ASPS0010

A RARE CASE OF INTRAUTERINE INTUSSUSCEPTION CAUSING ILEAL ATRESIA

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Introduction

Intussusception is a well-known cause of acute intestinal obstruction in infants and young children. Intrauterine intussusception on the other hand, is a rare occurrence. A review of 1500 cases of intestinal atresia by Evans, noted that intrauterine intussusception was the cause in only 0.6% of the cases[1].

Case Report

A district hospital in Sabah, Malaysia referred a neonate for delayed passage of meconium and vomiting. Mother had an uneventful antenatal history. He was born term at 38 weeks gestation with a birth weight of 2.8kg and allowed breastfeeding on demand immediately after birth. However, he did not pass meconium in the first 24 hours of life, and only had bowel opening at 40 hours of life, which was of normal dark greenish colour. He also developed bilious vomiting within the first 24 hours. On examination, he was non-syndromic, there was no heart murmur, his abdomen was distended, without any palpable mass or abdominal discoloration, his anus was patent, and per rectum there was no gush of air/explosive stools. Abdominal x-ray showed dilated bowels centrally with sudden paucity of bowel gas. We proceeded with a lower GI contrast study (Figure 1).

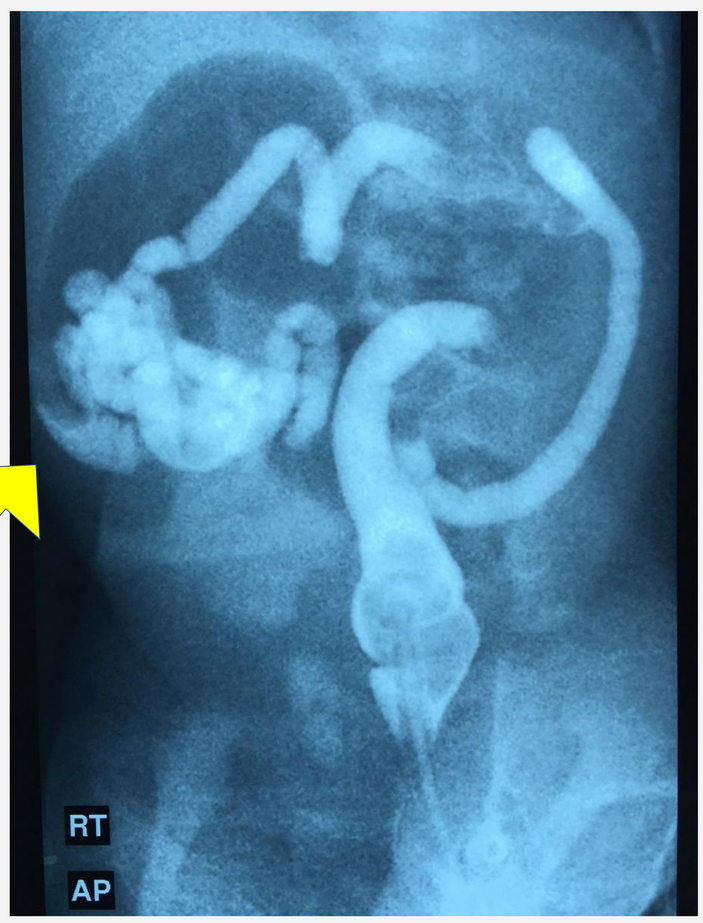

Fig 1. Contrast enema showing patent but small calibre colon, with contrast refluxing beyond the ileocaecal valve and abruptly stops with a claw sign (arrow). Small bowel appears dilated proximally.

With the working diagnosis of small bowel atresia, we proceeded with laparotomy via a right upper transverse incision.

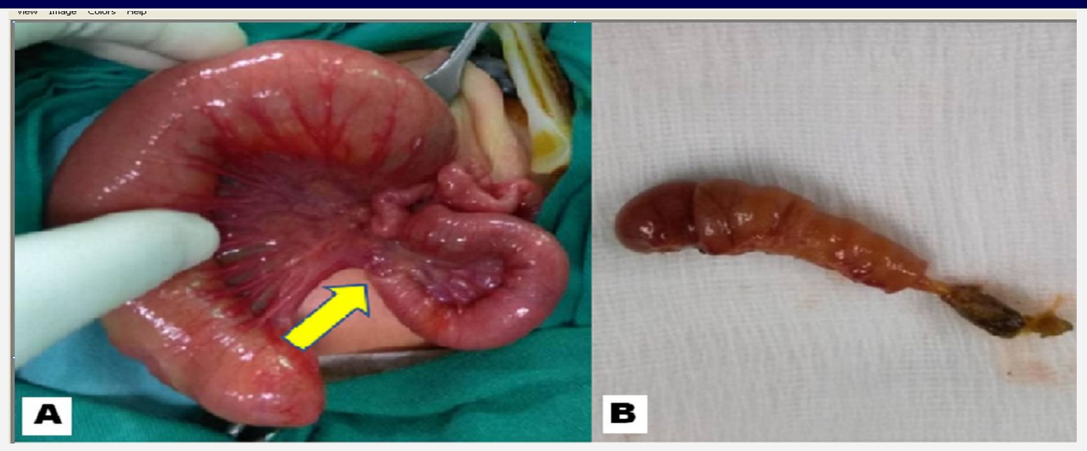

Fig 2. Intraoperative finding. (A) Ileal atresia with mesenteric defect 123cm from DJ, dilated proximal pouch. Bowel intussusception (arrow) noted at the distal atretic segment (28cm from ICV), with the apex around 5cm from the atretic end. (B) Resected distal atretic segment showing the gangrenous intussusceptum bowel protruding out of the intussuscipien bowel.

The atretic ends were resected followed by primary end to end anastomosis. On post op day five tube feeding was initiated and breastfeeding on demand the next day. He was discharged home well on post op day 10.

Discussion

The lower GI contrast study, which showed a claw sign was a good clue that this neonate had intrauterine intussusception. Despite the passage of normal meconium after 40 hours of life, intestinal atresia could not be ruled out. In Todani et al's review, 45% of the cases of intrauterine intussusception causing intestinal atresia passed normal meconium as well[2], consistent with the assumption that the intrauterine intussusception may have occurred in late fetal life. It impedes the blood flow to the affected bowel segment, and results in gangrene and resorption. It was shown by Tsujimoto et al that intestinal atresia could develop in 4-5 days after an intussusception in rabbit fetal models[3]. In some cases, it was complicated with meconium peritonitis. Intrauterine intussusception causing small bowel atresia remains an uncommonly reported entity. However, the cause as to why intrauterine intussusception occurs, remains a mystery. Can the same causes of post natal intussusception be said for intrauterine intussusception? There have been isolated reports of meckel's diverticulum playing a role as the lead point to intrauterine intussusception, but in this case, it was not demonstrable.

Conclusion

Intrauterine intussusception is a rare but evident cause of intestinal atresia. Claw sign seen on a neonatal contrast study should raise the suspicion of intrauterine intussusception. It has a good prognosis if treated surgically. More studies are needed to better understand this pathology.

REFERENCE:

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2. Todani T, Tabuchi K, Tanaka S. Intestinal Atresia due to Intrauterine Intussusception: Analysis of 24 Cases in Japan. J Paediatric Surg. 1975;10(No.4):445–50.
3. Tsujimoto K, Sherman F, Ravitch M. Experimental intestinal atresia in the rabbit fetus. Sequential pathological studies. Johns Hopkins Med J. 1972;131(4):287–97.