Lactobezoar and Necrotizing Enterocolitis in a Preterm Infant

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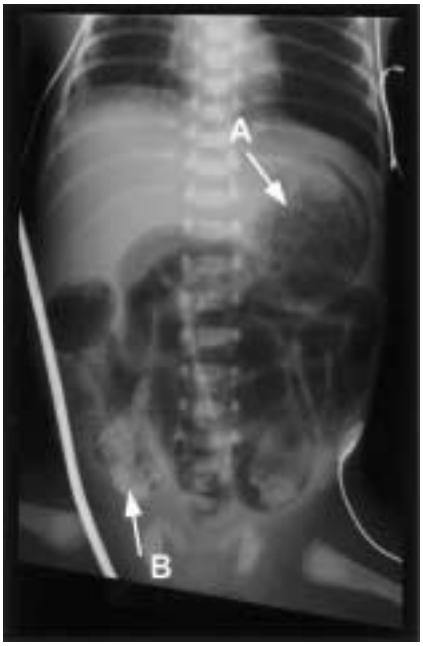
Necrotizing enterocolitis occurs in a sizable proportion of preterm infants. In a report from the National Institute of Child Health and Development Neonatal Research Network, 7% of very low birth weight infants (<1,500 g birth weight) suffered from proven NEC [1]. Despite many theories on the pathogenesis of NEC, the mechanism(s) by which preterm infants develop NEC are still unclear and are probably multifactorial. Another gastrointestinal complication of prematurity is the formation of lactobezoars [2]. To the best of our knowledge, and according to a Medline search including the key words "lactobezoar," "bezoar" and "necrotizing enterocolitis," the coexistence of lactobezoar and NEC has not been reported previously. We present here the case of an infant who presented with both lactobezoar and NEC, and discuss the possible relationship between the two entities.

Patient Description

A 29 week gestation male infant was delivered at the Lis Maternity Hospital at the appropriate birth weight for gestational age of 1,230 g. Maternal history was unremarkable, although the pregnancy was complicated by urinary tract infection and spontaneous preterm labor. The infant was delivered in a spontaneous vaginal vertex fashion. The immediate post-delivery course was complicated by respiratory distress syndrome (managed by high frequency oscillatory ventilation and exogenous surfactant administration) and patent ductus arteriosus (which was closed successfully after one course of indomethacin). The infant developed pulmonary interstitial emphysema, with subsequent moderate chronic lung disease necessitating ventilation until day 21 of life. The infant was started on feeds on day 5 of life, reaching 120 ml/kg weight of human milk and diluted Similac Special Care (12 cal/oz) (Ross Products Division, Abbott Laboratories, Columbus, OH, USA) within 5 days.

On day 10 of life he developed significant abdominal distension. Abdominal anterior-posterior X-ray [Figure] revealed a grainy gastric mass, outlined by air (arrow A), compatible with lactobezoar. It also revealed extensive intestinal pneumatosis (arrow B) and distended bowel, characteristic of necrotizing enterocolitis. Repeated gastric lavage with 0.9% NaCl solution resulted in suctioning of undigested milk curds and, radiographically, in successful evacuation of the bezoar. Three weeks later, after reintroduction of enteral feeding, the infant developed intestinal obstruction that presented as an abdominal mass on palpation, abdominal wall hyperemia and edema. During surgery, intestinal perforation was diagnosed that necessitated resection of a long portion of the small and large bowel. The infant died shortly after surgery. Postmortem examination confirmed the diagnosis of NEC, and the histologic examination of the specimen obtained during surgery revealed extensive necrotizing enterocolitis with multiple perforations.



Abdominal anterior-posterior X-rays showing a grainy gastric mass, outlined by air (arrow A) compatible with lactobezoar, and extensive intestinal pneumatosis (arrow B) and distended bowel, characteristic of necrotizing enterocolitis

Comment

The term bezoar is derived from Persian (meaning antidote for poisons), and refers to a gastrointestinal foreign body. Lactobezoar (undigested milk curds) was first described in 1959 [3] and is the most common type of bezoars in infants as compared to other bezoar types – trichobezoars (hair matter), phytobezoars (vegetable matter), and trichophytobezoar (combination of the two) [2,4]. Infants with a lactobezoar may be asymptomatic or may produce a wide range of gastrointestinal symptoms and findings, including feeding intolerance. The most common gastrointestinal symptoms are abdominal distension, gastric residuals, emesis and bloody stools, which may mimic necrotizing enterocolitis. The pathogenesis of lactobezoars is unclear. Several possible contributing factors have been suggested. The majority of lactobezoars has been described in preterm infants who were fed "preterm formulas" with high casein/whey ratios, protein content, calcium, and med-

NEC = necrotizing enterocolitis

ium chain triglycerides. The role of a high casein/whey ratio has been challenged by the fact that lactobezoars have been described also in infants who were fed whey-dominant formulas or even human milk. This was the case in our patient since he received human milk and a whey-dominant "preterm" formula. Also, as we reported previously, gastric emptying time of whey-dominant and of casein-dominant formulae are almost identical. Physiologic factors that may contribute to the formation of lactobezoars in preterm infants include delayed gastric emptying and decreased gastric acid output. Nevertheless, the common denominator of most reported cases or mini-series of lactobezoars is that they occur almost exclusively in preterm infants [2–4].

On the basis of our Medline search, the coexistence of lactobezoar and necrotizing enterocolitis has not been reported previously. In theory, if the two conditions are associated, it may be through various mechanisms: if the bezoar preceded NEC, it is possible that intestinal distension due to obstruction, induced by fragments of the bezoar that migrated into the intestine, may have contributed to ischemic lesions of the intestinal mucosa. Alternatively, if NEC preceded the lactobezoar, it may have done so through paralytic ileus resulting in the presence of undigested clotted milk in the stomach that may have facilitated the lactobezoar formation. Since both conditions coexisted on the first abdominal X-ray, we can only speculate about the chain of events.

Also, in theory, one may wonder whether the intestinal perforation that occurred later in the course of the disease may have been related to the lactobezoar, as published in one report [5]. This is not very likely, since in our case extensive *intestinal* necrosis and perforation was observed on pathologic examination, while in the case reported by Levkoff et al. [5] the perforation was punctual, *gastric* and may have been a complication of gastric lavage. We conclude that lactobezoars and NEC may coexist and are possibly causally related.

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