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Ceacal perforation in a premature newborn infant complicating milk curd syndrome: Case report

Milk curd sendromu'nun komplikasyonu olarak prematür yenidoganda çekum perforasyonu: Olgu sunumu

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A 34-weeks-old female baby having a closed ceacal perforation due to milk curd syndrome is reported. The obstructing milk curd was located in the transvers colon and the perforation was in the ceacum. Primary repair of the ceacal perforation, aspiration of the material from an enterotomy and a protective ileostomy were performed. The patient died in the second postoperative day as a result of cardiopulmonary arrest and septic shock.

Key words: Milk Curd Syndrome, newborn, intestinal perforation

Milk Curd Sendromu'na bağlı olarak kapalı çekum perforasyonu gelişen 34-haftalık kız bebek sunulmaktadır. Tıkayıcı süt kalıntısı transvers kolonda, perforasyon ise çekumda saptandı. Operasyonda, perfore alan primer olarak tamir edildi. Enterotomi yapılarak tıkayıcı süt kalıntısı aspire edildi ve koruyucu ileostomi açıldı. Hasta operasyon sonrası ikinci günde septik şok ve kardiopulmoner arrest sonucu kaybedildi.

Anahtar Kelimeler: Milk Curd Sendromu, yenidoğan, intestinalperforasyon

INTRODUCTION

Milk curd syndrome, known as lactobezoar or inspissated milk syndrome is a relatively common disorder, which is first described, by Wolf and Bruce in 1959 (1). The condition has been reported for gastric outlet obstruction and perforation in stomach, terminal ileum and Meckel's diverticulum (1-7). Ceacal perforation has not been mentioned up-to-date.

CASE REPORT

A 34-weeks-old and low birth weight (1460 g) premature female baby was admitted to the premature ward of our hospital in the first day of her life. She passed meconium in the first 24 hours and was started with oral feeding with a low-birth-weight infant formula fortified with medium-chain triglycerides and glucose polymers (Similac 24 LBW) at 3 days of age. Oral feeding was continued for several days and normal milk stool was observed on days 5, 8, and 9. The patient didn't pass stool for the consecutive 3 days and on day 12 she rejected oral feeding. Abdominal distension occurred and a solid mass became palpable at left upper and right lower quadrant. Abdominal wall tenderness and erythema were other physical examination findings at the time we consulted the patient. She was transferred to surgical intensive care unit in a septic state, with low platelet counts, high total bilirubin levels and failure to thrive. Plain abdominal radiography revealed a decline in the gas extent and "soap bubble" appearance in the right lower quadrant (Figure 1). After preoperative stabilization, explorative laparotomy was performed through a median incision with the indication of closed perforation. The perforation was assumed to occur due to necrotising enterocolitis. Operative findings included subserosal hemorrhagic foci on both small and large intestinal segments and localized dense fibrin collection at the appendicle lodge. There were a 2 cm ceacal perforation and a very dense, sticky and gray-white colored intraluminal material in the mid-portion of transverse colon. The ceacal perforation was primarily repaired and the dense material was aspirated through an enterotomy over the mass. Finally a protective ileostomy was performed.

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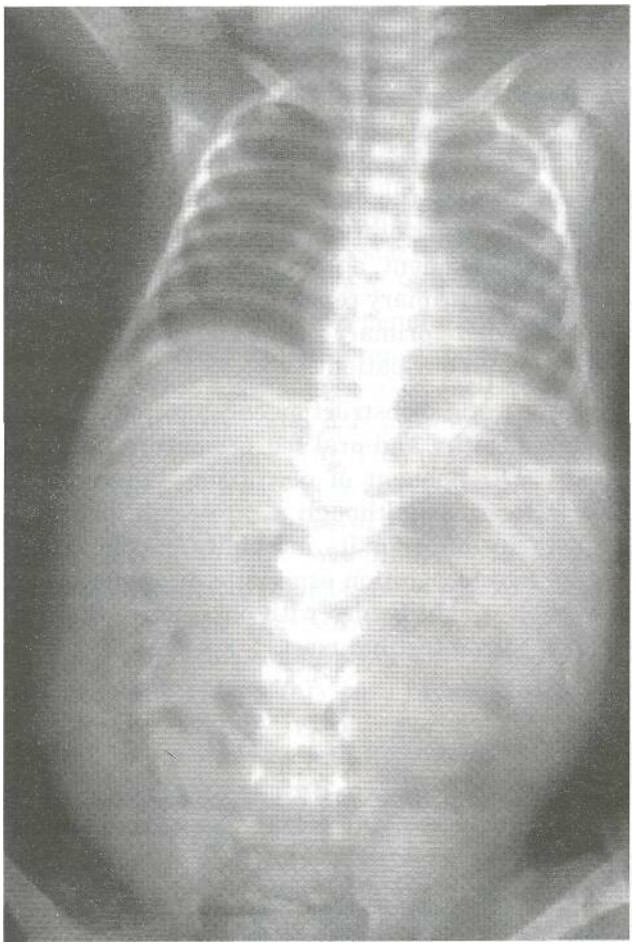

Figure 1. Plain abdominal radiography showed a marked decline in the abdominal gas extent and soap bubble appearance in the lower right quadrant

The postoperative course was eventful with respiratory and septic problems. On the second day of operation the patient died because of cardiopulmonary arrest and septic shock.

DISCUSSION

Lactobezoar formation is an uncommon complication of routine infant feeding. Wolf and Bruce first described this syndrome in 1959, in which a milk coagulum has led to gastric obstruction (1,8). Later on, Cook and Rickham described small bowel obstruction due to inspissated milk in 1969 (2). The term "inspissated milk syndrome" was first used by Cremin et al where a syndrome of intestinal obstruction in premature infants fed on high-calorie powdered milk formula (2). A case of ileal obstruction due to milk curd formation in a giant Meckel's diverticulum and two recent cases of gastric outlet obstruction were reported in 1993, 1997 and 1999 respectively (3, 7, 9). In previous studies, the male to female ratio was given as 5:1 and the overall incidence is about 6% (5, 6).

Typically, the syndrome represents as a distal small bowel obstruction in a male, premature or low-birth-weight infant who has passed first meconium and then milk stools. One half of the patients have passed blood via the rectum. The obstruction generally occurs as early as 2 days and late as 6 weeks of age with a peak of 4 to 10 days (5). In our patient the onset of obstruction is suggested to be day 10.

According to several investigations; although the etiology of lactobezoar formation is based upon the composition of the formula used in oral feeding (1, 4, 5, 10-14), in recent years, especially after 1980's, the syndrome seemed to have multifactorial predisposing factors such as; 1) a trend to earlier and more rapid advancement of feeding, 2) high-caloric-density formulas, 3) continuous-drip feeding techniques, 4) the use of additives like calcium, medium-chain triglycerides, carbohydrates, and vitamins, 5) the physiology of the premature infant including gastric emptying, gastric acidity, and gastrointestinal hormones, 6) low birth weight, 7) respiratory distress, and, 8) cows milk protein intolerance (12, 15). In light of these factors, our patient had the findings of prematurity, low birth weight and probably other additive factors causing lactobezoar formation. High-caloric formulas are accepted to be a significant predisposing factor in the formation of lactobezoar. Similac 24 LBW is such a formula (10) and it was used in our patient.

When biochemically investigated, the coagulum is found to be insoluble in 1N HC1. It dissolves in 1N NaOH. The resulting solution gives positive biuret and ninhydrin reactions. Following acid hydrolysis of the solution, the hydrolysate is ninhydrin positive and biuret negative. On microscopic examination, the coagulum revealed crystals of cholesterol, lactose and triple phosphate. All these findings conclude for the coagulum to be a calcium paracaseinate (1). The biochemical analysis of the aspirated material in our patient was proved to be calcium paracaseinate.

When suspected, inspissated milk obstruction can be diagnosed and treated nonoperatively based on characteristic radiographic findings (5). In plain radiographs, an opalescent intraluminal mass surrounded by a halo of air may be seen or signs of obstruction with air-fluid levels can be detected

but the graphs may be normal at all. In recent years, abdominal ultrasonography has been successful in detecting such a mass leading upper or lower gastrointestinal obstruction (8, 15). Barium contrast studies are also helpful with signs of filling defects and narrowed but no micro colon. In our case, a decline in the abdominal gas extent and a soap bubble appearance was seen in plain radiography as pathological findings.

In management, operative or nonoperative treatment or both are performed. Nonoperative treatment consists of gastrografin enemas. Gastrografin is a hypertonic solution, which can attract three times its volume of water intraluminally. It also contains 0.1% polysorbate, a surface acting emulsifying agent, which decreases the surface tension of the plug. Surgical exploration usually reveals that the most common location of the milk plug$_x$is the terminal ileum (5). Some other forms of reported locations are gastric (1, 3, 9) and Meckel's diverticulum (7). Sometimes the plug involves the proximal small bowel in which enterostomy and acetylsistein irrigation can be used in management. In our case the coagulum was localized in the mid-portion of the transverse colon. When perforation occurs proximal to the obstructed segment, a proximal stoma can be performed with primary repair or resection and anastomosis, where primary repair and ileostomy were performed in our patient.

Gastrointestinal obstruction encountered in a previously healthy and oral fed premature infant is mostly a component of necrotising enterocolitis. But lactobezoar, although a rare condition, must be kept in mind for its morbidity and mortality due to late recognition especially in premature or low birth weight babies with obstructive findings of the gastrointestinal tract.

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