



Transanal protrusion of intussusception can be sign of Waugh syndrome

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ABSTRACT

Intussusception rarely occurs with transanal prolapse of intussusception (TAPI), this presentation may be a sign of Waugh's syndrome (WS), an association between intestinal malrotation and intussusception. The authors present the case of infant with an episode of TAPI, resolved with air enema, who required later diagnostic tests that showed the presence of WS, for which surgery was required after the resolution of the intussusception. At now we found only 72 cases reported of WS and some of them clinically presented with TAPI. In our opinion, patients with this type of presentation require a thorough radiological study of the intestine to rule out intestinal malrotations.

1. Introduction

Intussusception is an acute condition in which part of the intestine enters the lumen of the adjacent segment [1]. It is the most common cause of intestinal obstruction in childhood. It takes on different forms depending on the tract affected: the most common is the ileo-colic one [2]. It typically presents with intestinal obstruction in the course of enteritis, vomiting, lethargy and blood in the stool. Rarely, in 8–29% of cases [3–6], the tract affected by intussusception can prolapse through the anus: transanal protrusion of intussusception (TAPI). This presentation, more frequent in females (female/male ratio: 3/2) and in some countries [6–8], may be due to an abnormal position of the intestine and defined as Waugh's syndrome (WS) [9,10]. Classically the treatment of the intussusception is based on the enema reduction with fluoroscopy, reserving the surgery for the cases of failed reduction. However, in the cases of TAPI, even after enema reduction of the intussusception, surgery may be required to treat intestinal malrotation that is frequently associated, as WS, in TAPI [7]. The Authors report a review of the Literature and their experience on a clinical case presented as TAPI in which surgery was required, after air enema reduction, to treat the intestinal malrotation.

2. Case presentation

A 3-month-old girl, born at 26 weeks by caesarean section, first meconium emission in 24 h, breastfeeding. She was admitted to neonatal intensive care for prematurity. In the previous month he had 3 episodes, recorded as rectal prolapse with "auto" reductions. Subsequently she had a trans-anal protrusion of intussusception (Fig. 1) so that an easy reduction with air enema was performed twice because of recurrence 24 hours later. Because of the recurrent TAPI, once intussusception was resolved, diagnostic investiga-

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Fig. 1. Image of transanal protrusion of intussusception.

tions were performed to rule out some intestinal anomalies causes of prolapse. Suction rectal biopsies for Hirschsprung's disease were negative, as well as Magnetic Resonance Imaging of the cauda equina, and test for cystic fibrosis. One week later a study of the colonic anatomy by contrast enema (Iopamidol diluted with sterile solution, ratio 1: 4) showed an intestinal malrotation with the colon fully positioned to the left) (Fig. 2). After resolution of the intussusception she started breastfeeding and for about 2 weeks had irregular evacuations and transient abdominal distension. Because of this difficulty in feeding and intestinal malrotation, the girl underwent laparotomy that showed an extreme mobility of the cecum (placed anteriorly in the left hypochondrium and colon; therefore a repositioning of the intestine in the abdomen with the cecum in the left iliac fossa was performed. The patient was fed up, had no problems and was fine at the 6-month follow-up.

3. Discussion

Intussusception is one of the most frequent causes of intestinal obstruction, which must be diagnosed and treated early to avoid serious consequences. The clinical presentation may vary according to the age of the patient and the underlying pathology (diverticu-

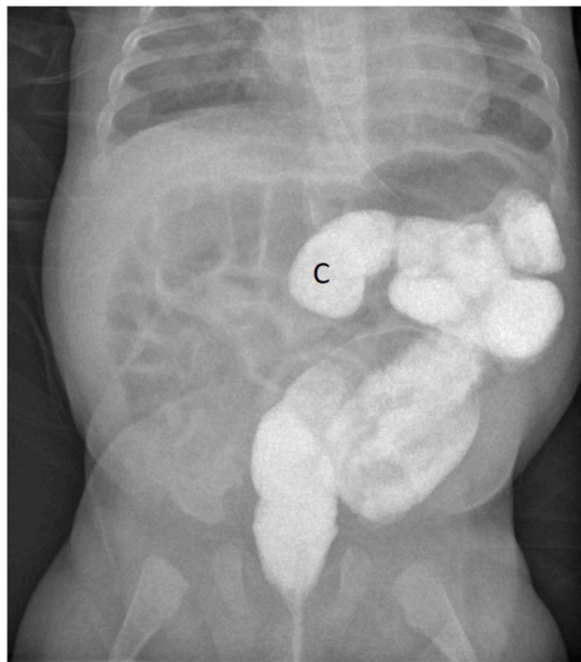


Fig. 2. Image of contrast enema that shows the intestinal malrotation. C: cecum.

lum, intestinal angioma, etc.). An infrequent form of clinical presentation is TAPI, a clinical condition with the presence of a mass (made up of the invaginated intestine) that prolapses through the patient's anus and creates diagnostic difficulties with rectal prolapse [11,12]. Distinguishing the two conditions is very important for treatment. Some signs are characteristic for the diagnosis of TAPI such as the “dance sign”, ie the lower right quadrant empty on palpation, and the possibility of exploring with the finger the space between the prolapsing mass and the anal wall [7,13]. Also in this case-report there was a probable misdiagnosis of rectal prolapse which resolved spontaneously. This patient shows an association between intestinal malrotation and intussusception that occurs with TAPI, known as Waugh's syndrome, described for the first time in 1911 [10]. Recurrent rectal prolapse in infant can be caused by various pathologies such as Hirschsprung's disease, cystic fibrosis, nervous anomalies of the sacrum and intestinal malrotation. All these disorders were investigated in our case due to the reported episodes of rectal prolapse. Waugh's syndrome is a rare association between bowel malrotation and intussusception and has been reported a long time ago. After resolution of the intussusception, the treatment consist in the repositioning the bowel in the abdomen and it is possible to perform also laparoscopically, in this case we preferred a laparotomic approach because of the recurrent intussusception and the infrequent association between TAPI and WS. Analyzing the Literature to date we have found 71 cases reported in pediatric age and one in an adult [11,14,15]. Not in all patients with WS, intussusception occurs with TAPI, but this sign may indicate the presence of association [16,17]. For this reason we believe it is important to make a correct differential diagnosis between TAPI and rectal prolapse, also after the resolution of the intussusception with enema. It is important in these patients to perform a radiological study to evaluate the possibility of intestinal malrotation which must be treated surgically, to avoid further intussusception that may require intestinal resections as reported by other authors [7].

Consent to publish the case report was obtained

This report does not contain any personal information that could lead to the identification of the patient.

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All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

References

- [1] Huppertz H.I, Soriano-Gabarro M, Grimprel E, et al. Intussusception among young children in Europe. *Pediatr Infect Dis J* 2006;25(suppl 1):S22–9.
- [2] Ekenze S, Mgbor S Childhood intussusception: the implications of delayed presentation. *Afr J Paediatr Surg* 2011;8(1):15.
- [3] Mburu E, Akello W, Kimani W, et al. Trans-anal protrusion of intussusception. *J Pediatr Surg Case Rep* April 2020;55:101405.
- [4] Ramachandran P, Vincent P, Prabhu S, Sridharan S Rectal prolapse of intussusception - a single institution's experience. *Eur J Pediatr Surg* 2006;16(6):420–2.
- [5] Obiora E.U, Sebastian E.O, Ogundu II. Transanal protrusion of intussusceptions in children. *Afr J Paediatr Surg* 2014;11(3):229–322.
- [6] Ameh E.A, Mshelbwala P.M Transanal protrusion of intussusception in infants is associated with high morbidity and mortality. *Ann Trop Paediatr* 2008;28(4): 287–92.
- [7] Ibrahim I.A Prolapsed ileocolic intussusception. *Ann Pediatr Surg* 2011;7(2):76–8.
- [8] Ndour O, Ndoye M Prolapsed intussusception in children: clinical study and therapeutic aspects. 2013. December 2015.
- [9] Brereton R.J, Taylor B, Hall C.M Intussusception and Malrotation in infants: Waugh's syndrome. *Br J Surg* 1986;73:55–7.
- [10] Waugh G.E Referred penile pain in intussusception with notes of three cases. *Lancet I* 1911;1492–4.
- [11] Al-Momani Hashem. Waugh syndrome: a report of 7 patients and review of the published reports. *Ann Saudi Med* 2014;34(6). Nov-Dec.
- [12] Shahramian I, Parooie F, Mirabbasi S.A, et al. Colonoscopic reduction of a transanal prolapsed ileocolic intussusception. *Wien Med Wochenschr* 2022. <https://doi.org/10.1007/s10354-022-00915-4>. Feb 15, In press.
- [13] Tennant S, Halliday K Intussusception mimicking rectal prolapse. *Pediatr Radiol* 2008;38:700–2.
- [14] Saxena Rahul, Puri Archana, Pinnamaneni Ramya. Waugh syndrome in preterm infant: diagnostic clues. *Pediatr Neonatol* 2015 Jun;56(3):203–4.
- [15] Gandhi Vidhyachandra, Pai Nitin, Kashiva Reema. Adult with intestinal malrotation and colocolic intussusception: an unusual combo. *BMJ Case Rep* 2019;12(7):e226398. Jul 22.
- [16] Ezomike U.O, Ekenze S.O, Igwilo I.O Transanal protrusion of intussusceptions in children. *Afr J Paediatr Surg* 2014;11(3):229–32. Jul-Sep.
- [17] Joob B, Wiwanitkit V Transanal protrusion of intussusceptions. *Afr J Paediatr Surg* 2014;11(4):372–3. Oct-Dec.