Intestinal atresia due to intrauterine intussusception of a Meckel's diverticulum

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A R T I C L E   I N F O

Article history:
Received 23 May 2013
Received in revised form 6 July 2013
Accepted 9 July 2013

Key words:
Meckel's diverticulum
Ileal atresia
Intrauterine intussusception

A B S T R A C T

It is widely acknowledged that jejunoileal atresias result from late intrauterine mesenteric vascular accidents as shown in Louw and Barnard's classic study. These accidents include intestinal volvulus, malrotation, intussusception, internal hernia, and strangulation in association with gastrochisis and omphalocoele. The incidence of intussusception as the cause of intestinal atresia is between 0.6 and 13.1%. Few case reports have demonstrated Meckel's diverticulum as the lead point of the intussusception leading to a jejunoileal atresia. We present such a case, with an intestinal atresia resulting from intra-uterine intussusception of a Meckel's diverticulum. The neonate was treated with resection of the atresia with primary bowel anastomosis and a Meckel's diverticulectomy with a full recovery. © 2013 The Authors. Published by Elsevier Inc. Open access under CC BY license.

Meckel's diverticulum is the most common congenital abnormality of the GI tract, occurring in 1–2% of the population. The lifetime risk of becoming symptomatic is 4%, with over 90% of symptomatic patients presenting with painless rectal bleeding, obstruction, or inflammation in a child. Rarely, a symptomatic Meckel's diverticulum may present in the neonatal age group, with case reports in the literature of obstruction, inflammation, perforation, and intestinal atresia [1]. We present a case of an intestinal atresia due to an intussuscepted Meckel's diverticulum, the fifth described case in the English literature and review of the literature [2–5].

1. Case report

A 3146-g white male was born after an uneventful pregnancy at 39 weeks gestation by spontaneous vaginal delivery. At birth, the Apgar scores were unremarkable. Initially, he tolerated feeds and passed meconium. However, on day of life 2, he developed abdominal distention with bilious emesis. On exam, the abdomen was non-tender but distended with visible loops of dilated bowel (Fig. 1). Abdominal X-rays showed a large stomach bubble with dilated loops of small bowel suggestive of a high-grade obstruction (Fig. 2). Given the presentation, a jejunoleal atresia was suspected, and the patient was taken to the OR. Laparotomy revealed an atresia of the terminal ileum with proximal and distal loops of bowel ending blindly. The proximal bowel was markedly distended. There was a dilated proximal end of ileum and a decompressed distal end with a small mesenteric defect (Type IIIa atresia) (Fig. 3). In opening the distal segment to perform an anastomosis, a blind intussuscepted segment of intestine was encountered within the lumen. The location, size, and appearance of the intussuscepted segment were consistent with a Meckel's diverticulum (Fig. 4). About 15 cm of the dilated proximal bowel and 2 cm of the distal bowel were resected and an end-to-end anastomosis was made. The child had an uneventful postoperative period and was discharged on post-operative day 14.

2. Discussion

The theory of the origin of intestinal atresia has evolved from Tandler's theory of non-recanalization of the gut to the current most accepted theory of vascular disruption of mesenteric blood flow leading to bowel necrosis and resorption [6]. Louw and Barnard confirmed this hypothesis in 1955 after producing intestinal atresia in puppies following in utero ligation of mesenteric vessels. The intestinal atresia seen in these animals was a Type IIIa atresia...
(Fig. 5), with a grossly distended proximal bowel loop followed by a short V-shaped atresia and small distal bowel.

Dalla Vecchia et al. study reported the characteristics of 128 jejunoileal atresias from a single institution [7]. Disruption of mesenteric blood flow in utero leading to the atresia was associated with volvulus in 34 neonates (27%), malrotation in 24 (19%), gastroschisis in 21 (16%), omphalocele in 2 (1.6%), Meckel’s diverticulum (without intussusception) in 3 (2.4%), and intussusception in 2 neonates (1.6%) [7]. A comprehensive meta-analysis of intestinal
intestinal atresia. The first three cases were described in Turkey, with the first two cases from the same institution [3–5]. The fourth case was described in Taiwan [2]. This rare presentation of a common congenital abnormality has an excellent prognosis with early identification and surgical correction. While this is only the fifth case of intestinal atresia due to a Meckel’s diverticulum described, ileal atresias resulting from idiopathic intrauterine intussusception or other vascular accident in the neonatal period are more common. We suspect that some or many of these previously described ileal atresias may be the result of an intrauterine intussusception of a Meckel’s diverticulum rather than an isolated idiopathic intussusception or other vascular accident.

3. Conclusion

The theory of jejunoileal atresia as a result of disruption of mesenteric blood flow is now widely accepted doctrine. Although the vascular accident is rarely associated with either a Meckel’s diverticulum or intussusception, a Meckel’s diverticulum acting as the intussusceptum is even more rare. We reported such a case in a 2-day old neonate.

Sources of funding

None.

Conflict of interest statement

None.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Acknowledgment

None.

References


atresia studies by Todani et al. shows the incidence of intussusception as the cause of intestinal atresia is 0.6–13.1% [8]. To date, less than 40 cases of intrauterine intussusception leading to intestinal atresia have been adequately documented in the world’s literature [9]. Within these case reports, a recognizable cause, such as a Meckel’s diverticulum, of the intussusception is very infrequent.

Meckel’s diverticulum results from persistence of the omphalomesenteric duct. Normally obliterated in the 5th–7th week of gestation, failure to do so results in an omphalomesenteric duct remnant, which includes a Meckel’s diverticulum. A Meckel’s diverticulum can act as a lead point for an intussusception, resulting in a vascular accident and necrotic bowel. Resorption of this necrotic bowel causes a type Ila intestinal atresia [6].

Similarly to previous case reports of intussusception induced intestinal atresia, meconium was passed shortly after birth in our patient. Since meconium does not reach the colon until the fourth month of gestation, it is thought that Meckel’s diverticulum causes a late intrauterine intussusception and bowel obstruction [10].

The neonate in this case was treated with resection of the atresia with primary bowel anastomosis and a Meckel’s diverticulectomy. Dalla Vecchia et al. report similar treatment in 45 of their patients (35%) [7]. Additional treatments included resection with tapering enteroplasty in 23 neonates (18%), temporary ostomy in 54 (42%), enterotomy with web excision in 5 (4%), and a Bianchi longitudinal intestinal lengthening procedure in 1 (0.8%). Postoperative course was uneventful and the neonate made a full recovery in our case.

This is the fifth case report in the English literature of intrauterine intussuscepted Meckel’s diverticulum leading to an