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Case Report

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Retrograde intussusception and Giant Meckel's diverticulum: a rare occurrence in Waugh's syndrome

Abstract

Retrograde intussusception (RINT) can occur anywhere in the gastro-intestinal tract. We report a case of ileo-ileal RINT with giant Meckel's diverticulum having Waugh's syndrome. Usually giant Meckel's diverticulum presents as volvulus in neonates and adhesions/ mass effect in adult. Here we discussed a case of ileo-ileal RINT with giant Meckels' diverticulum.

Keywords: Intestinal obstruction, Meckel's diverticulum, retrograde ileo-ileal intussusception, reverses intussusception, Waugh's syndrome

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Introduction

Intussusception is defined as telescoping of proximal bowel segment into immediately adjacent distal bowel segment. Thorek and Lorrimer broadly classified intussusception into ante grade and retrograde variants (Table 1).[1] Retrograde/ anti peristaltic/ reverse intussusception (RINT) is defined as "anti peristaltic telescoping of distal bowel into proximal bowel.[1,2] Usually RINT occurs in patients with Roux-en-Y gastric bypass, long intestinal tubes, metastatic bowel malignancy, and Achalasia Cardia etc.[1–7] Here, we describe a case of ileo-ileal RINT predisposed by a giant Meckel's diverticulum (MD) in a patient with Waugh's syndrome..

Table 1 Classification of Intussusception by Thorek and Lorrimer et al.

Ante grade		Retrograde	
1	Gastro duodenal	1	Gastro-oesophageal
2	Enteric	2	Duodeno-gastric
3	Enterocolic	3	Enteric
4	Colic	4	Caeco-ileal
		5	Colic
		6	Jejuno-gastric

Case report

A 1.5-month-male infant, weighing 5-Kg presented with features of acute intestinal obstruction for 2 days. There was no history of bleeding per rectum. On examination, the abdomen was grossly distended. No intra-abdominal lump was felt. Bowel sounds were diminished. Per-rectal examination didn't reveal any bleeding, mass or mucus discharge. Haematological investigations showed leucocytosis and hypokelamia. Intravenous fluid and parenteral antibiotics were administered and the dyselectrolytemia was corrected. Abdominal X-ray (erect) and ultra sonography documented a huge gas shadow occupying most of the abdominal cavity and dilated bowel loops. The infant underwent exploratory laparotomy under general anaesthesia. A giant MD (14 cm long axis, 5cm transverse axis, 3cm vertical axis, and 2cm at base) was found arising from anti-mesenteric border of terminal ileum (30cm proximal to ileocaecal junction). The MD had a very short mesentery (4 cm along long axis and 1cm perpendicular to it) containing feeding vessels. The diverticulum and adjacent loop of ileum were fixed to retro peritoneum. The caecum and colon were not retro peritoneal in location. The duodeno-jejunal flexure was on the right of midsaggital plane; The terminal ileum (5cm proximal to ileo-cecal junction) had telescoped into proximal ileum reaching up to the base of MD (Figure 1) (Figure 2). Proximal ileum and MD were grossly dilated. Manual reduction of RINT was successful, but involved bowel had some necrotic patches. The giant MD was excised along with the adjacent segment of non-viable ileum. End to end ileo-ileal anstomosis was carried out along with appendectomy. The abdomen was closed in layers. Laying open of excised specimen didn't reveal any pathological lead point. The MD was seen arising from ante mesenteric border of ileum and freely communicating with the lumen of ileum. Histopathological examination showed inflammatory infiltrate in MD and ileum. The postoperative period and follow up were uneventful.

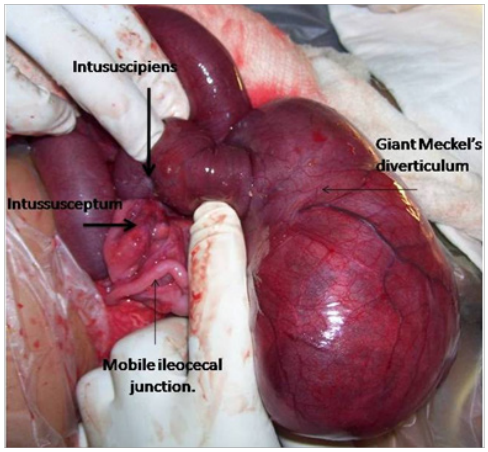

Figure 1 Intra- operative photograph showing retrograde/reverse ileo-ileal intussusception (bold vertical arrows), giant Meckel's Diverticulum and mobile caecum.

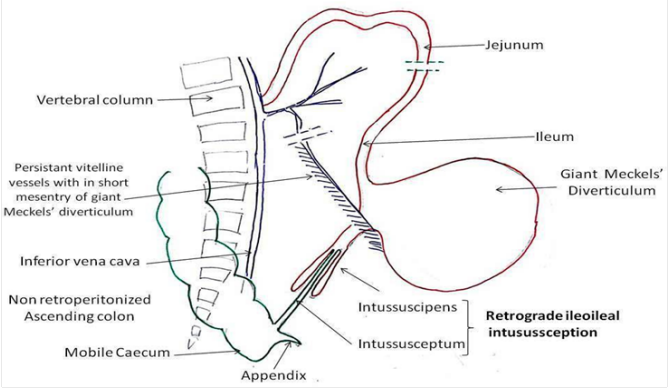

Figure 2 schematic diagrams showing giant Meckel's diverticulum, having short mesentery containing persistent vitelline vessels, retrograde ileo-ileal intussusception.

Discussion

Meckle Diverticulum is the remnant of proximal portion of vitelline/ omphalomesenteric duct. Usually MD is short and wide mouthed (average 2.9cm long and 1.9cm wide); however, in our case, the MD was huge in proportion with dimensions of 14cm×5cm×3cm. Further, the feeding vessels of MD (vitelline vessels) which usually obliterates during phase of fetal development could be identified within the narrow mesentery.[9] It has been well described in literature, that appearance of MD is a clue to the type of complication it might undergo.[8] Diverticulitis and torsion are common complications associated with a long and narrow based MD, while broad, stumpy MDs are susceptible to intestinal obstruction.[9,10] Intestinal obstruction may occur via herniation of bowel through a ring formed by diverticulum attachment to abdominal wall. The other noteworthy complications in association with MD are volvulus, axial torsion; diverticular adhesions, incarcerated litter's hernia, foreign body/phytobezoar/ enterolith impaction, and benign/ malignant tumors.[8] The MD may inverts within the lumen of bowel, simulating an intestinal polyp, which acts as a pathological lead point for subsequent development of intussusception.[10] The giant MD more often causes intestinal obstruction in all age groups. In neonates, giant MD may cause intestinal obstruction by volvulus; while in adults, it usually leads to adhesive obstruction and mass effect.[10] In our case the proposed aetiology might be, anti-peristalsis initiated by mild gastrointestinal infection in the ileum and coexistent anatomical factors such as mobile ileo caecal junction and MD predisposed the intussusception to proceed in retrograde direction.[6] The precise incidence of intestinal mal rotation is unknown., It is estimated that it occurs between 0.0001% and 0.19 % of adults.[10,11] Intestinal mal rotation encompasses various anatomic anomalies ranging from complete non rotation to normal position and broad-based mesentery i.e. incomplete rotation, mixed rotation, atypical mal rotation, and other variants of mal rotation. Association of idiopathic intussusception with mal rotation is not widely reported, yet few authors quote this association in as many as 40 % of cases.[7] Brereton et al. termed this association "Waugh's syndrome" on the name of "George A. Waugh", who first identified the association.[7] The importance lies in the fact, that whenever pneumostatic/hydrostatic enema reduction of intussusception is successful, the mal rotation component should be ruled out by radiology as these patients are also at risk of volvulus or recurrent intususception.[8] Our patient had defect in second stage of midgut rotation (duodeno-jejunal flexor to the right of superior mesenteric vessels and mobile ascending colon with caecum).

Usually intussusception in Waugh's association proceed in ante grade direction, but here intussusception occurred in reverse direction.[10–12] Unfortunately, we were unable to detect presence of RINT on ultra sonography as most of the abdomen was obscured by the huge gas shadow of Giant MD. We didn't suspect a preoperative diagnosis of RINT, hence gastro-intestinal contrast study was not advised for this patient. Most common type of RINT is jejuno-jejunal following Roux-en-Y gastro-jejunostomy.[1] Spontaneous RINT can be encountered after cardiomyotomy as the hyper mobile distal bowel segment (stomach) telescope within the dilated proximal esophagus.[2] Spontaneous colo-colic RINT is an usual occurrence in older age. Loss of anti-peristaltic activity of colon coupled with exaggerated ante grade peristaltic waves in proximal bowel due to ante grade intussusception leads to sliding of proximal bowel over the ante grade intussusception;[6] however, in our case the mechanism was entirely different. The index case is unique in many ways. It is the first reported case of Waugh's syndrome having RINT. This is also the first case of primary ileo-ileal variety RINT in infants.[5] Giant MD having persistent remnants of primitive vitelline vessels which lead to fixation of loop of ileum to the retroperitoneal is never described in the literature. The case also proposes a newer mechanism of intestinal obstruction by MD. To conclude, giant MD is almost always associated with complications, one of which can be RINT. Thus, we recommend surgical excision of giant MD in all patients.[8,10]

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None.

Conflict of interest

The authors declared there is no conflict of interest.

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