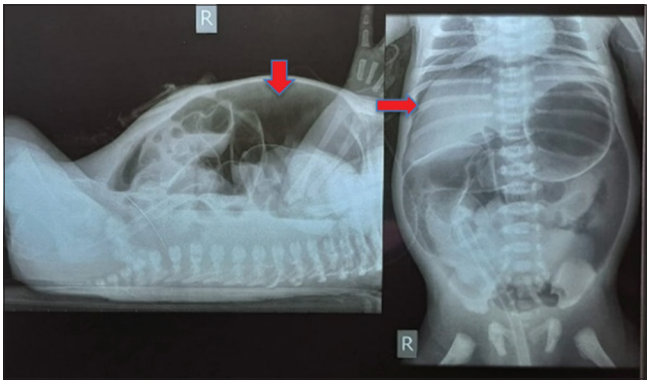

Figure 1: Two position abdominal X-ray shows a massive pneumoperitoneum, shown in red arrow

made. The vital signs during surgery were 120×/min in pulse, 38°C in temperature, and 140×/min in pulse, 37.5°C in temperature after surgery was done. But then the patient died 4 h after the completion of the operation. Anatomical histopathology analysis of the edge of the caecal perforation sample did not reveal either Hirschsprung's disease or NEC, as shown in Figures 2 and 3.

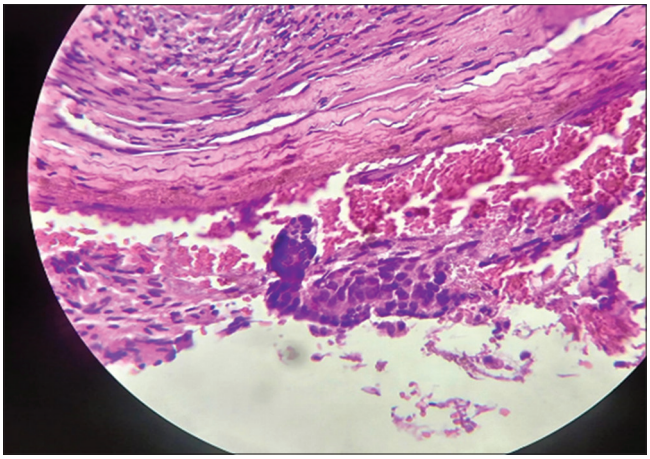

Figure 2: In 400× magnification, erosive mucosa and ulceration was found

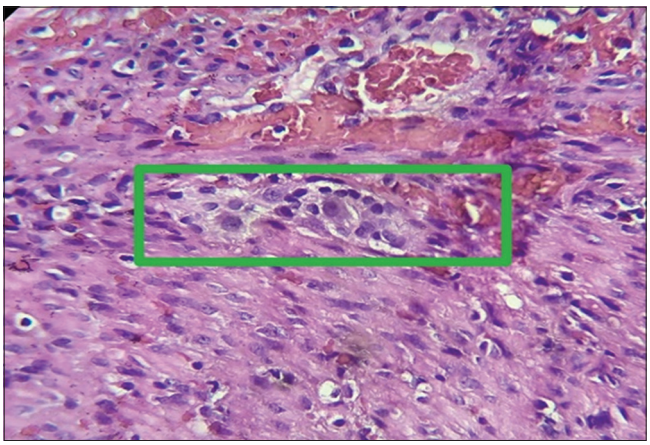

Figure 3: In 400× magnification, a mature ganglion in the Auerbach plexus was seen between the longitudinal and circular muscle layer

Discussion

Hirschsprung's disease is probably the only condition that correlates with caecal perforation that has been studied in case studies [1]. However, an incidence of 3–4% occurs in the long segment of Hirschsprung's disease. Another etiology that is closely related to this case is NEC. However, in general, NEC is usually found in cases of pre-mature babies. If there is perforation, the ileum and jejunum are usually also involved in cases of pre-mature infants [1], [3]. The most significant etiology in the pathophysiology of NEC is intestinal mucosal damage due to hypoxemia and sepsis [2], [3], and [9]. According to Resch *et al*., hypoxic conditions resulted in ischemic injury to the intestine along with intestinal immaturity, which could lead to perforation in this baby [10]. A classic sign, namely a pneumoperitoneum, can be visualized on an upright X-ray of the abdomen. Neonates with spontaneous intestinal perforation can be treated by primary peritoneal drainage or combined with laparotomy [11], [12].

The optimal management plan depends on the location of the perforation, degree of contamination, and hemodynamic stability of the neonate [3], [4]. In hemodynamically stable patients with a single well-localized perforation, primary closure or anastomotic resection may be performed. In caecal perforation, unless Hirschsprung's disease is ruled out, primary repair alone is impossible, such that a protective stoma is usually performed. A biopsy is recommended in all cases [13]. A frozen section (FS) can be used to assist in decisions in a short amount of time, but not all hospitals can do a FS. Because the ileocaecal valve cannot be salvaged, an ileostomy must be performed. The morbidity in these patients remains high, with a stoma requiring extensive resuscitation in the intensive care unit [1], [2], and [3]. Despite these efforts, the mortality rate in most studies is close to 50% [3], [5]. The final diagnosis, in this case, was idiopathic caecal perforation because there is no evidence of another differential diagnosis such as NEC, Hirschsprung's disease, and meconium ileus clinically, radiologically, or histopathologically.

Conclusion

Spontaneous caecal perforation in neonates without any known cause is a rare case. A detailed history, physical examination, and histopathological analysis are essential for determining the right course of action. Even with the best care possible, babies with perforated colons tend to have a poor prognosis.

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