Case Report

Unusual Ileal Anomalies Associated with Omphalomesenteric Duct Remnants

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Abstract

The association between omphalomesenteric duct remnants and other digestive tract malformations is not rare. Most associated anomalies are reported with Meckel's diverticulum. We report two associated anomalies which were never reported: an ileal stenosis and an ileal duplication. Surgeons must be aware of associated anomalies to prevent post-operative complications.

Keywords: Duplication, ileum, omphalomesenteric remnants, stenosis

Introduction

Omphalomesenteric duct (OMD) is the normal embryological communication between the yolk sac and the primitive midgut.[1] The closure of the OMD is followed by its resorption at 8th or 9th week of gestation.[1] Absence of closure and incomplete resorption occur in 2% of the population. It may remain silent or lead to various anomalies called OMD remnants: umbilicoileal fistula (patent OMD), umbilical sinus, Meckel's diverticulum, umbilical cyst, umbilical mucosal polyp or a fibrous cord connecting the ileum to the umbilicus.[1] Association between omphalomesenteric remnants and digestive tract malformations is not rare. We report two cases of associations which were never reported: omphaloileal fistula (OM fistula) and umbilical sinus associated with ileal stenosis and ileal duplication, respectively. Analysis of these cases emphasises that the OMD remnants can have an association with the ileal anomalies. Potential technical hazards make it important that this association is recognised.

Case Reports

Case 1

A 5-day-old male was admitted for faecal umbilical discharge. After the first normal meconial evacuation, the mother noticed the absence of the normal faecal evacuation through the anus. The patient was born through normal vaginal delivery with an uncomplicated prenatal history. On examination, a mucosa with a lumen was noticed in umbilicus with a feculent discharge [Figure 1a]. The diagnosis of omphalomesenteric fistula was made. Through a periumbilical approach, we found a particular OMD with a stenotic distal part of the ileum. We have noticed a fibrosis within the umbilical ring entangling and strangulating the ileum [Figure 1b]. We performed a resection of the duct, tapering of the ileum followed by end-to-end anastomosis and excision of the mucosa from the umbilicus without omphaloplasty. The patient had an uneventful recovery and was discharged on the 7th post-operative day. Pathologic analysis found a patent OMD with ileal stenosis caused by a fibrous and inflammatory thickening of the strangulated ileal wall.

Case 2

A 1-year-old male was admitted for an 11-month history of umbilical discharge. On examination, we noticed an umbilical mucosa with a very small lumen. Abdominal ultrasound showed a retro-umbilical mass (13.6 mm × 9.5 mm). Fistulography had not been performed because of difficult

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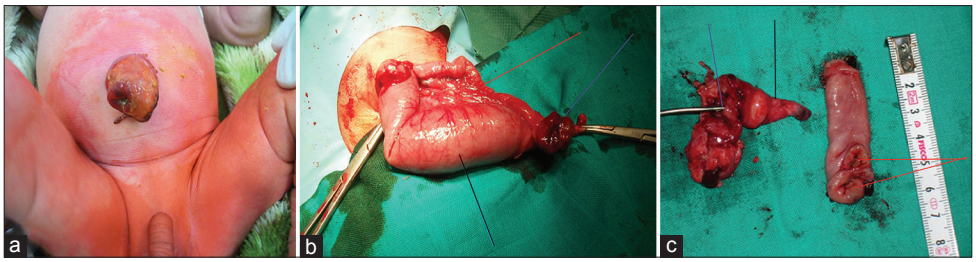


Figure 1: (a) Umbilical fistula with protrusion of intestinal mucosa and persistent umbilical discharge. (b) Intra-operative photograph before excision of omphalomesenteric fistula. Blue arrow: Omphalomesenteric fistula with intestinal mucosa. Red arrow: Distal stenotic ileum. Black arrow: Proximal ileum. (c) Gross specimen showing: Blue arrow: Umbilical mucosa with lumen. Red arrow: Duplicated ilium. Black arrow: Sinus

catheterisation. Through a periumbilical approach, we noticed an umbilical sinus connected to the duplicated ileum by a fibrous cord. The length of the tubular duplication was 6 cm. Excision of the umbilical sinus, the fibrous cord and the 6 cm duplicated ileum was performed [Figure 1c]. The post-operative course was uneventful, and the patient was discharged on the 6th post-operative day. Pathologic analysis found an umbilical sinus with a small intestine mucosa and heterotopic gastric epithelium.

Discussion

OMD remnants occur in 2% of the population. The association with digestive tract malformations was already reported, especially in the case of Meckel's diverticulum.[2] The associated digestive tract malformations reported are oesophageal atresia, anorectal atresia, duodenal atresia, ileal atresia and exomphalos.[2] To the best of our knowledge, only a single case of association between OM fistula and ileal atresia has been reported in the literature;[3] ileal stenosis and atresia are the consequence of a compromise of the blood supply to the small bowel that can be caused by a volvulus, intussusception or snaring at the umbilical ring.[4] Data from animal studies have shown that a partial obstruction of the superior mesenteric artery caused a stenosis of the small bowel.[3] In the first case, the stenosis of distal ileum can be explained by the snaring at the umbilical ring. The association between umbilical sinus and ileal duplication had never been reported. It can be explained by the persistence of endomesenchymal tract between the yolk sac and amnion.[5] The treatment of umbilical sinus alone could have let the ileal duplication with severe complications.

Conclusion

We have reported two ileal anomalies associated to OMD remnants which were never reported before. Surgeons must be aware of associated anomalies to prevent technical hazards.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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