Unusual reduction of an unusual form of intussusception: A Case Report

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Citation

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Abstract

We report a case of an 8-month-old infant with intussusception and malrotation with three perforations of the colon. The treatment options in case of a perforation and problems with manual reduction of the intussusception are discussed.

INTRODUCTION

Intussusception is a frequent cause of bowel obstruction in infants and toddlers. The association of intussusception and malrotation has been named Waugh's syndrome by Brereton et al. (1), after George E. Waugh, who first described the association in a report in 1911 (2). Few case reports of this syndrome have been published in the literature the last fifteen years (3141576). The purpose of this report is to document the combination of intussusception and intestinal malrotation with bowel perforation and to highlight the treatment used in this case.

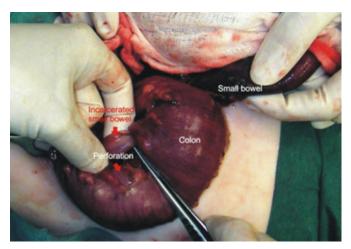
CASE REPORT

A previously healthy 8-month-old girl presented with vomiting and bloody stools since one and a half week. The last days, the vomiting was bilious and the abdomen became distended. She was first seen at a regional hospital where an ultrasound of the abdomen was performed showing signs of intussusception. Subsequently, the child was referred to our pediatric surgical center. On physical examination, we saw an apathic and ill baby, with a heart rate of 160 bpm and very poor peripheral circulation. The abdomen was firmly distended and peristalsis was almost absent and highpitched. Because of her serious clinical condition an exploratory laparotomy was performed immediately. Abdominal exploration showed some unclear abdominal fluid and Ladd's bands with nonrotation. Ileocolic intussusception was seen over a large trajectory; the intussuscepted ileum reached into the distal colon. There were three perforations of the colon; one of ten centimeters in length at the proximal colon and two perforations of three centimeters in length on the mesenteric side at the distal

colon.

Figure 1

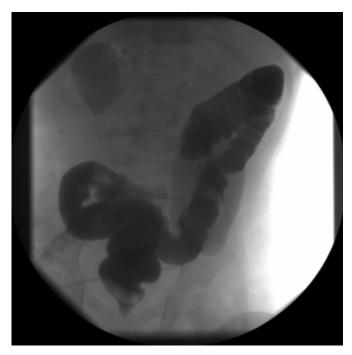
Photo 1: Intraoperative view: ileocolic intussusception over a large traject with perforations of the colon and incarcerated small bowel.



Ladd's procedure was performed. Because the intussusception was too incarcerated, a normal retrograde manual reduction by compression of the colon was not possible. Therefore, the incarcerated small bowel was manually inverted by placing our fingers through the perforations pushing from the luminal side while pulling from the outside. In this way we could redress the intussusception without causing additional perforations. The original perforations were closed and only the ileocecal region was resected because of persistent ischemia. An ileostomy and a mucous fistula of the colon were created. Two days later, a planned second-look laparotomy showed a vital ileum and colon with no signs of necrosis. Postoperatively, our patient stayed on our pediatric intensive care unit. She stayed in the ICU for 3 weeks and was admitted to a pediatric ward afterwards from which she was discharged after another 3 weeks. Three months later, we performed a barium enema study which showed no signs of obstruction (photo 2).

Figure 2

Photo 2: Barium enema study after three months



Subsequently, intestinal continuity was restored without any further complications. Four months after the initial laparotomy, the patient returned to our outpatient clinic. She was in good health and showed no gastrointestinal signs suspect for bowel obstruction, like feeding disorders or constipation.

DISCUSSION

Intussusception of the bowel is a relatively common entity in childhood with an incidence of 1.9 to 4 per 1000 live births in the western world $(_7)$. The combination of intussusception with malrotation has not been widely reported. The etiology of the association of intussusception and malrotation is not completely known. Breckon and Brereton suggest that malrotation by its nature is associated with a mobile right hemicolon, which may be a prerequisite for intussusception. Mustafa Inan shares this opinion $(_{4})$ and we also agree upon this.

Hydrostatic reduction of intussusception by a barium or air enema is the treatment of choice in an intussusception. A surgical approach is chosen in patients with signs of shock,

perforation or peritonitis (8). Our patient showed signs of shock upon arrival in our hospital and therefore a laparotomy was performed. Three large perforations of the colon were found. According to current literature the intussusception should be palpated and reduced intraabdominally by pushing the lead point rather than putting traction on it, since this may result in serosal tears and perforation. If manual reduction cannot be achieved without creating significant serosal tears, resection is required $(_7)$. In our case, the intussusception was so incarcerated that a conventional redressing manoeuvre was not possible. Because the incarceration affected such a large segment of the colon and the ileum, we decided not to primarily resect this entire part of the bowel. A bowel resection of this size might have led to considerable water, electrolyte and shortchain fatty acid resorption disorders (9,10,11). Instead, we used the existing perforations to place our fingers intraluminally. We were then able to redress the intussuseption by pushing the ileum from the inside, while pulling from the outside. After this manoeuvre we did not resect the perforated bowel segment but chose to close the perforations instead.

If classical manoeuvres to redress an intussusception of the bowel are unsuccessful or impossible, we feel that putting traction on the affected segment is worth trying before deciding to perform a bowel resection. Any existing perforations may be useful in this setting and can be closed afterwards. Postoperative bowel stenosis may not be a problem as this case has shown.

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References

1. Brereton RJ, Taylor B, Hall CM: Intussusception and malrotation in infants: Waugh's syndrome. Br J Surg 1986; 73: 55-57

2. Waugh GE: Referred penile pain in intussusception with note of three cases. Lancet I 1911; 1492-1494

3. Luo CC, Wang CR, Chiu CH: Intussusception and intestinal malrotation in an infant: a case report. Pediatr Surg Int 2003; 19: 413-414

4. Mustafa Inan, Umit Nusret Basaran, Suleyman Ayvaz, et al.: Waugh's syndrome: report of two cases. J of Pediatr Surg 2004; 39: 110-111

5. Breckon VM, Hadley GP: Waugh's syndrome: a report of

six patients. Pediatr Surg Int 2000; 16: 370-373 6. Sarin YK, Singh VP: Waugh's syndrome. Indian Pediatr 1995; 32: 108-9

7. O'Neill JA Jr, Grosfeld JL, Fonkalsrud EW et al. In: O'Neill JA Jr (ed.); Principles of Pediatric Surgery, second edition. Mosby, Missouri, 2004; chapter 54: 527-530 8. Sorantin E, Lindbichler F: Management of intussusception. Eur Radiol 2004; 14: L146-L154 9. Treen,WR: Short Bowel Syndrome. In: Wyllie R, Hyams JS (eds.): Pediatric Gastrointestinal Disease, second edition. WB Saunders, Philadelphia, 1999; 315-333 10. Ruppin H, Par-Meir S, Soergel KH, et al: Absorption of short-chain fatty acids by the colon, Gastroenterology 1980; 78: 1500-1507

11. Binder HJ, Mehta P: Short-chain fatty acids stimulate active sodium and chloride absorption in vitro in the rat distal colon. Gastroenterology 1989; 96: 989-996

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