



Case report

Ileal lactobezoar in extreme premature infant complicated by intestinal perforation: A case report

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ABSTRACT

Introduction and importance: Lactobezoar is a rare complication that has been reported more in the stomach, however it may be located anywhere in the intestine.

Case report: Reported here, is a case of ileal lactobezoar which was complicated by perforation and was mimicking necrotizing enterocolitis in presentation, ex preterm (26 weeks) male infant who presented at day of life 18th (2 days after BM fortification) with hemodynamic instability and intestinal perforation, which was diagnosed by Abdominal X-ray and Ultrasound necessitating urgent laparotomy.

Clinical discussion: Laparotomy revealed an area of ileal perforation and an inspissated mass which was confirmed to be lactobezoar by pathology, ileostomy was performed. The baby had an acute postoperative status of hypovolemic shock which was managed clinically, then was restarted on feeds, and the stoma was reversed 9 weeks later.

Conclusion: Lactobezoar, although rare, but numbers increased especially with the rise in numbers of extremely preterm infants worldwide, it most commonly presents later in life but in some cases, such as our case it may happen in 1st 2–3 weeks after birth and may cause significant complications as perforation making its differentiation from common GI problems in neonates as NEC more challenging.

1. Introduction

In neonatal intensive care units (NICUs), preterm infants especially those who are born extremely prematurely are in increased risk of serious life threatening GI problems, of which Necrotizing Enterocolitis (NEC) is the most frequent [1], occurring in 15 % of low birth weight and premature infants admitted to NICUs, [2,3]. Although, most cases are managed medically, around 20–40 % of these infants may progress and develop complications such as intestinal perforation necessitating laparotomy [1]. However, other rare conditions, which may present with clinical and radiologic features that mimic NEC also requiring surgical intervention, maybe spontaneous intestinal perforation, volvulus without malrotation, or as in our case, lactobezoar [4–6]. Lactobezoar, is a rare GI problem in infancy, and is primarily found in the stomach, but, it may occur in any part of the intestinal tract [4]. Lactobezoar often occur 14 days after adding human milk fortifier to the breast milk [7].

Here we present a case of a premature infant with a pre-operative diagnosis of intestinal perforation from NEC, with intra-operative findings of ileal lactobezoar complicated by intestinal perforation. The infant was originally managed and transferred from a perinatal center to The Hospital for Sick Children for surgical evaluation and management.

2. Methods

A single case report following the SCARE 2020 criteria [8].

3. Case presentation

A male infant of Indian descent was delivered vaginally in one of the perinatal centers in Toronto, at 26-weeks' gestation with a birth weight of 900 g and APGAR scores of 8 and 8 at 1 and 5 min, respectively. He received a dose of surfactant by minimally invasive techniques and then

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was maintained on NCPAP for the treatment of respiratory distress syndrome. The infant was treated with caffeine as a part of preterm routine care. Maternal history was negative for drug exposures. Family history was negative for GI diseases. There was no history of known allergies. He continued to be managed at the perinatal center.

Gavage feeding with the mother's expressed breast milk (EBM) was started on the day of life (DOL) 2. On DOL 16, the EBM was fortified to 24 kcal/oz. using mother's(hindmilk and extensively hydrolyzed formula. On DOL 18, he presented with significant apneic episodes for which he was intubated and mechanically ventilated, kept NPO, a wide pore nasogastric tube was placed for abdominal decompression. Physical exam was remarkable for abdominal wall erythema and distention.

Feeds were held and a full septic work-up was initiated; intravenous triple antibiotics (vancomycin, cefotaxime and metronidazole) were started. Initial laboratory data was remarkable for leucopenia with WBCS $5.81 \times 10^9/L$, anemia with hemoglobin of 113 g/l and thrombocytopenia with PLT of $54 \times 10^9/L$ and C-reactive protein of 73 mg/L. Additionally, arterial blood gases showed severe metabolic acidosis (PH 7.05) elevated lactic acid (7 mmol/L).

Abdominal X-rays (Anteroposterior and lateral cross-table views) were done in the bedside and revealed distended bowel loops in ileocolic region containing a non-specific opacity (meconium or a bezoar) as in (Figs. 1 & 2). Abdominal ultrasound indicated multiple distended bowel loops containing echogenic debris in the mid and left abdomen.

He continued to deteriorate with evolving signs and symptoms of peritonitis complicated by septic shock requiring volume expanders and vasopressors support which warranted an eminent transfer to the hospital for sick children for surgical evaluation, in the context of clinical deterioration, laboratory derangement and radiologic concerns, decision was to take the baby for urgent laparotomy 8 h after the intital deterioration.

At the time of surgery, hemodynamic status had improved after volume expansion with crystalloid and colloid and vasopressor support with norepinephrine. The infant was given morphine for sedation and pain control. Parents were counseled by NICU and surgical team about the gravity of the condition which mandates surgical intervention.

3.1. Intra-operative findings

Exploratory laparotomy was performed at the hospital for Sick Children by a pediatric surgeon with 15 years of experience. The infant was properly sedated, analgesia was given and the procedure was done

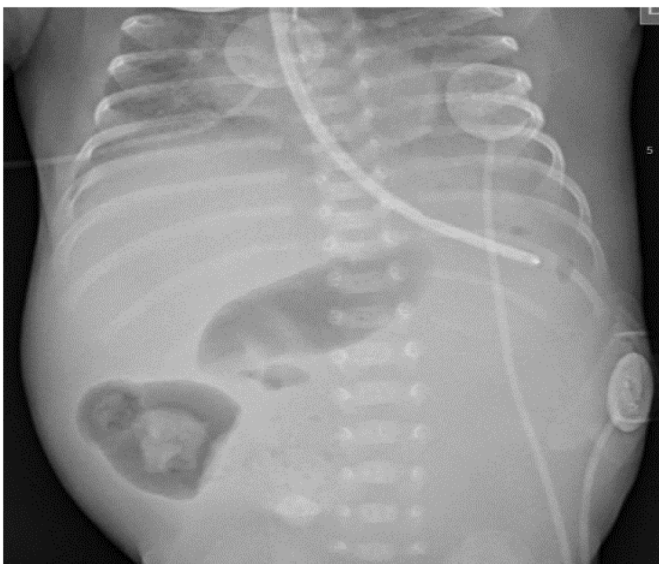


Fig. 1. AP view.

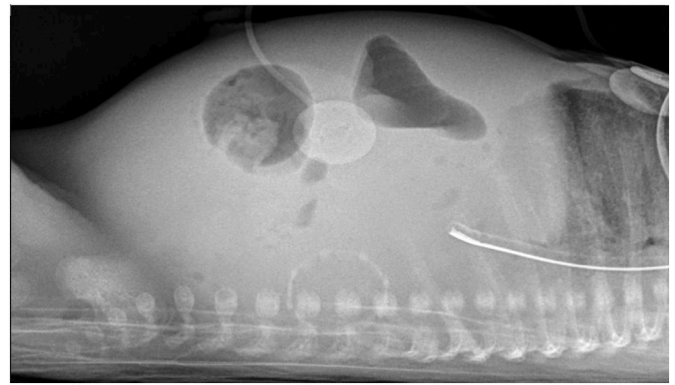


Fig. 2. Lateral cross-table view.

under general anesthesia. Upon entry into the abdominal cavity, significant serous ascites and partially digested feeds were found. A bowel segment, approximately 12 cm from the ileocecal valve was frankly perforated with approximately 75–80 % disruption of the intestinal luminal circumference (Fig. 3). Distal to the perforation site, a firm mass within the lumen of the terminal ileum was milked out through the perforation site. The mass appeared to be a bezoar of inspissated milk (Fig. 4) that was lodged at the site of bowel perforation.

There was a distinct change in the ileum caliber, with a transition from markedly dilated bowel proximal to the bezoar and collapsed, smaller caliber bowel distal to the bezoar. No web or atresia was identified in that bowel. We hypothesized that this bezoar caused mechanical small bowel obstruction resulting in perforation of the proximal bowel. The remaining small bowel was normal in appearance without evidence of necrotizing enterocolitis. In total, 7 cm of ileum including the transition zone were resected and the specimens sent to pathology. A double-barreled ileostomy was created because the infant was difficult to ventilate and was hemodynamically unstable, and thus restoring bowel continuity was not attempted at that time. Over all, the procedure was well tolerated with no surgical complications.

3.2. Pathological findings

Examination of the resected portion of the bowel revealed ischemic

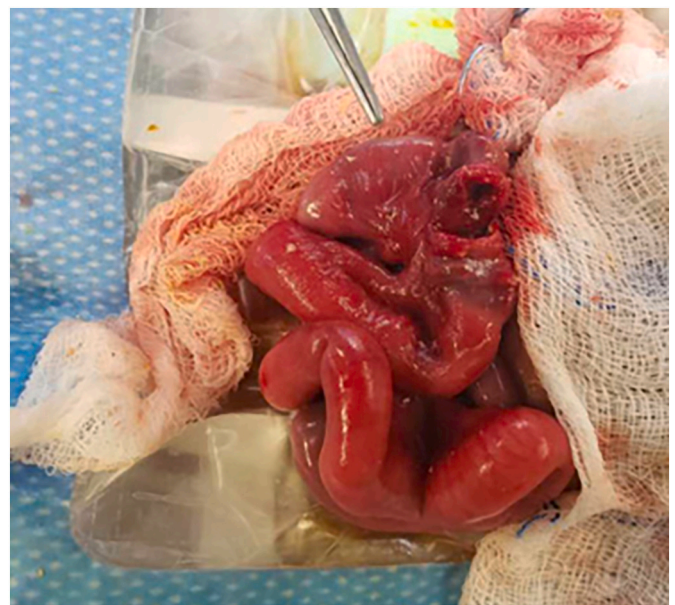


Fig. 3. Image of dilated ileum proximal to obstruction.

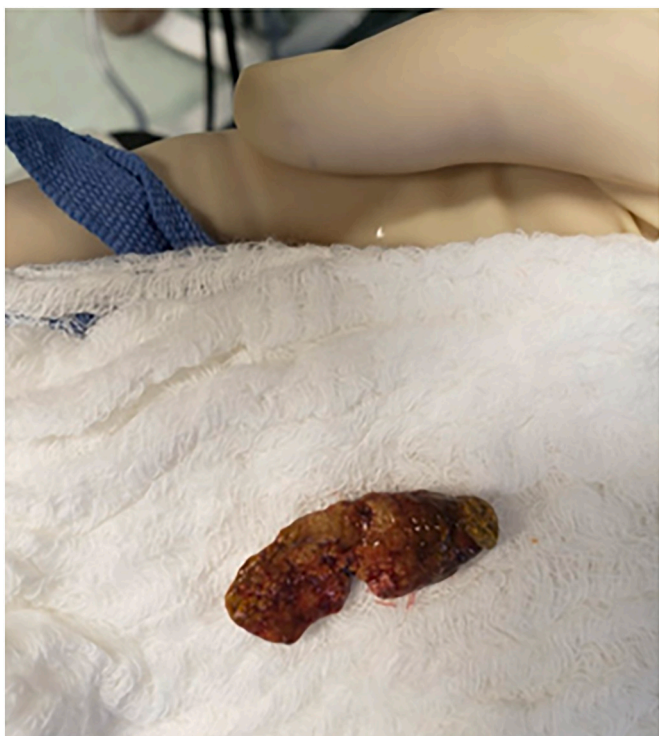


Fig. 4. Lactobezoar extracted from ileum.

changes, ganglion cells present, and no evidence of atresia or web.

3.3. Post-operative course

Patient presented to NICU in a persistent state of instability and developed a picture of hypovolemic shock, requiring further blood products, volume expanders, and vasopressors, over the next 12 h his condition stabilized, and the vasopressors were discontinued. The surgical team advised to keep NPO and on IV triple antibiotics for 10 days and the patient was adherent to the plan guided by NICU team, Full enteral feeds were achieved on postoperative day 50 and stomal reversal was performed 9 weeks after the initial laparotomy.

4. Discussion

Our case adds to the current literature because our case was a pre-term baby, weighing 900 grams who presented with lactobezoar located in the ileum, an unusual location for a lactobezoar. And with the onset of symptoms 48 hrs after fortification of the BM, an earlier time of presentation than reported in older neonates, and lastly, an atypical presentation similar to NEC making the preoperative diagnosis difficult, and our case was critically ill. Successful case management was the collective effort of multiple teams including NICU teams consisting of neonatologist, respiratory therapists, pharmacists and neonatal nurses at 2 different sites, and the surgical and anesthesia teams, and dieticians as feeds were advanced after intestinal recovery.

A lactobezoar is a milk coagulum that causes mechanical obstruction of the gastrointestinal tract. Obstructing lactobezoars are clots, consisting entirely of inspissated milk, or fatty acid calcium stones [9]. It was first described in 1959 by Wolf and Bruce in a report a term neonate fed infant formula containing higher levels of casein [14]. Later, in the 1970's, the problem was addressed by the introduction of formulas with a more physiologic casein and whey ratio similar to human milk, resulting in a reduction in the occurrence of lactobezoars.

In the last twenty years, there has been a resurgence of lactobezoars after the introduction of high-caloric- density -feeds to extremely low

birth weight infants [9]. Premature infants show delayed gastric emptying and bile acid secretion, with diminished gastric fluid volume, acid secretion, pepsin activity, and bile salt concentration. These factors affect bile composition and function, resulting in slow absorption and delay in gastric and intestinal transit times [10]. Protein, calcium, phosphorus, and sodium concentrations in term-infant formulas and breast milk are insufficient to meet the daily requirements of premature infants. Thus, fortifiers were introduced to compensate for these deficiencies [10]. Stasis and precipitation of the fortifier components within the gastrointestinal tract can lead to bezoar formation in the lumen. It is relevant to be mentioned, that such condition can develop in both preterm or full-term infants even with the smallest amounts of enteral feeds, so considering it in the differential diagnosis of the conditions causing feeding intolerance may prevent further progression and clinical deterioration [4].

Clinical presentation includes abdominal distention, abdominal wall erythema, apnea, and desaturation in premature infants [7]. The onset of symptoms varied from 3 to 47 days after the fortifier was added to feeds with a mean of 14 days [7]. In the retrospective studies we reviewed, the diagnosis of lactobezoar was generally made intra-operatively at the time of the surgical management of intestinal obstruction and/or perforation [7,9,11]. In some cases, which are similar to our case, misrecognizing the condition in its early stages, may progress into intestinal obstruction and even perforation causing a picture of peritonitis and septic shock [4].

Radiologically, some reports connect lactobezoar to a characteristic appearance on x-rays (intraluminal mass with a halo of air) [12] which is similar to the finding in our patient, but this finding did not help to establish diagnosis preoperatively and was only linked to it on retrospective manner.

In most of the reports, lactobezoars if diagnosed preoperatively, can be managed in a conservative manner by periods of bowel rest, Intravenous nutrition, gastric lavage in cases of gastric pathology [4], Gastrografin enemas used in lactobezoars in the small and large bowel, but may be complicated by bowel perforation [12,13], Surgical intervention should be preserved to those infants who have failed medical management or whom developed complications as intestinal perforation [4] which is consistent with our case.

5. Conclusion

We describe a case of ileal lactobezoar complicated by intestinal perforation in premature infant. We can conclude based on our patient that lactobezoar can develop very early following initiation of fortified feeds in a premature infant, and the lactobezoar can be in the distal small bowel which is more likely to lead to various problems as intestinal perforation, peritonitis and septic shock if not detected and managed earlier. This presentation differs from what has been described in term infants in whom it is commonly located in the stomach. Differential diagnosis of intestinal obstruction caused by lactobezoar should be considered in premature infants with the recent fortification of feeds with signs and symptoms of intestinal obstruction. Early recognition of this potential complication of feed fortification prevents the evolution of more serious complications.

5.1. Patient perspective

Our patient is in neonatal age group, so we substituted this section by his parents' perspectives. They were so concerned about the disease itself and its evolving and overwhelming course, especially when they faced that the problem is different about the presumed diagnosis before the operation. They were communicated in a conference meeting preoperatively through NICU and surgical team about the procedure, technique, steps and possible complications, they expressed their worrisome but also their hopes that the procedure could eliminate the concurrent problem and improve their baby's general conditions.

Provenance and peer review

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Ethical approval

The case report was exempted from our institution.

Consent

Written informed consent was obtained from the patient parents for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Mohamed Elkhoul and Farid Aleali had equal contribution as co-authors in collecting the data of the patient, checking the literature review and writing the discussion.

Ayah Alzamrooni was responsible for the surgical part of the history of the patient.

Dr. Chiu and Dr. Gauda helped in writing and revising the paper with supervision of the team.

Research registration

N/A.

Guarantor

Dr. Estelle Gauda.

Declaration of competing interest

There is no conflict for the author and co-authors.

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