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Case Report

Perforated Bowel after Obstruction in a Neonate with Milk Curd Syndrome following Ready to Feed Infant Formula

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ABSTRACT

Milk Curd Syndrome (MCS) is a known iatrogenic disease and an unusual and uncommon cause of neonatal intestinal obstruction, particularly in premature babies. MCS is caused by feeding infant formula rich in calcium and fat. The disease is known for its morbidity and death rate particularly when the disorder is not diagnosed in good time. The overall incidence has decreased because of improvement in infant feeding formula preparations and feeding practices. We are reporting a case of MCS in a premature baby who was fed a newer brand of ready-to-feed infant formula. The baby required surgical intervention for perforation of his large bowel and survived without complications.

1. INTRODUCTION

Constipation is not an uncommon disorder in infants. In neonates, "constipation" may be caused by intestinal obstruction, which could be congenital or iatrogenic. Milk Curd Syndrome (MCS) is a rare iatrogenic form of infantile intestinal obstruction because of inspissated milk. The condition is potentially preventable by using better feeding formulas and feeding techniques. MCS must always be suspected in formula-fed babies who have prolonged constipation. The condition is curable with morbidity-free survival after surgical intervention. In this report, we present a case of transverse colon perforation caused by MCS in a premature baby with morbidity-free survival after surgical intervention.

2. CASE REPORT

A premature male baby born at 33 weeks' gestation by vaginal delivery with a birth weight of 1.37 kg, received noninvasive ventilation for moderate respiratory distress for 5 days. Nasogastric tube feeding, using a 24 kcal preterm Ready-to-Feed (RTF) formula, was started on day 5 of life, and the feeding volume was gradually increased as per standard feeding protocol. The feeding was frequently interrupted by episodes of feeding intolerance with large gastric aspirates, which was sometimes greenish in color. Full feeding was achieved on day 21 of life. On day 28, the baby developed an episode of clinical sepsis with C-reactive protein gradually rising to 177. The associated respiratory deterioration required ventilation. On day 32, the abdominal X-ray showed free peritoneal gas. An urgent laparotomy was performed. The transverse colon had a perforation with curd milk inside the lumen and in the peritoneum (Figure 1). The transverse colon was resected with end-to-end anastomosis. A diverting ileostomy was done. Full oral feeds were gradually built up using elemental formula. The ileostomy was closed after 3 weeks, and the baby was discharged home with a normal growth rate.

3. DISCUSSION

Milk curd syndrome or inspissated milk syndrome, also called lactobezoar, was first recognized in 1959 [1]. Its radiological features were first reported in 1970 by Cremin et al. [2]. MCS, an iatrogenic feeding disorder caused by formula feeds rich in calcium and fat, is an unusual cause of neonatal intestinal obstruction. The incidence was high during 1970s, the era during which infant feeding with formula feeds was common, and milk powders were prepared by drying cow's milk. The incidence remained high until 1980 [3], and 70 cases were described in the world literature by then. The incidence declined drastically after 1980 as a result of better understanding of neonatal physiology, neonatal nutrition, and better preparation of infant formulas containing low Ca and fats [4]. Lohn et al. [3] described 43 cases of lactobezoar up to 1980 but only two cases in the next two decades. Since 2000, there has been a resurgence in the incidence of MCS, probably because of the increasing number of low-birth-weight babies using formula feed [5]. Although the majority of described cases were attributed to cow's milk-based infant formula, a case of MCS attributed to goat's milk has also been described [6]. MCS has also been described with high caloric fortified expressed breast milk [6]. Our patient was fed one of the newer brands of RTF high caloric preterm formula (24 kcal/oz). We did not find any other case in the literature associated with feeding RTF formula. Ours is probably the first case. The calcium, phosphate, and fat content of RTF is similar to that of the standard powder form of preterm formula. However, what is not clear is how the companies have

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Figure 1 | Intraoperative photograph of inspissated milk in perforated transverse colon.

ensured the stability of RTF at room temperature (not exceeding 25°C) at which it can be kept for 1 year as written on each bottle. The manufacturer may be adding some kind of stabilizer to achieve this target. This is a new area that needs to be explored and which, we hope, our case will highlight.

The incidence of MCS is slightly higher in males than in females. The incidence was higher in term babies in the earliest reports [1,3]. However, in subsequently described reports, nearly all cases were in premature babies in whom high calorie feeds were initiated soon after birth [7,8]. Our case was also a preterm baby fed on a high calorie formula. The usual clinical presentation of MCS is with features of small bowel obstruction, that is, bilious vomiting and abdominal distension after the neonate has passed through the stages of passage of normal meconium followed by transition stools and normal milk stools. Our patient had a long period of "feeding intolerance," a clinical feature that otherwise is very common and nonspecific among premature babies. Our patient had obstruction in the transverse colon. The peak incidence of MCS occurs between 5 and 14 days of life [4]. Our patient was diagnosed on day 32 during a laparotomy for an accidental and unexpected finding of free peritoneal gas on abdominal X-ray, which was done for cardiorespiratory instability and investigation for "clinical sepsis." Reports of MCS presenting as intestinal obstruction, gastric perforation, cecal perforation and necrotizing enterocolitis have been published [4,8]. The cecum is the most common site of large bowel perforation due to MCS [4]. Our patient had perforated transverse colon, which has also been described in the literature [4].

The differential diagnoses are meconium ileus and total colonic aganglionosis. Abdominal X-ray usually shows the characteristic features of air intermixed with stools in the distal small bowel with proximal distended bowel loops. Like meconium ileus, there is scarcity of air fluid levels on abdominal X-ray [4]. Hyperechoic masses may be seen on ultrasound. A contrast enema should be done, both for diagnostic as well as therapeutic purposes. The enema should enable clinicians to distinguish MCS from the microcolon found in ileal atresia or proximal colonic atresia.

In MCS, a trial of gastrografin enema may be both diagnostic as well as therapeutic [4]. Watanabe et al. [9] successfully treated a preterm baby with MCS using olive oil enemas and successive colonic lavage for 3 days. In general, the conservative methods fail and surgical intervention becomes imperative [5]. The terminal ilium is the most common site of inspissated bolus, sometimes extending into the colon. Another common site is the mid-ileum. During laparotomy, it may be possible to break up the bolus through the bowel wall and milk it through the ileocecal valve into the colon [7]. This is possible only in cases that are diagnosed early. Milking can be assisted by intraluminal injection of isotonic saline. However, many cases require enterotomy and removal of the inspissated milk bolus. Bowel resection with or without diverting ileostomy have also been required, particularly in cases with perforation [7]. Our case presented with perforation and required bowel resection, end-to-end anastomosis, and a diverting ileostomy, which was closed 3 weeks later. The long-term prognosis is generally very good. A high index of suspicion in high-risk babies (prematurity, high caloric infant formula, clinical features of small bowel obstruction) can help clinch an earlier diagnosis with early intervention and better outcomes.

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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