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Perforated Ileal Lactobezoar in a Preterm Neonate with Trisomy 21: Rare Case presentation and Literature Review.

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Abstract

Lactobezoar or milk curd syndrome is a compact mass of undigested milk concretions located within the gastrointestinal tract. Most often found in infants, gastric lactobezoars are the most common form in neonates. Intestinal lactobezoar are less common and can cause intestinal obstruction and perforation. We are presenting a unique case of perforated intestinal lactobezoar in a preterm neonate with trisomy 21, who was fed with a low birth weight (LBW) ready-to-feed milk formula together with non-fortified expressed breast milk (EBM).

Keywords: *Perforation, Intestinal, Premature, Lactobezoar, Milk curd syndrome.*

Introduction

Bezoar is a compact mass of foreign material located anywhere within the gastrointestinal tract. Bezoars may contain hair, vegetable matter, or undigested milk masses **[**1**]**. Bezoars composed of undigested

milk aggregates are called lactobezoars and are almost exclusively found in infants. Lactobezoar rarely causes intestinal obstruction. Only 26 cases in total have been reported in the English literature after 1985 **[**2,3,4**]**. One case was reported in KSA in 2019 **[**5**]**.

Aim of work

To present a unique case of perforated intestinal lactobezoar to increase awareness about the clinical presentation and management in this age group.

Case Presentation

A preterm 31-week gestation, boy was born by emergency cesarean section to a 34-year-old mother. The Apgar score was 6 and 8 at 1 and 5 minutes respectively, breech presentation, weighed 1.745 kg, dysmorphic features suggestive of trisomy 21, which was confirmed later by chromosomal analysis.

Upon admission, an X-ray showed mild respiratory distress. No survanta was given and the baby was kept on nasal CPAP for 2 days. Echo showed only a closing PDA. Feeding was started with LBW formula together with non-fortified EBM at the age of 5 days. Feeding was tolerated and was increased gradually according to our feeding protocol. On day 12 of life, the baby started to be hypoactive and had mottled skin. Abdominal distention was the only GIT symptom. A full sepsis screen was done and second line antibiotics were started. An X-ray showed pneumoperitoneum (figure 1,2).

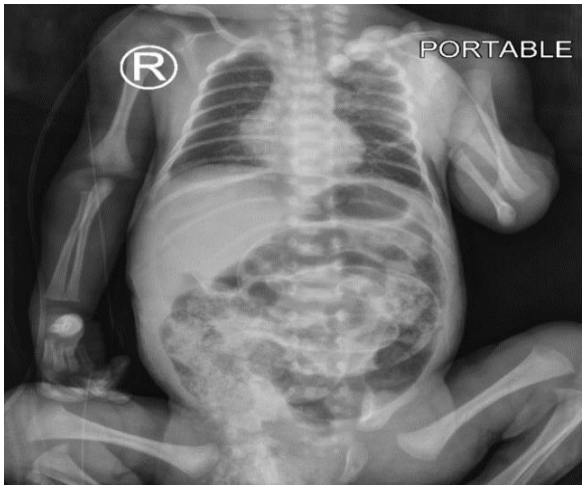

Figure 1

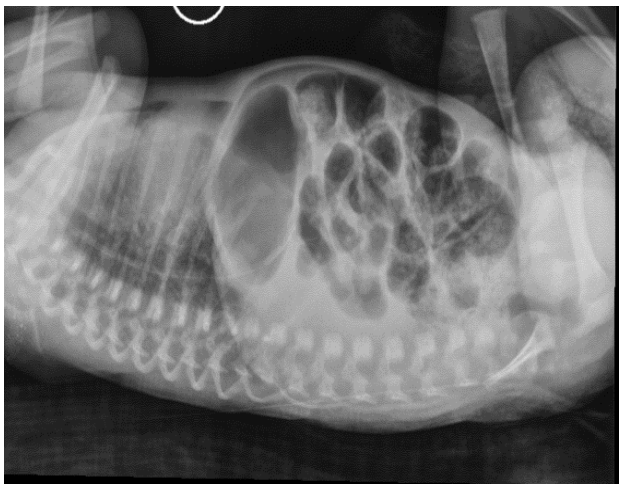

Figure 2

Surgical laparotomy exploration was planned. Exploration laparotomy revealed the diagnosis of intestinal lactobezoar causing intestinal perforation. The bezoar was found at the terminal ileum, ileocecal junction, and proximal colon. The peritoneal cavity was soiled with stony-hard yellow bezoar

materials. Intestinal wash and irrigation with warm normal saline was done to clean out the impacted hard bezoar through the perforation (figure 3).

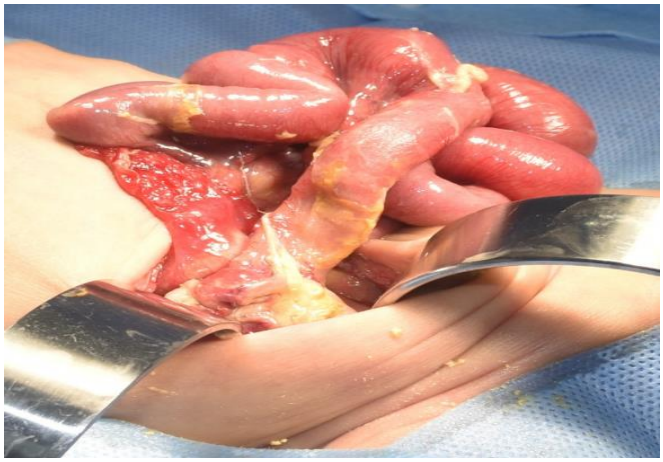

Figure 3

A proximal ileostomy was created to facilitate the healing of the edematous inflamed ileum. On day 18 of life (day 6 post ileostomy) feeding was restarted using the Monogen formula, which is a nutritionally complete special formula, low fat, powdered feed containing whey protein, carbohydrate, fat soluble vitamins, minerals and trace elements. Feeding again was tolerated and was increased gradually. On day 54 of life, the baby reached 2.625 kg and was in a stable general condition, so closure of the ileostomy was done. Feeding was restarted on day 60 of life with the same formula and was tolerated. The regular formula was introduced and was increased gradually until it replaced the Monogen formula. The baby was discharged in good condition on day 74 of life, with a discharge weight of 3 kg.

Discussion

Wolf and Bruce described lactobezoar for the first time in 1959 **[**6**]**. Since that time, many case reports and large studies examining lactobezoars and their formation have been published. Bakken and his associates published a review in 1997 about the lactobezoar which did not report the number of previously documented cases **[**7**]**. Most cases were reported during the seventies, and after 1986, only twenty-six cases were reported in the English literature **[**8**]**.

Lactobezoar is a rare cause of intestinal obstruction and the etiology is uncertain, however prematurity, LBW, thickened formulas enriched with fat, casein and calcium, the addition of fortifiers, gastro-intestinal hypomotility and rapid feeding increase the risk of developing lactobezoars **[**2,8**]**. Lactobezoar obstruction should be suspected in all premature babies with signs of bowel obstruction who are fed EBM with caloric fortification **[**10**]**. In our case, the baby was fed with LBW ready-to-feed formula together with non-fortified EBM. Lactobezoars present with a variety of symptoms and signs. However, a review

of the documented cases revealed several common manifestations. These manifestations included diarrhea, non-bilious vomiting, abdominal distension, a palpable mass, gastric residual and dehydration. Diarrhea, vomiting and abdominal distention were encountered in most of cases. In two of the largest series of patients compiled by Schreiner and associates **[**11,12**]**, some infants presented with increased abdominal girth, abdominal distension was the only clinical sign in seven patients. Gastric residuals or frank vomiting were the main presentation in sixteen patients. Lactobezoar was discovered accidentally during a routine chest x-ray in four patients. Emesis and/or a palpable mass were described as the most frequent symptoms in other case reports **[**13,14**]**. An abdominal radiograph confirms the diagnosis of gastric lactobezoar **[**9**]**. Abdominal ultrasound and an upper gastrointestinal water soluble contrast series should be considered if abdominal radiographs cannot confirm the diagnosis **[**2,9**]**. In about ten percent of cases lactobezoar can be missed and discovered retrospectively during explorative laparotomy.

Conservative treatment and expectant management are the core therapy. Good parental hydration and withholding enteral feeding for several days led to resolution of the bezoar in most cases **[**15**]**. Using contrast enemas in a trial to shift the lactobezoar has a low success rate and may be risky. If diagnosed early, enterotomy and bolus removal are safe and effective treatments. Resection with or without diverting ileostomy may be necessary in case of late diagnosis **[**16**]**.

Conclusion

Lactobezoar is a rare cause of intestinal obstruction which can be benign with early diagnosis. Early diagnosis requires an index of suspicion and good radiological insight; otherwise, it can be easily missed. Any delay in diagnosis and/or management can be fatal. It is very important to keep this entity in mind when dealing with a baby with one or more of the risk factors encountered in the formation of lactobezoar.

Conflicts of interest

The authors declare they have no conflicts of interest.

Authors' contribution

All authors were involved in literature search and preparation of the manuscript.

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