

INTRAUTERINE INTUSSUSCEPTION: A RARE CAUSE OF ILEAL ATRESIA WHICH CAN LEAD TO CONFUSION IN DIAGNOSIS*

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A case of ileal atresia resulting from intrauterine intussusception is presented to stress the confusing differences between this condition and typical ileal atresias. Key words: *ileal atresia, intrauterine intussusception, neonatal intestinal obstruction.*

Ileal atresia is a significant cause of intestinal obstruction encountered in the neonatal period. Its incidence varies from one in 330 to one in 1500 live births^{1,2}. Intrauterine compromise of the vascular supply to the ileum is suggested to be the most common cause of ileal atresia. Volvulus, congenital bands, internal herniation and intussusception lead to vascular compromise and, thus, to atresia. The typical case of ileal atresia presents with bilious vomiting, inability to pass meconium, and air-fluid levels on plain erect abdominal radiographs. Diagnosis is easily established with the appearance of a microcolon on barium enema examination¹.

Intrauterine intussusception is an infrequently reported cause of ileal atresia. Fifty such cases have been reported in the literature. Ileal atresia, resulting from intrauterine intussusception, presents with different clinical and radiological signs³⁻¹³. A case of ileal atresia, resulting from Intrauterine intussusception, is presented to stress the confusing differences between this condition and typical ileal atresias.

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Case Report

A full-term female weighing 3500 g was referred to the Hacettepe University Children's Hospital at 24 hours of age with a history of bilious vomiting and abdominal distension. Past history of the passage of meconium was not obtained. Physical examination was normal except for abdominal distension. A plain erect abdominal radiograph revealed multiple, wide-based air-fluid levels, suggesting low intestinal obstruction (Fig. 1). A barium enema examination revealed a colon of normal caliber (Fig. 2). Later, the patient passed a dark green, somewhat dry stool measuring from 5-10 Cm³. At laparotomy, a fibrous cord type of ileal atresia (Grosfeld Classification-Type II) was detected about 20 cm proximal to the ileocecal valve (Fig. 3). Upon entering the post-atretic segment, a polypoid mass was discovered (Fig. 4). Resection and an end-to-end anastomosis were performed.



Fig. 1: Plain erect abdominal radiograph at admission revealing multiple wide-based air-fluid levels.



Fig. 2: Barium enema examination revealing colon of normal caliber.

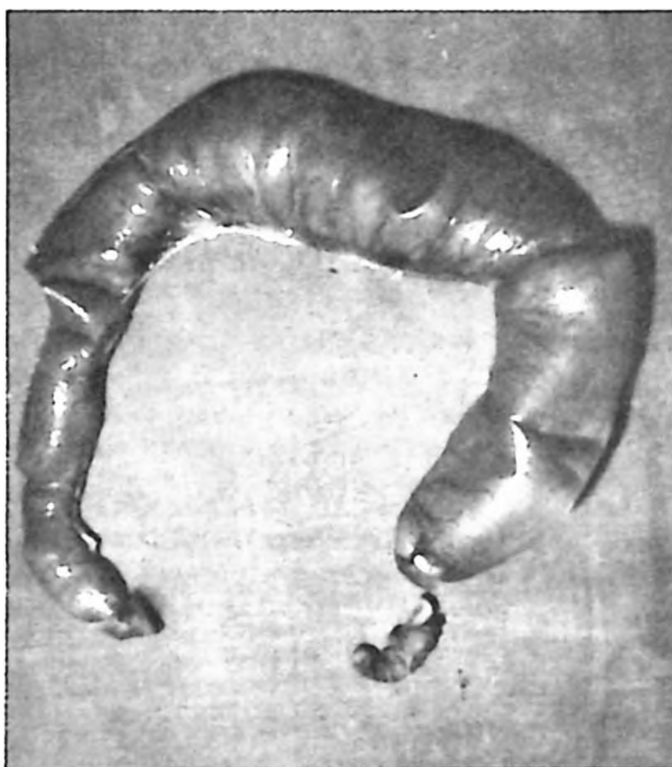


Fig. 3: Specimen of the atretic bowel segment with proximal dilatation.

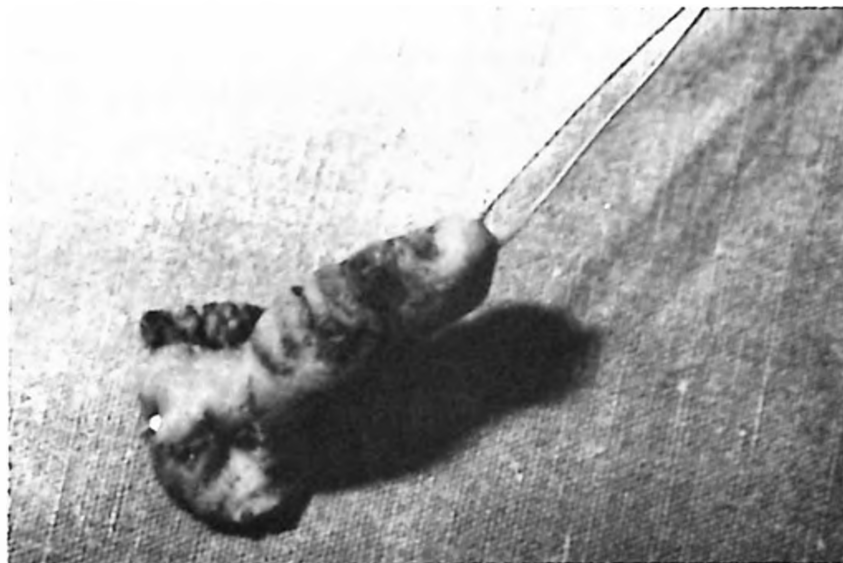


Fig. 4: Polypoid mass in the post-atretic segment.

Microscopically, the lumen and epithelium were not present in the cord, as in the atretic segment. However, the muscular layer was present, and there were focal calcifications in the serosa. The intestinal wall was recognized in the polypoid mass in the post-atretic segment. Ganglion cells were seen in the myenteric plexus of the pre-and post-atretic segments.

The postoperative course was uneventful. The patient was followed for one year, and she showed neither symptoms nor pathologic findings.

Discussion

Of the 50 reported cases of ileal atresia resulting from intrauterine intussusception, data is available for 35³⁻¹³, and of these, 85 percent are mature babies as in our case. The passage of bile-stained meconium, despite surgically proven atresia, implies that atresia occurred after the passage of meconium to the colon, and most probably late in fetal life. Such atresias may occur as a result of intrauterine intussusception³⁻¹³. Another characteristic finding in these atresias is the presence of a polypoid mass in the distal atretic segment³⁻¹³. In view of the foregoing, a diagnosis of ileal atresia caused by intrauterine intussusception was made in our patient.

Since the colon functions before intrauterine intussusception occurs, contrary to atresias resulting from other causes, barium enema reveals a colon of normal caliber, and meconium is found in the distal segment.

The passage of bile-stained meconium and the presence of a normal caliber colon on barium enema examination should not exclude the possibility of ileal atresia caused by intrauterine intussusception.

REFERENCES

1. Grosfeld JL. Jejunoileal atresia and stenosis. In Welch KJ, Randolph JG, Ravitch MM, et al (eds). *Pediatric Surgery* (4th ed), Vol 2. Chicago: Year Bk Med, 1986, pp. 838-848.
2. Büyükpamukçu N, Lister J. A review of ileal atresia. *Turk J Pediatr* 21:11, 1979.
3. Evans CH. Atresia of gastrointestinal tract. *Surg Gynecol Obstet* 92:1, 1951.
4. Rachelson MH, Jernigan JP, Jackson WF. Intussusception in the newborn infant with spontaneous expulsion of the intussusceptum: a case report and review of the literature. *J Pediatr* 47:87, 1955.
5. Parkkulainen KV. Intrauterine intussusception as a cause of intestinal atresia. *Surgery* 44:1106, 1958.
6. Gherardi GJ, Fisher JH. Atresia of the small intestine produced by intussusception in utero. *N Engl J Med* 264:229, 1961.
7. Spencer R. The various patterns of intestinal atresia. *Surgery* 64:661, 1968.
8. Grosfeld JL, Clatworthy HW Jr. The nature of ileal atresia due to intrauterine intussusception. *Arch Surg (Chicago)* 100:714, 1970.
9. Todani T, Tabuchi K, Tanaka S. Intestinal atresia due to intussusception: analysis of 24 cases in Japan. *J Pediatr Surg* 10:445, 1975.
10. Pavri DR, Marshall DG, Armstrong RF, Gorodzinsky FP. Intrauterine intussusception: case report and literature review. *Can J Surg* 26:376, 1983.
11. Ein SH, Venugopal S, Mancor K. Ileocaecal atresia. *J Pediatr Surg* 20:525, 1985.
12. Woodall DL, Birken GA, Williamson K, Lobe TE. Isolated fetal-neonatal abdominal ascites: a sign of intrauterine intussusception. *J Pediatr Surg* 22:506, 1987.
13. Şenocak ME, Büyükpamukçu N, Hıçsönmez A. Ileal atresia due to intrauterine intussusception caused by Meckel's diverticulum. *Pediatr Surg Int* 5:64, 1990.