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Cluster of Gastric Lactobezoar in One NICU: 7 Cases in 37 Days, Consequence of Aggressive Enteral Nutrition?

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Abstract

Gastric Lactobezoar (GLB) is a conglomeration of milk and mucus that may cause gastric outlet obstruction. Seven cases appeared in 37-day period in premature babies and one died. GLB appeared on average 18 days postnatally, when enteral feeding reached 130 ml/kg/day. The first symptoms were seen 2.4 days before diagnosis, including abdominal distension, vomiting and regurgitation of curd milk. Diagnosis was made by X-ray in each case. Treatment was medical for three cases and surgical in four cases. Prematurity, consumption of Premature Formula milk (PF), enhanced caloric density, and combining PF with medium chain triglycerides were the main predisposing factors.

Keywords: Bezoar; Newborn; West indies; Enterocolitis; Necrotizing; Enteral nutrition

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Introduction

A bezoar is a trapped mass located in the gastrointestinal system, usually in the stomach, classified according to its major constituents: trichobezoar (hair), phytobezoar (plant material) and lactobezoar (milk). A lactobezoar is an aggregation of mucus with undigested milk constituents. There are several factors that are associated with increased likelihood of lactobezoar formation, including prematurity, low birth weight, an immature gastro-intestinal tract, dehydration, and enteral nutrition with formulas that have a high caloric density or that contain casein or Medium Chain Triglycerides (MCT) [1]. Lactobezoar may occur in full term babies that have been breastfed, but this has only been reported very rarely. Gastric Lactobezoar (GLB) is the most common type of bezoar but it is a rare pathology, only described in medical literature around 100 times since 1959 [2]. Classical symptoms of GLB are abdominal distension, emesis, diarrhea, and an abdominal mass. Diagnosis is most easily made by X-ray, which may show a large radio-opaque mass resembling a "sandwich loaf" or "gingerbread" located in the stomach; this description is pathognomic for GLB. Diagnosis can otherwise be made using ultrasound, which should demonstrate a large, heterogeneous, hyperechoic mass in the stomach [3]. Medical treatment comprises discontinuation of oral feeding, intravenous fluids and parenteral nutrition, gastric lavage and N-acetyl-cysteine in order to encourage disintegration of the bezoar [4]. Sometimes, lactobezoar may result in gastric outlet obstruction and complications such intestinal pneumatosis that require surgery [1,5].

In 2013 we saw seven cases of this rare neonatal pathology in a time period of just 37 days in our NICU. Four of the affected neonates required surgery and one died. This was a notable event not only for the severity of the outcomes and the rarity of such a cluster, but also that it was the first time we had seen this pathology in our unit since it opened in 1990, despite the fact that there had been no changes to our enteral feeding protocols for four years.

The aim of our study was to analyze each case individually to look for predisposing factors, as well as analyzing the nutritional process to find any anomalies in milk reconstitution or potential sources of contamination, in order to understand the cluster and to prevent further cases.

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A retrospective analysis was performed for all cases of GLB in our unit. Inclusion criteria for the analysis were defined as any patient with an abnormal radiological picture consistent with GLB, as well as one of the following: gastric perforation, necrotizing enterocolitis (according to

Table 1: Cases of gastric lactobezoar: potential predisposing factors.

GA	Type of diet	Type of Milk	Duration PF (d)	Caloric density (Kcal/100 ml)	ml/kg/d at diagnosis	Duration MCT+PF (d)	Surgery
27	C	PF+HM	5	87	153	9	Y
30	C+D	PF	9	86	129	15	Y
33	D	PF	9	80	145	0	Y
36	C	PF+HM	5	87	153	4	Y
32	C + D	PF+ HM	3	80	120	2	N
33	D	PF	9	80	112	0	N
35	D	PF + HM	3	80	160	0	N

GA: Gestational Age; C: Continuous enteral feeding; D: Discontinuous enteral feeding; PF: Premature formula; HM: Human Milk; MCT: Median Chain Triglyceride; Y: Yes; N: No; d: Day; Kcal/100 ml: Kilocalories per 100 milliliters; ml/kg/d: milliliters per kilogram per day

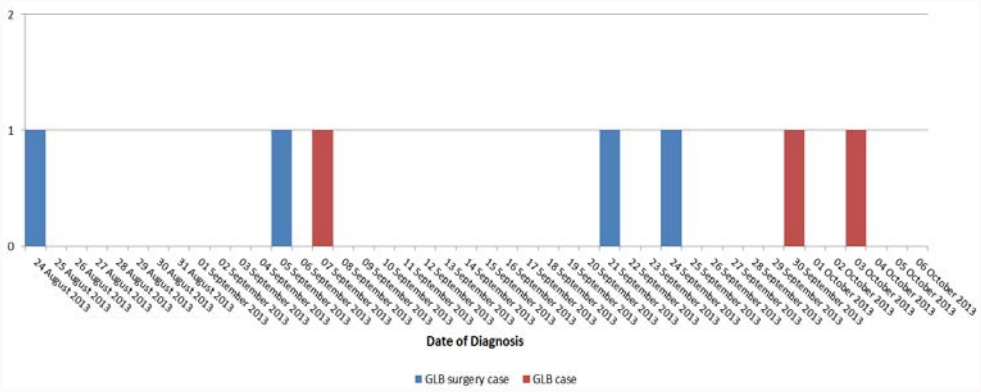

Figure 1: Date of Gastric Lactobezoar's Diagnosis.
GLB: Gastric Lactobezoar

the modified Bell's stage) [6]. We only included neonates that were hospitalized in our NICU (Fort de France, Martinique and French West Indies) between 1st July 2013 to 3rd October 2013. We collected the following data for each patient: gestational age, birth weight and size, mode of delivery, neonatal resuscitation, type of nutrition (parenteral or enteral) at the time of diagnosis, abdominal signs at diagnosis (regurgitation, emesis, abdominal distension), and the need for surgical intervention. We also collected data on some possible predisposing factors: whether enteral feeding was continuous or discontinuous, the type of milk used enteral supplementation, milk composition, use of MCT, discrepancy between the milk composition and medical prescription, possible sources for contamination, and bacteria cultured from the milk or patients. Descriptive statistics comprising counts and proportions, means, medians, were done as appropriate with Excel 2013. IRB approved the study.

Results

Description of cases

Seven cases were diagnosed in a 37 day period, from 24th August to 3rd October 2013 (Figure 1). The affected neonates were all male and premature, ranging from extremely premature to moderately premature (mean: 32.3 GA; 27 to 36 GA). We did not find that the mode of delivery was a risk factor: four of the neonates were born by vaginal delivery and three by caesarean-section. Two of them required immediate resuscitation, both recovering within a few minutes. On average the diagnosis of GLB was made 18 days postnatally (minimum 5 days, maximum 56 days); the first signs appeared 2.4 days before diagnosis and commonly included abdominal distension, regurgitation and/or vomiting of curd milk (in 5 cases, 71%). X-ray imaging was performed in every case, showing a "sandwich loaf" image which is pathognomic for GLB (Figure 2). No gastric ultrasounds were performed because the X-ray results were sufficiently convincing. In terms of treatment, we initially used a medical approach: enteral nutrition was stopped and parenteral nutrition commenced, and we performed gastric lavage with N-acetyl-cysteine. Antibiotics were used in four cases due to suspicion of abdominal sepsis mimicking necrotising enterocolitis. These cases all required surgery as they did not respond despite best medical management. The diagnosis of GLB was confirmed during these surgeries by macroscopic observation (Figure 3 and 4). The least premature baby (36 GA) died of multiple organ failure a few days after surgery.

Potential predisposing factors

All the affected neonates were fed before their diagnosis by continuous or discontinuous enteral nutrition using reconstituted milk powder (premature formula, PF); two of them had breastfeeding in complement. In addition, four of them received MCT enterally (Liprocil®). We analyzed the nutritional composition of the milk given to all babies in the unit and found no discrepancies between the milk given and the milk prescribed. The typical composition of the PF milk for the neonates on our unit was (by % of total mass) casein 14.5%, total protein 15.8%, with a protein/casein ratio of 0.069 and a sodium content of 0.39 d/100 ml. The diagnosis of GLB was made in these babies whilst the volume of their enteral feeds was being increased, occurring on average when the volume reached 130 ml/kg/day. Three out of the four babies who underwent surgery were fed with more than 80 kcal/kg/day with PF supplemented by MCT (Table 1). All of the bacterial cultures taken from milk bottles and the affected children's secretions or blood were negative. We did not identify any sources of contamination in the neonatal unit.

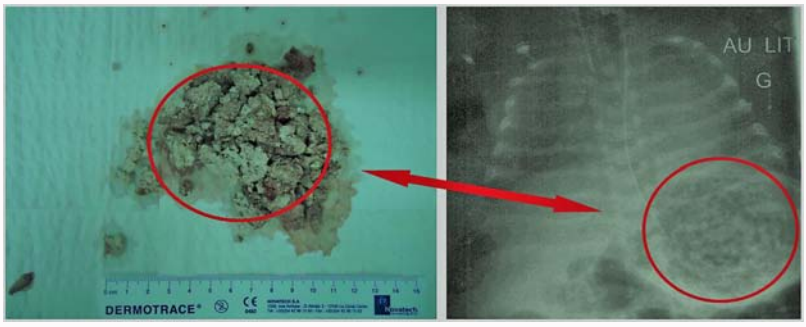

Figure 2: Macroscopic view of lactobezoar and his X-ray picture.

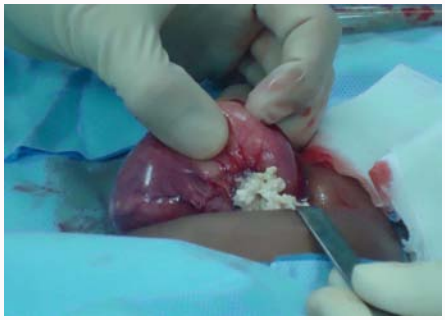

Figure 3: Surgical aspect of lactobezoar with gastric perforation.

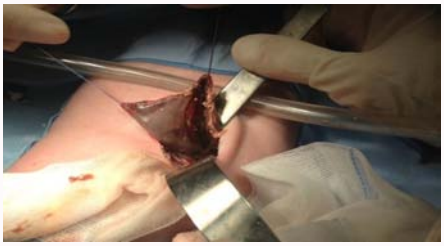

Figure 4: After lactobezoar's removal, visualization of ischemic stomach tissue.

Discussion

This is the first publication describing a GLB cluster. As it is a rare pathology it usually occurs as an isolated case, in different units and with a long timeframe which introduces many variables to consider. It was for this reason that we decided to investigate the cluster to understand what could be causing it, make changes to our protocols and stop this cluster as soon as possible. The death of one of our patients demonstrates the highly dangerous nature of the condition. It can be difficult to make a diagnosis of GLB because the clinical signs are commonplace in neonates, and many occur in perfectly healthy babies with no underlying pathology. However, early suspicion of GLB is imperative, as the pathology is severe and requires timely management, and the more severe signs that lead to diagnosis may be preceded by subtler clinical signs. The presence of both abdominal distension and vomiting/regurgitation of curd milk should indicate a possible diagnosis of GLB and should be investigated with imaging such as abdominal X-ray or ultrasound.

The severity and rare nature of the disease and the unusually high number of patients with the condition in our unit in a very short time frame required us to do some detective work to ascertain the most likely culprit. Due to the clinical presentation and the frequency of sepsis amongst the cases, we looked for a bacterial sepsis or contamination of the enteral nutrition that could have led to the formation of a GLB, but were unable to isolate any germs or likely sources of contamination. We then collected data from all the cases on all previously documented predisposing factors described in medical literature, in order to find out if one or more factors could be implicated in the onset of this cluster. Neither dehydration nor were electrolyte disturbances associated. However, we did find a correlation between other classical factors such as prematurity and use of Premature Formulas and MCT [7]. Interestingly, the more severe cases that required surgery were those fed with a combination of PF and MCT, giving a caloric density higher than 80 kcal/100 ml.The rapid increase of high calorie enteral nutrition in the context of prematurity and immature gastrointestinal function could probably explain the sudden onset of this unfamiliar pathology in our unit.

To make sure that there was no other explanation; we tracked a potential manufacturing error by precisely analyzing milk composition and eliminated this as a potential cause. An expert assessment of our enteral nutrition protocols didn't show any difference between our protocols and international recommendations (European Society for Pediatric Gastroenterology Hepatology and Nutrition (ESPGHAN)) [8]. They did however hypothesise that the combination of PF and MCT (Liprocil®) could have led to a saponification reaction favoring lactobezoar formation.

We favored this approach to enteral nutrition because in Martinique, as in many other parts of the French West Indies, many children are born with fetal growth restriction due to placental insufficiency [9]. Thus, to improve their growth and future development, they are fed with an aggressive strategy, and enteral nutrition is introduced as soon as possible to avoid the risks of central line infection associated with parenteral nutrition [10]. Despite the risks associated with poor growth, the severity of GLB is such that we decided to change our enteral nutrition protocols. Since the cluster, we have modified our strategies, limiting enteral nutrition to <80 kcal/100 ml, prohibiting the combination of MCT and PF, and using pasteurized maternal milk for all premature neonates born at <32 GA or weighing <1500 g.

Since introducing these changes, we have not had any further cases of GLB. To evaluate the impact of our new enteral nutrition protocol on the neonates in our unit, we are currently carrying out a study to analyze their growth and track any adverse effects.

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Institutional Review Board Statement

The institution research ethics board approved the study procedures.

Informed Consent

The need for informed consent was waived given the non-interventional study design by the Institutional Review Boards.

Data Sharing Statement

The data set is available from the corresponding author-Olivier Fléchelles.

References

1. Bos ME, Wijnen RMH, de Blaauw I. Gastric pneumatosis and rupture caused by lactobezoar. Pediatr Int. 2013;55(6):757-60.
2. Heinz-Erian P, Gassner I, Klein-Franke A, Jud V, Trawoeger R, Niederwanger C, et al. Gastric lactobezoar - a rare disorder? Orphanet J Rare Dis. 2012;7:3.
3. Vallabhaneni P, Mansour M, Hutton K, Banerjee S. Lactobezoar-not so bizarre! Arch Child Fetal Neonatal Ed. 2011;96(2):F127.
4. Heinz-Erian P, Klein-Franke A, Gassner I, Kropshofer G, Salvador C, Meister B, et al. Disintegration of large gastric lactobezoars by N-acetyl-cysteine. J Pediatr Gastroenterol Nutr. 2010;50(1):108-10.
5. Gambart M, Breinig S, Breton A, Vial J, Herbault-Barres B, Bouali O. [Gastric perforation caused by a lactobezoar in an infant: a case report]. Arch Pediatr. 2012;19(9):927-31.
6. Walsh MC, Kliegman RM. Necrotizing Enterocolitis: Treatment Based on Staging Criteria. Pediatr Clin North Am. 1986;33(1):179-201.
7. Prahl M, Smetana D, Porta N. Lactobezoar formation in two premature infants receiving medium-chain triglyceride formula. J Perinatol. 2014;34(8):634-5.
8. Agostoni C, Buonocore G, Carnielli VP, De Curtis M, Darmaun D, Decsi T, et al. Enteral Nutrient Supply for Preterm Infants: Commentary From the European Society of Paediatric Gastroenterology, Hepatology and Nutrition Committee on Nutrition. J Pediatr Gastroenterol Nutr. 2010;50(1):85-91.
9. Saunders L, Guldner L, Costet N, Kadhel P, Rouget F, Monfort C, et al. Effect of a Mediterranean diet during pregnancy on fetal growth and preterm delivery: results from a French Caribbean Mother-Child Cohort Study (TIMOUN). Paediatr Perinat Epidemiol. 2014;28(3):235-44.
10. Boutaric E, Gilardi M, Cécile W, Fléchelles O. [Impact of clinical practice guidelines on the incidence of bloodstream infections related to peripherally inserted central venous catheter in preterm infants]. Arch Pédiatr. 2013;20(2):130-6.