

Meckel's diverticulum: the lead point of intrauterine intussusception with subsequent intestinal atresia in a newborn

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Intussusception is a known complication of Meckel's diverticulum. However, intrauterine intussusception secondary to Meckel's diverticulum has unknown incidence. Our case describes the medical and surgical management of a newborn with intrauterine intussusception of a Meckel's diverticulum as a cause of the vascular malformation leading to ileal atresia. To our knowledge, this is the fifth case report of this phenomenon. In all cases, definitive management required exploratory laparotomy, bowel resection, and intestinal primary anastomosis. *Ann Pediatr Surg* 12:164–166 © 2016 Annals of Pediatric Surgery.

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Case report

Our patient was a male, weighing 2400 g, born at 37 weeks of gestation to a 20-year-old G1P1 African-American mother by vaginal delivery. A third-trimester prenatal ultrasound revealed intra-abdominal calcifications and dilated loops of the bowel. The patient was delivered without complications, and his Apgar scores were 8 and 9 at 1 and 5 min, respectively. By day 3 of life, the newborn had still not passed meconium. He was transferred from the delivery hospital to our tertiary-care institution for further evaluation and management.

Physical examination revealed a soft, but distended abdomen. The patient was initially managed by nil per os, nasogastric tube decompression, hyperalimentation, and broad-spectrum antibiotics. Plain abdominal radiography showed multiple dilated bowel loops with air fluid levels, but no evidence of free air (Fig. 1). A water-soluble enema was administered the following day (Fig. 2), which revealed no contrast passage beyond 5 cm proximal to the terminal ileum, indicative of bowel obstructive process.

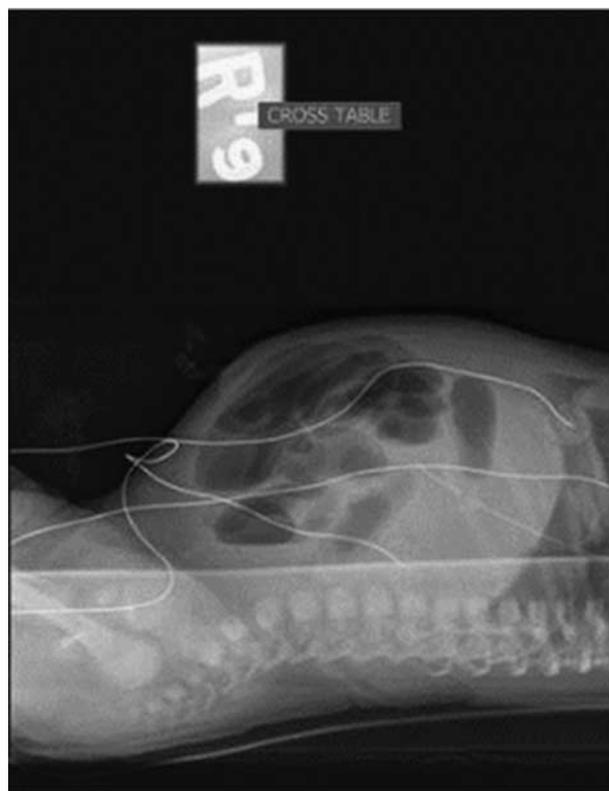
With failure of progression, the patient was taken to the operating room for an exploratory laparotomy. Upon entering the abdomen, a small amount of meconium-tinged ascitic fluid was encountered. The small bowel was eviscerated and examined from the ligament of Treitz to the terminal ileum. Near the terminal ileum, there was a transition zone with a dense collection of small bowel and a small rim of proximal necrosis. This segment of the bowel was resected back to viable margins, and an end-to-end primary ileo-ileal hand-sewn anastomosis was performed. Further exploration of the abdominal cavity revealed no additional abnormalities, and the abdomen was closed.

Examination of the specimen (Fig. 3) was performed on the back table. A longitudinal incision showed ileo-ileal small bowel intussusception with a rim of proximal necrosis. Meckel's diverticulum was discovered to be

intraluminal, consistent in location with the lead point of the intussusception.

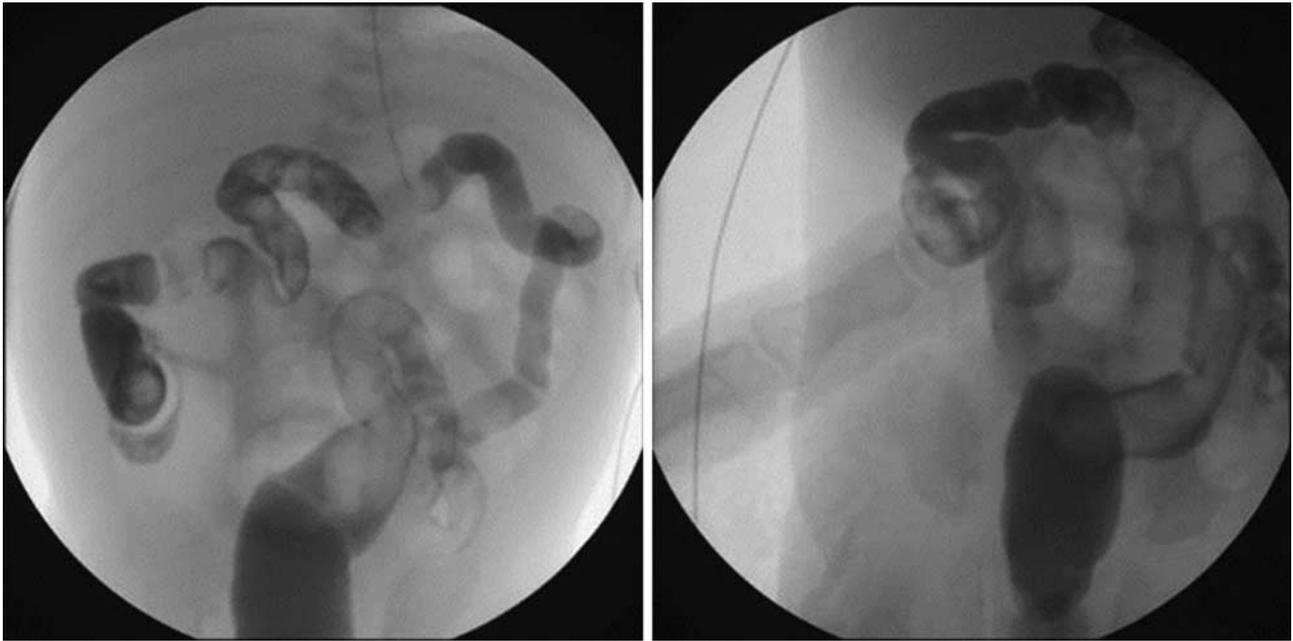
The patient was transferred to our neonatal ICU post-operatively and had an uneventful postoperative course. His bowel function gradually returned, and feeding was

Fig. 1



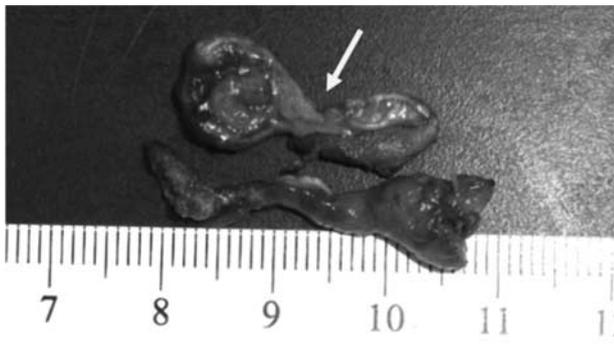
Plain abdominal radiograph showing multiple dilated bowel loops with air fluid levels, but no evidence of free air.

Fig. 2



A water-soluble enema revealed no contrast passage beyond 5 cm proximal to the terminal ileum, indicative of bowel obstructive process.

Fig. 3



Resected specimen showing ileo-ileal small bowel intussusception.

initiated on postoperative day 6. His diet was advanced and well tolerated. The patient was discharged home on postoperative day 12. Pathology examination revealed no evidence of heterotopic tissue.

Discussion

First described in 1508, and later characterized by the German anatomist Johann Friedrich Meckel, Meckel's diverticulum is a remnant of the omphalomesenteric duct that fails to obliterate during the 50th week of fetal development. Occurring in 1–3% of the population, it is the most common congenital anomaly of the gastrointestinal tract [1]. It is a true diverticulum, possessing all three layers of the intestinal wall. During development, the Meckel's diverticulum contains pluripotent cells that are capable of differentiation to various tissue types, predominantly gastric or pancreatic.

Often found incidentally during radiological studies or at the time of surgery, the majority of Meckel's diverticula remain asymptomatic [2]. The largest case series to date, by following-up 202 patients over the course of 15 years, has revealed a 4–6% lifetime risk of developing a complication, of which gastrointestinal bleeding, obstruction, intussusception, diverticulitis, and perforation are the most common [3]. In children, gastrointestinal bleeding due to the presence of acid-secreting gastric mucosa is the most common complication, with a peak incidence between 3 months and 3 years of age [4].

Intrauterine intussusception is a pathological event scarcely described in the literature. In 1960, Talwalker [5] archived 26 known cases of intussusception in newborns that were reported in the literature. At the time, the author noted a 'cause and effect' relationship between prenatal intussusception and intestinal atresia. In 1975, Todani *et al.* [6] reported 24 cases from Japan of intestinal atresia due to intrauterine intussusception. Since then, additional isolated cases of intrauterine intussusceptions in relation to intestinal atresia have been reported [7–11]. Surgical intervention remains the definitive for childhood intussusception in the presence of complete small bowel obstruction, failure of hydrostatic reduction, the third recurrence following radiographic reduction, or peritonitis on examination.

Intussusception is a known complication of Meckel's diverticulum. However, to date, intrauterine intussusception secondary to Meckel's diverticulum has unknown incidence. In fact, only four reported cases are known to exist [12–15]. The exact contribution of the Meckel's diverticulum to the intrauterine intussusception remains unclear – whether it is by inversion or en-bloc invagination. It is presumed, however, that the Meckel's

diverticulum serves as a pathological lead point to which the bowel intussuscepts with failure for spontaneous reduction. It is likely that ischemia of the intussusceptions led to perforation and circumferential intestinal wall loss, which healed with an interposing segment having necrosed, leading to atresia being discovered at birth. Ileal atresia and/or perforation had occurred in all four reported cases. Furthermore, all cases required exploratory laparotomy, bowel resection, and intestinal primary anastomosis. To our knowledge, ours is the fifth case report of this phenomenon.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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