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A rare case report of a neonatal idiopathic intussusception in a full-term newborn

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ABSTRACT

INTRODUCTION: In contrast with the usual idiopathic intussusception appearing in infants, neonatal intussusception in full-term newborns is a rare entity and usually due to an organic lead point lesion. It has a misleading and variable presentation. This manuscript reports a very rare case of neonatal idiopathic intussusception in a full-term male newborn in order to highlight the difficulties in establishing an early diagnosis.

PRESENTATION OF CASE: We present a full-term male newborn who was referred to our department at day 7 of life with fecaloid vomiting, distended abdomen and absence of intestinal transit. No bloody stool was identified. The newborn was in poor condition, dehydrated and hypothermic. He was promptly resuscitated and an abdominal ultrasound was quickly performed, it showed an ileocecal intussusception located in the right hypochondriac region. An emergency laparotomy was performed confirming the ultrasound findings. The pathological exam of the resected bowel confirmed the presence of an intussusception with areas of hemorrhagic rearrangements but no lead point was detected. The patient died from septicemia.

CONCLUSION: Intussusception occurring in the neonate is still difficult to assess, because of its rarity and its uncommon mode of presentation. This case report underlines the importance to make early diagnosis, because once a critical condition develops, the mortality rate is likely to rise.

1. Introduction

Intussusception is one of the most common causes of intestinal obstruction in children it is found most frequently in infants from 6 to 18 months. However, during the newborn period, it is an exceedingly rare clinical entity. It accounts for only 3% of intestinal obstruction causes in neonates and represents only 0.3% of all cases of intussusception [1] and it's usually due to an organic lesions.

The unusual age group and variable presentation explain the challenge of suspecting this disease in newborns and contribute to the frequent delay in diagnosis. Early diagnosis may be enhanced with abdominal ultrasonography to reduce the morbidity and mortality.

We present a very rare case of neonatal idiopathic ileocecal intussusception diagnosed on the seventh day of birth in the department of pediatric surgery B and medical imaging department at Children's hospital of Tunis. The report has been arranged in line with SCARE guidelines [2].

2. Presentation of case

A 3300 g, full-term male was born at 38 weeks of gestation by normal spontaneous vaginal delivery. Pregnancy was uneventful. Prenatal ultrasound, neonatal physical examination at birth, and the first meconium passage were normal. He tolerated oral feeding and had daily defecation.

On the fifth day of life, he began to have bilious vomiting and progressive abdominal distention; Consequently, enteral feeding was discontinued. He was referred to our department at day 7 of life with fecaloid vomiting, distended abdomen and absence of intestinal transit. The newborn was in poor condition, dehydrated and hypothermic. A nasogastric tube was inserted and fecaloid gastric residuals came out. There was no palpable mass and no bloody stool was identified on rectal examination.

The baby was intubated immediately due to respiratory distress and was admitted to the neonatal intensive care unit. An abdominal ultrasound was performed. The ultrasound showed small bowel

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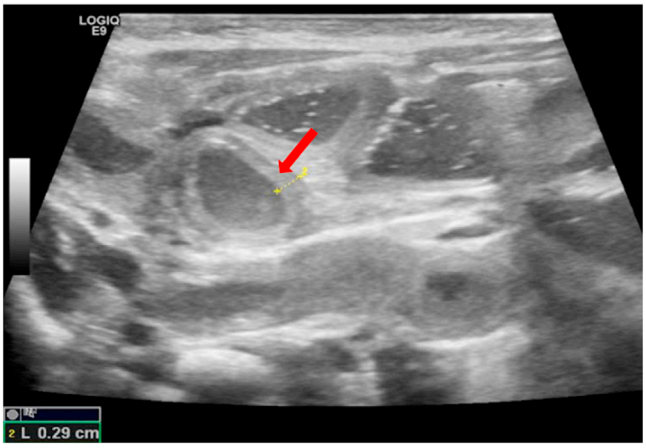

Fig. 1. Ultrasound images showing a small bowel distension and bowel wall thickening (red arrow) upstream the intussusception.

distension and diffuse bowel wall thickening (Fig. 1) upstream a sub hepatic ileocecal intussusception. The latter showed the classic target and pseudo kidney signs extending on 35.5 mm without blood flow within it (Fig. 2). There was a diffuse mesenteric fat infiltration without any intra-abdominal fluid nor an abdominal mass. No mesenteric malrotation was found. The abdominal ultrasound did not conclude to any other abnormality.

Complete blood count indicated anemia with thrombocytopenia and mild leukocytosis. Blood biochemistry showed hyponatremia hyperkalemia and renal impairment. After promptly resuscitation, an emergency laparotomy was performed. The access to the abdominal cavity was obtained through a midline upper abdominal incision. Exploration revealed dilated small bowel upstream of a five-centimeter-ileocecal intussusception (Fig. 3), that was reduced with retrograde manual pressure. There were no congenital anomalies, no ischemia or necrosis. The palpation of a focal thickening of the cecal wall required the resection of 3 cm of terminal ileum and the cecal segment (Fig. 4) followed by an ileocolic end-to-end anastomosis. The remaining bowel was normal.

The pathologic study confirmed the presence of an intussusception with areas of hemorrhagic rearrangements within the ileal wall. No lead point was detected and there was no proof of concomitant infection.

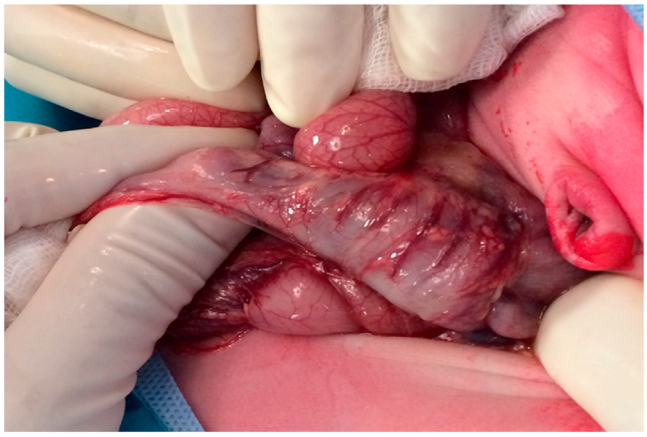

Fig. 3. A five-centimeter-ileocecal intussusception.

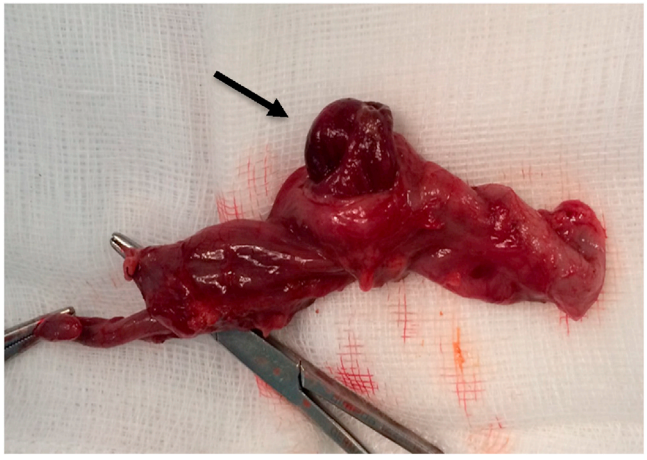

Fig. 4. Resection of 3 cm of terminal ileum and cecal segment with a suspected hemorragic lesion of the cecum (black arrow).

The infant was followed up in the neonatal intensive care unit. The post-operative period was stormy: The child succumbed on the third post-operative hour to septicemia, persistent hypotension and multiple organ dysfunction syndrome.

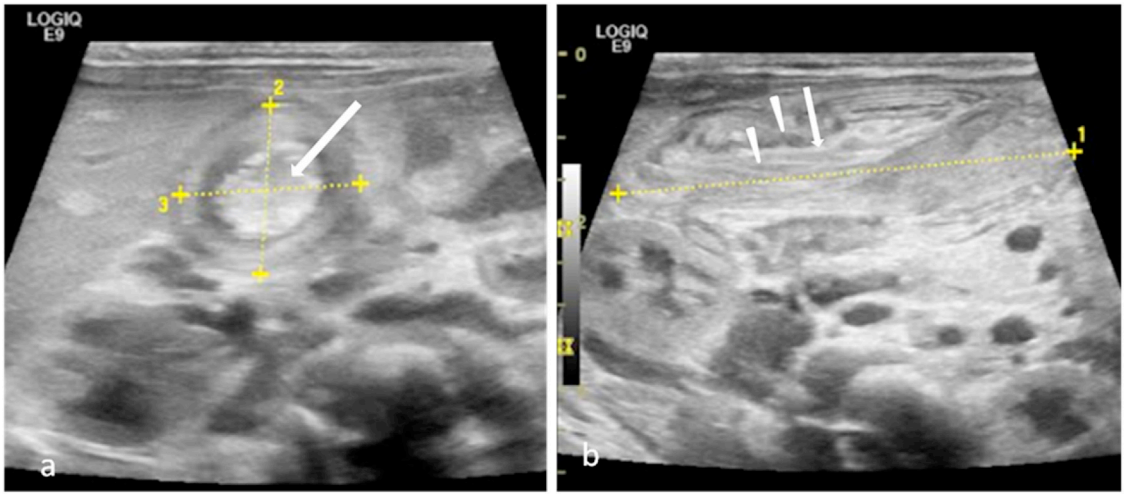

Fig. 2. a: Ultrasound transverse view showing a target sign consisting of a peripheral hypoechoic ring with central echogenicity (white arrow) measuring 19 mm × 19.8 mm. b: Ultrasound longitudinal view showing a pseudo kidney sign consisting of a hyperechoic mesenteric fat (white arrow) contained between an external cecal wall and an internal ileal wall (white arrowheads) extending on 35.5 mm.

3. Discussion

Intussusception is the invagination of an intestine segment within a more distal one. It is the most common cause of bowel obstruction in children [3]. Intussusception is extremely rare in neonates; it represents only 3% of all cases of neonatal intestinal obstruction. In a 150-year review of the world's literature by Rachelson et al., in 1955, reported that intussusception in the 1 st month of life had an incidence ranging from 0% to 2.7% with an average incidence of 0.3% [4].

The etiology of intussusception remains unknown in the majority of cases. Many reports suggest that full term intussusception shows a high frequency of organic causes [5]. Avansino et al. reported lead point lesions in 10–58% of cases in term newborns [6]. The reported causes in literature are duplication cyst, hamartoma, Meckel's diverticulum and mesenchymoma [6,7].

Cases of congenital infantile fibrosarcoma [8], jejunal atresia [7], cecal duplication cyst have been reported to cause intestinal intussuscepton in full term babies. Neoplasms remain exceptional [8]. Our case seems to be exceptional as it reports an idiopathic neonatal intussusception in a full term male boy. In fact, operative exploration as well as pathological exam of the resected bowel specimen did not show any lesion or tumoral tissue nor an abnormality of the bowel wall rather than hemorrhagic rearrangements due to its compromise after a long lasting intestinal obstruction. Neonatal infection has been reported as cause of this pathology [6,9]. However, in our report, no proof of infection was observed.

To our knowledge, this is the first report of idiopathic ileocecal intussusception appearing in a full-term newborn.

Intussusception occurring in the neonate is still difficult to assess. This is partly because of its rarity and because of its uncommon mode of presentation. Yoo and Touloukian [10], reported in an analysis of 22 cases that the classic tetrad commonly found in older infants with idiopathic ileocolic intussusception is replaced by signs of intestinal obstruction. Rectal bleeding, though present in 75% of cases, often occurs late. A palpable abdominal mass is less often found and irritable crying is rare.

Because symptoms might be misleading, early diagnosis may be enhanced with abdominal ultrasonography to reduce the morbidity and mortality [11]. This imaging mean was first described for the diagnosis of this disease in 1980 [12]. It is the best imaging modality thanks to its lack of radiation, its availability, its low cost and mainly its high sensitivity, specificity and negative predictive value respectively estimated at 97.9%, 97.8% and 99.7% [13].

Ultrasonography shows, as exactly reported in our case, a target sign on transversal view and a pseudokidney sign on longitudinal view [14,15]. In our case, ultrasound was considered as a first screening test rather than an abdominal radiography. This diagnostic strategy seems to agree with the most recent reports. In fact, abdominal radiography is controversial in the diagnosis of intussusception and should be used before enema reduction to exclude a pneumoperitoneum but not as a first screening test [14]. The Doppler use during ultrasound exam might be useful. In our case, no Doppler flow was found within the intussusception. According to Lim et al., this feature correlates with bowel ischemia and predicts necrosis [16]. This agrees with our pathological test on surgical specimen showing hemorrhagic changes revealing a bowel wall compromise.

Although previous reports indicate the value of ultrasonography and contrast enema in early diagnosis, in the majority of the cases, a definitive diagnosis is made at surgery [17]. Prompt laparotomy following diagnosis is crucial for achieving better outcomes. Simple reduction should be attempted, though if there is any difficulty in reducing the intussusception or if there is any doubt about the viability of the gut, the intussusception should be resected [18]. Primary anastomosis can be performed successfully, and stomas can be created in the critically ill patients or those with late detection and septicemia [19].

With regards to patients' outcome, there are no clear information about mortality of neonatal intussusception taken apart in the literature. The delay of consultation is a common problem in poor countries deteriorating patients' outcome.

4. Conclusion

Neonatal intussusception is a rare entity appearing mainly in preterm infants. In full-term babies, this abdominal emergency is usually ileoileal and usually due to an organic lesion. To our knowledge, this is the first case of an idiopathic ileocecal intussusception in a full-term newborn. The overall prognosis for neonates with intussusceptions depends on early diagnosis, because once a critical condition develops, as in this case, the mortality rate is likely to rise.

Declaration of Competing Interest

The authors report no declarations of interest.

Source of funding

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Ethical approval

The study is exempt from ethical approval.

Consent

It's a neonatal case report so we asked a parental consent.

Author contribution

1. Study concept and design: Meriem Oumaya.
2. Data collection and interpretation: Meriem Oumaya and Mariem Marzouki.
3. Interpretation and organisation of figures: Asma Souid and Lilia Lahmer.
4. Writing the paper: Meriem Oumaya.
5. Critical revision of the manuscript for important intellectual content: Yosra ben Ahmed and Wiem Douira.
6. Study supervision: Said Jlidi.

All authors read and approved the final manuscript.

Registration of research studies

NA.

Guarantor

Meriem Oumaya.

Provenance and peer review

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References

[1] M.H. Rachelson, J.P. Jernigan, W.F. Jackson, Intussusception in the newborn infant with spontaneous expulsion of the intussusceptum; a case report and review of the literature, J. Pediatr. 47 (juill (1)) (1955) 87–94.

[2] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A.J. Fowler, D.P. Orgill, et al., The SCARE 2018 statement: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 60 (2018) 132–136, 1 déc.
[3] V. Restivo, C. Costantino, F. Tramuto, F. Vitale, Hospitalization rates for intussusception in children aged 0–59 months from 2009 to 2014 in Italy, Hum. Vacc. Immunother. 13 (2) (2017) 445–449, 11 janv.
[4] N.L. Wang, M.L. Yeh, P.Y. Chang, J.C. Sheu, C.C. Chen, H.C. Lee, et al., Prenatal and neonatal intussusception, Pediatr. Surg. Int. 13 (avr (4)) (1998) 232–236.
[5] H.B. Patriquin, E. Afshani, E. Effman, T. Griscom, F. Johnson, S.S. Kramer, et al., Neonatal intussusception. Report of 12 cases, Radiology 125 (nov (2)) (1977) 463–466.
[6] J.R. Avansino, S. Bjerke, M. Hendrickson, M. Stelzner, R. Sawin, Clinical features and treatment outcome of intussusception in premature neonates, J. Pediatr. Surg. 38 (déc (12)) (2003) 1818–1821.
[7] N.L. Wang, M.L. Yeh, P.Y. Chang, J.C. Sheu, C.C. Chen, H.C. Lee, et al., Prenatal and neonatal intussusception, Pediatr. Surg. Int. 13 (avr (4)) (1998) 232–236.
[8] J. Obayashi, H. Koizumi, M. Hoshikawa, M. Wakisaka, A. Fujikawa, H. Kitagawa, et al., A case of congenital infantile fibrosarcoma of the bowel presenting as a neonatal intussusception, Pathol. Int. 67 (déc (12)) (2017) 644–648.
[9] R. Ma, M. Alam, M. Pacilli, E. Hook, A. Nagy, D. Carroll, A rare case of neonatal intussusception caused by cytomegalovirus, Eur. J. Pediatr. Surg. 24 (avr (2)) (2014) 184–186.
[10] R.P. Yoo, R.J. Touloukian, Intussusception in the newborn: a unique clinical entity, J. Pediatr. Surg. 9 (août (4)) (1974) 495–498.
[11] O.O. Ogundoyin, D.I. Ogunlana, O.M. Onasanya, Intestinal stenosis caused by perinatal intussusception in a full-term neonate, Am. J. Perinatol. 24 (janv (1)) (2007) 23–25.
[12] R.A. Bowerman, T.M. Silver, M.H. Jaffe, Real-time ultrasound diagnosis of intussusception in children, Radiology 143 (mai (2)) (1982) 527–529.
[13] A.L. Hryhorczuk, P.J. Strouse, Validation of US as a first-line diagnostic test for assessment of pediatric ileocolic intussusception, Pediatr. Radiol. 39 (oct (10)) (2009) 1075–1079.
[14] E.A. Edwards, N. Pigg, J. Courtier, M.A. Zapala, J.D. MacKenzie, A.S. Phelps, Intussusception: past, present and future, Pediatr. Radiol. 47 (août (9)) (2017) 1101–1108.
[15] L.E. Swischuk, C.K. Hayden, T. Boulden, Intussusception: indications for ultrasonography and an explanation of the doughnut and pseudokidney signs, Pediatr. Radiol. 15 (6) (1985) 388–391.
[16] H.K. Lim, S.H. Bae, K.H. Lee, G.S. Seo, G.S. Yoon, Assessment of reducibility of ileocolic intussusception in children: usefulness of color Doppler sonography, Radiology 191 (juin (3)) (1994) 781–785.
[17] M. Martínez Biarge, A. García-Alix, M. Luisa del Hoyo, A. Alarcón, M. Sáenz de Pipaón, F. Hernández, et al., Intussusception in a preterm neonate; a very rare, major intestinal problem–systematic review of cases, J. Perinat. Med. 32 (2) (2004) 190–194.
[18] V.C. Talwalker, Intussusception in the newborn, Arch. Dis. Child. 37 (avr (192)) (1962) 203.
[19] D.P. Mooney, G. Steinthorsson, N.A. Shorter, Perinatal intussusception in premature infants, J. Pediatr. Surg. 31 (mai (5)) (1996) 695–697.