

Giant Pseudopolyp as a Cause of Life-Threatening Anemia and Colocolic Intussusception in an Infant: A Case Report and Review of the Literature

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Introduction: Ileocolic intussusceptions and colonic pseudopolyps are well known entities in childhood. Intussusception appears within the first year of life, while pseudopolyps appear in preschool children. In contrast, colocolic intussusception is very rare in infants. **Case Presentation:** A 5-month-old male infant was admitted to our hospital, with severe anemia and episodes of colicky abdominal pain. Abdominal ultrasound examination revealed an intussusception. As ultrasound guided hydrostatic reduction achieved partial reduction, emergency laparotomy was performed, showing a narrow-based polyp, as a lead point of a colocolic intussusception. **Conclusions:** In contrast to ileocolic intussusception, colocolic intussusceptions usually occur in adults or in late childhood. A solid colonic pseudopolyp, serving as a lead point for colocolic intussusception in an infant, has not been described so far.

Keywords: Anemia; Colonic Polyps; Diagnosis, Differential; Infant; Intussusception

1. Introduction

Juvenile polyps become symptomatic in early childhood, with a peak incidence between 5 and 9 years and, usually, not before the age of two (1). Occurrence is sporadic or within defined syndromes, like Peutz-Jeghers syndrome (mainly small bowel) or juvenile polyposis syndrome (2, 3).

Intussusception is a common entity in early childhood, with a peak incidence between 5 and 7 months of age (1). However, most cases are ileocolic and idiopathic. In contrast, colocolic intussusception usually has a lead point and tends to present later in life (4).

We report on a 5-month-old boy, with severe anemia due to chronic intestinal blood loss, caused by a large solitary pseudopolyp, located in the transverse colon and serving as a lead point for colocolic intussusception. The combination of an intestinal pseudopolyp with colocolic intussusception in an infant, has, to the best of our knowledge, not been reported so far.

2. Case Presentation

2.1. History and Physical Findings

A 5 months old male infant of non-consanguine parents was admitted to our hospital, with severe anemia and episodes of colicky abdominal pain.

He was pale and, on cardiac auscultation, a loud systolic murmur and mild tachycardia were noted. The ab-

domen was soft, with normal bowel sounds, and there was no tenderness, no palpable mass and no hepatosplenomegaly. Both testicles were fully descended with a testicular hydrocele on the right. On digital rectal examination there was no obvious blood and no other abnormality. Neither skin lesion nor other evidence of external or internal blood loss was detected on further physical examination. Nutritional status was good, with a body weight of 7780 grams (75th percentile). Later in the day, the child had two bowel movements, mixed with dark and partially coagulated blood.

Delivery had been at term and uneventful. The child was exclusively breastfed and had two to four soft bowel movements per day. In the first two months of life, bloody stools had repeatedly been observed. This phenomenon had been attributed to gastroenteritis and resolved spontaneously.

There was no notion of adenomatous polyposis coli in the family.

2.2. Paraclinical Investigations

Red blood count revealed severe hypochromic microcytic anemia: Hemoglobin 2.4 g/dL, mean corpuscular volume 52 fl, mean corpuscular hemoglobin 11 pg, mean corpuscular hemoglobin concentration 22%. Erythrocyte morphology was normal. Reticulocytes were moderately increased, to 2%. Ferritin (1.8 ng/mL) and transferrin satu-

ration (1.9%) were extremely decreased. The C-reactive protein was mildly elevated, to 23 mg/L. White blood cell count, uric acid, creatinine, bilirubin and aminotransferases were within normal range.

Abdominal ultrasound examination revealed a target sign in the left upper abdomen, of approximately 3.5 cm in diameter, suggestive of intussusception (Figure 1).

2.3. Treatment

Primarily, 15 mL of packed red cells per kg body weight were administered. We then proceeded to ultrasound guided hydrostatic reduction. However, only partial reduction, as far as to the right side of the abdomen, could be achieved. During the manipulation, the infant developed increasing abdominal distension, with rebound tenderness and respiratory distress. The child was referred to the pediatric surgery for emergency laparotomy. The abdomen was opened via a transverse incision, in the right upper quadrant. The peritoneal cavity contained a large amount of fluid, which was mainly clear, was compatible with reactive serous peritonitis, except for a small amount of slightly turbid and greenish tinged exudate, in the vicinity of a 7 - 8 cm long intussusception, in the right half of the transverse colon, which appeared long-standing and could not be manually reduced due to firm adhesions between intussusceptum and intussusciptum (Figure 2). Incision of the outer wall revealed a rather narrow-based cauliflower-shaped mass, of about 5 cm in diameter, originating from the wall of the intussusceptum and protruding into its lumen (Figure 3). The affected segment of the transverse colon (7 - 8 cm) was resected and a tension-free end-to-end anastomosis performed.

To keep as much bowel in function as possible, we fashioned a defunctioning loop transverse colostomy, just oral to the inflammatory process, in the left flexure. We saw no reason for an ileostomy, which is associated with the risk of much more fluid loss, exclusion of enterohepatic circulation of bile acids, malabsorption of fat and fat soluble vitamins and all other possible sequelae.

Elective incidental appendectomy was added on, because of an atypically high position of the non-fixed cecum. Further transfusions were administered perioperatively. On histopathology, the mass was identified as a pseudopolyp. Focal hemorrhagic bowel wall infarction was described in the resected colonic segment. Microbiological work-up yielded *Escherichia coli* (*E. coli*) and two strains of enterococci in the abdominal fluid and *E. coli* in a blood culture taken after the surgical intervention. Despite adequate antibiotic treatment, postoperative course was complicated by development of a septic state, due to abscess formation beside the left colic flexure, which required reexploration on day 7. The abscess was drained and a protective loop transversostoma was fashioned in between the left colonic flexure and the intact anastomosis. Subsequent recovery was uneventful. There

was no further gastrointestinal bleeding, the parents soon noticed significantly increased alertness and physical activity, and the stoma was reversed 7 weeks later.

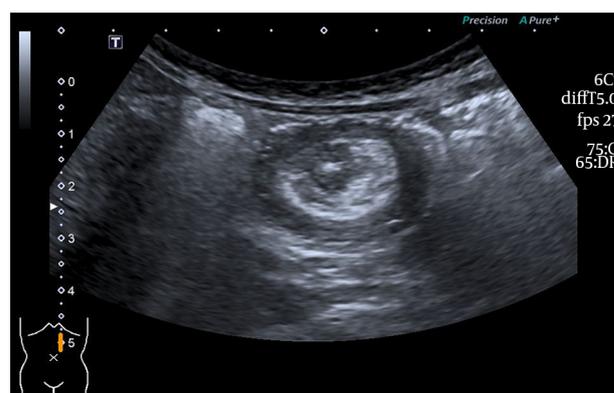


Figure 1. Abdominal Ultrasound on Admission

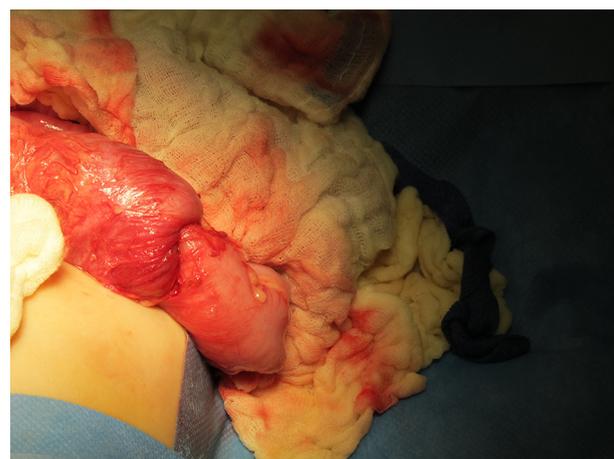


Figure 2. Intraoperative View of the Intussusception in the Transverse Colon

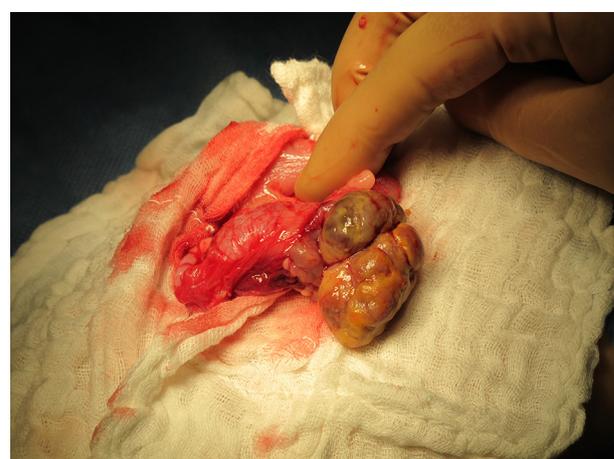


Figure 3. Exteriorized Huge Pseudopolyp

3. Discussion and Review of Literature

Based on a literature review of the past 30 years, the world wide incidence of intussusception in children < 1 year of age has been estimated to average 74 per 100000 (range: 9 - 328), with a peak incidence at 5 - 7 months (1). In a study, analyzing all infant death records in the USA, an annual intussusception mortality rate of 1.4/1000000 live births was calculated, for the year 2007, with a clear downward trend over the past 3 decades (5). In 90% of childhood intussusception cases, the cause is ileocolic, without a leading point (5).

The first rotavirus vaccinations introduced in the late 90s were accused to predispose to intussusception (6). Currently, used rotavirus vaccines seem to be safe and are therefore implemented into the regular vaccination schedule (7).

In contrast to ileocolic intussusception, colocolic intussusceptions usually occur in adults or in later childhood, and they are rarely idiopathic. Benign and malignant tumors, congenital duplications and also inspissated stool, in patients with cystic fibrosis, or a stricture in Crohn's disease, may trigger intussusception in this part of the bowel. A solid colonic pseudopolyp, serving as a lead point for colocolic intussusception, in an infant, has not been described so far (8). The youngest patient, with colocolic intussusception, we found in the literature was a premature girl, born at 28 weeks of gestation, presenting at the age of 38 weeks with an acute abdomen and hypovolemic shock syndrome, with cardiac arrest. An intestinal lymphangioma was found, as the lead point (9). The youngest children with a polyp-induced and idiopathic colocolic intussusception were 3 and 7 years old, respectively (10).

Solid colon polyps are encountered in patients, at a mean age of 5 to 9 years, and more than 80% are located in the sigmoid colon or rectum (11). They may cause significant anemia and only rarely require blood transfusions (12). Ninety percent of colonic polyps in children are benign. In a large observational study on 487 children with colonic polyps, from China, more than 90% of the patients had a juvenile polyp and all polyps in that study were benign (13). The therapy of choice is endoscopic removal, with a recurrence rate < 5% in patients with 1 - 3 polyps. In the patient, we describe the polyp was a juvenile pseudopolyp in the transverse colon. Pseudopolyps are most commonly reported as unusual sequels of inflammatory bowel disease, as a result of mucosal damage and regeneration (13, 14). Obviously, the patient we described did not suffer from inflammatory bowel disease. Therefore, the etiology of the pseudopolyp remains obscure.

Retrospectively, the unusual presentation of the patient, with an intussusception combined with severe anemia, is well explained by the polyp and subsequent rectal blood loss. The ultrasound was neither able to discriminate between colocolic and ileocolic intussusception, nor to

identify the lead point. In a series of 43 children with intussusception, due to a lead point, 2/3 of the lead points could be depicted by ultrasound while 1/3 could not (6).

Postoperative intra-abdominal abscess formation is also extremely rare, in children. In the reported case, it may be explained by bacterial transmigration through the partially necrotic bowel wall into the peritoneal cavity and translocation into the blood stream, as proven by the findings of the same bacteria in ascites, blood culture and abscess, in combination with immunodeficiency, secondary to extreme chronic anemia aggravated by a still immature immune system.

4. Conclusions

Rectal blood loss, in infants, must always be investigated. In the presented case, earlier diagnosis might have permitted endoscopic treatment and would have almost certainly prevented the need of blood transfusions and emergency surgery.

References

- Jiang J, Jiang B, Parashar U, Nguyen T, Bines J, Patel MM. Childhood intussusception: a literature review. *PLoS One*. 2013;**8**(7):e68482.
- Beggs AD, Latchford AR, Vasen HF, Moslein G, Alonso A, Aretz S, et al. Peutz-Jeghers syndrome: a systematic review and recommendations for management. *Gut*. 2010;**59**(7):975-86.
- Howell J, Pringle K, Kirschner B, Burrington JD. Peutz-Jeghers polyps causing colocolic intussusception in infancy. *J Pediatr Surg*. 1981;**16**(1):82-4.
- Arnold M, Sidler D, Moore SW. Compound colonic intussusception: a reason for failure of pneumatic reduction. *J Pediatr Surg*. 2010;**45**(10):e25-8.
- Navarro O, Dugougeat F, Kornecki A, Shuckett B, Alton DJ, Daneman A. The impact of imaging in the management of intussusception owing to pathologic lead points in children. A review of 43 cases. *Pediatr Radiol*. 2000;**30**(9):594-603.
- Murphy TV, Gargiullo PM, Massoudi MS, Nelson DB, Jumaan AO, Okoro CA, et al. Intussusception among infants given an oral rotavirus vaccine. *N Engl J Med*. 2001;**344**(8):564-72.
- Desai R, Curns AT, Patel MM, Parashar UD. Trends in intussusception-associated deaths among US infants from 1979-2007. *J Pediatr*. 2012;**160**(3):456-60.
- Ippolito RJ, Touloukian RJ. Colocolic intussusception in an older child caused by a polyp of the distal colon. *Clin Pediatr*. 1978;**17**(9):720-6.
- Al-Jahdali A, Lees GM, Gay DP, Al-Sairafi R. Colocolic intussusception in a preterm infant with intestinal malrotation. *J Pediatr Surg*. 2009;**44**(12):e17-8.
- Arthur AL, Garvey R, Vaness DG. Colocolic intussusception in a three-year-old child caused by a colonic polyp. *Conn Med*. 1990;**54**(9):492-4.
- Wei C, Dayong W, Liqun J, Xiaoman W, Yu W, Xiaohong Q. Colorectal polyps in children: a retrospective study of clinical features and the value of ultrasonography in their diagnosis. *J Pediatr Surg*. 2012;**47**(10):1853-8.
- Thakkar K, Fishman DS, Gilger MA. Colorectal polyps in childhood. *Curr Opin Pediatr*. 2012;**24**(5):632-7.
- Munchar J, Rahman HA, Zawawi MM. Localized giant pseudopolypoidosis in ulcerative colitis. *Eur J Gastroenterol Hepatol*. 2001;**13**(11):1385-7.
- Kirks DR, Currarino G, Berk RN. Localized giant pseudopolypoidosis of the colon. *Am J Gastroenterol*. 1978;**69**(5):609-14.