A rare cause of neonatal intussusception. Considering it might reduce the mortality. A case report and a review of the literature

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ABSTRACT

Full-term neonatal intussusception is rare. Cecal duplication as a lead point for intussusception is very rare. We report a case of full-term neonate with ileo-cecal intussusception due to cecal duplication. Although cecal duplication represents rarely a surgical emergency, intussusception always needs immediate attention. The awareness of this entity is essential for diagnosis and management.

1. Introduction

Gastrointestinal duplication is a congenital malformation occurring in the mesenteric side of the bowels [1]. It can occur at any site, from the mouth to the anus. The ileum represents the most common site. Colonic duplications, however, are rare, representing only 13% of cases. Cecal duplication is the rarest [2,3]. It usually presents with abdominal pain and distention, bleeding per rectum, and a palpable mass [1]. In this paper, we present a case of full-term neonatal intussusception secondary to cecal duplication, which has not been described before in the English literature. We aim to draw attention toward a very rare entity because, otherwise, it might be missed and the subsequent complications might be devastating.

2. Case report

A term male neonate, with a birth weight of 3650 g, presented to the neonatal care unit for the management of indirect hyperbilirubinemia and phototherapy. He was born of a nonfollowed pregnancy of a primigravida 25-year-old healthy woman. He tolerated regular feeds and passed meconium within the first 24 hours of life. In addition to jaundice, the physical examination upon presentation was normal. On the fifth day of life, the patient developed acute constipation, followed rapidly by abdominal distention and biliary vomiting. Further, his clinical examination showed tenderness in the right lower quadrant. No abdominal mass was palpable. No blood per rectum was noted. Blood workup showed mild leukocytosis with normal levels of inflammatory markers. Abdominal radiography was performed, showing distended bowel loops and a non-aerated colon, with no evidence of either pneumoperitoneum or pneumatosis intestinalis (Fig. 1). Bowel rest was initiated with a nasogastric tube in place, giving a biliary output. Triple intravenous antibiotics were started.

On the next day, because of the distended bowel loops, echography was attempted but not conclusive. Because the patient was hemodynamically stable and because we doubted NEC, retrograde water-soluble contrast colonography was performed. It showed a filling defect at the level of the proximal ascending colon (Fig. 2). Laparotomy was done and revealed ileo-colic intussusception. Manual reduction had failed. Therefore, resection was done, followed by ileo-colic end-to-end anastomosis. Upon studying the resected segment, a cecal cystic duplication as a lead point for the intussusception (Fig. 3) was found. Postoperative stay was smooth, and the patient was discharged home on the sixth postoperative day.

3. Discussion

Cecal duplication is a very rare congenital anomaly. As other variants of intestinal duplication, it can be associated with obstructive symptoms [4]. Because of its rarity, only 12 pediatric cases of intestinal obstruction secondary to cecal duplication have been reported in the literature [1,3,5-12]. Only two of the 12 were...
Fig. 1. Abdominal radiography showing diffusely distended small intestine with a non-aerated colon and rectum.
Neonatal intussusception, on the other hand, is also very rare, with only 50 cases reported in the literature [13]. It represents 3% of neonatal intestinal obstruction and 0.3% of all cases of intussusception [14,15]. The presentation of neonatal intussusception is vague, misleading, and nonspecific [16]. That is why more than 50% of cases were misdiagnosed as NEC, especially in premature infants [13]. In cases of full-term infants, intussusception is usually associated with a pathological anatomical lead point [15], which was a Meckel’s diverticulum, a polyp, or an intestinal duplication [11].

To the best of our knowledge, no cases were reported describing a full-term neonatal intussusception secondary to cecal duplication. That was the reason why we shared our experience. In our case, although the patient was first misdiagnosed as NEC, this did not delay our correct diagnosis. The fact that the patient never had sepsis, the absence of blood per rectum and the abdominal radiography finding oriented us toward mechanical obstruction [17]. The echography was not conclusive, which is usually the investigation of choice [18]. However, because of the filling defect on retrograde colonography, the diagnosis of a duplication cyst was rapidly thought of. No clear symptoms or signs could have oriented us toward the intussusception in this neonate [15]. Although duplication cysts rarely represent surgical emergency, any delay, however, in operating an intussusception could have severe consequences such as ischemia, perforation, and peritonitis. That is why we need to lower our threshold to not to miss an easily correctable malformation early one.
Because the duplication cyst shares a common wall with the adjacent intestinal segment and because it is located on its mesenteric side, segmental excision of the involved segment of the bowel with end-to-end anastomosis is the surgical procedure of choice, when possible [19].

4. Conclusion

This is a case of full-term neonatal intussusception secondary to a cecal duplication cyst. Lowering the diagnosis threshold helped us reduce the mortality and morbidity.

Ethical statement

I declare that there are no ethical issues pertaining to this paper.

Declaration of competing interest

I declare that there is no conflict of interest.

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