OPEN ACCESS

Case Report

Submitted: 26-08-2023
Accepted: 06-01-2024

DOI: https://doi.org/10.47338/jns.v13.1243

A remarkable case of intrauterine intussusception, ileal atresia, and complicated meconium ileus: A case report

Jonathan Hencke,* Oliver Diez, Steffan Loff

Department of Paediatric Surgery, Olgahospital, Klinikum Stuttgart, Germany

Correspondence*: Jonathan Hencke, Kinderchirurgische Klinik, Olgahospital, Kriegsbergstraße 62, D-70174 Stuttgart, Germany. **E-mail**: j.hencke@klinikum-stuttgart.de

KEYWORDS

Fetal ascites,
Ileal atresia,
Cystic fibrosis

ABSTRACT

Background: Intrauterine intussusception, although very rare (<2%), is a potential cause of intestinal atresia. It usually manifests as bowel obstruction during the first hours or days of life.

Case Presentation: We report a case with fetal ascites prompting early Cesarean section, with subsequent percutaneous abdominal drainage and laparotomy. Intraoperative findings showed meconium peritonitis and type IIIa ileal atresia with intussusception of the distal part, with the necrosed intussusceptum likely causing perforation and ileal atresia; in addition, the distal ileum demonstrated signs of meconium ileus. After ileostomy creation, the postoperative course was uneventful. Cystic fibrosis was excluded via a sweat test.

Conclusion: Prompt diagnosis and management of neonatal intestinal obstruction secondary to the unusual combination of fetal ascites, intussusception, complicated meconium ileus, and ileal atresia results in a favorable outcome.

INTRODUCTION

Several prenatal intestinal events may necessitate surgical intervention shortly after birth; among them are prenatal volvulus, meconium ileus (MI), or intestinal ischemia. These events can result in perforation with meconium peritonitis and in some cases, the bowel necrosis causes jejunal or ileal atresia (IA). Additionally, in-utero intussusception has similar consequences, albeit relatively uncommon (less than 2% of jejunal or ileal atresias) [1].

CASE REPORT

A male newborn, at 36+1 weeks of gestation, was transferred to our NICU from a rural hospital following a Cesarean section due to suspected hydrops. The fetus had developed ascites and gained about 1 kg within the prior week. After birth, the abdomen was enormously distended, however, other features of hydrops were absent. A pigtail catheter was placed in the left lower quadrant draining more than 100 ml of clear yellow liquid. An abdominal radiograph showed only a few air-filled bowel loops (Fig. 1). Due to suspicion of a surgical cause, the patient was urgently planned for laparotomy.

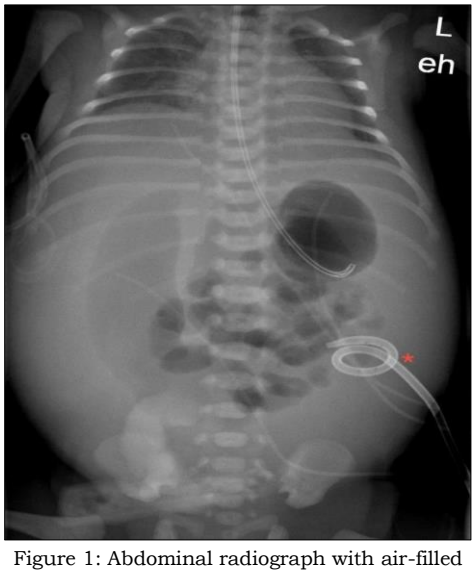

Figure 1: Abdominal radiograph with air-filled bowel loops and ascites despite drainage with pigtail catheter (*)

Upon opening the peritoneum, more fluid gushed out and the bowel exhibited typical coatings for meconium peritonitis. A type IIIa IA was discovered 66 cm after Treitz's ligament; the distal part measured 23 cm up to the ileocecal valve. The colon and the remaining abdomen appeared normal apart from the

meconium peritonitis. The discrepancy difference between the proximal and distal bowel at the atresia was approximately 3:1. The configuration of the ileal atresia was remarkable: while the proximal end was completely closed, the distal part was invaginated (Fig. 2); upon enterotomy, the invaginated part continued into a dry green tissue (Fig. 3). The distal ileum contained hard fragments of meconium reminiscent of meconium ileus. These intraoperative findings in conjunction with the prenatal ascites may fit the diagnosis of an intrauterine intussusception (IUI) with the subsequent necrosis of the affected bowel and perforation, or a complicated meconium ileus with additional intrauterine intussusception.

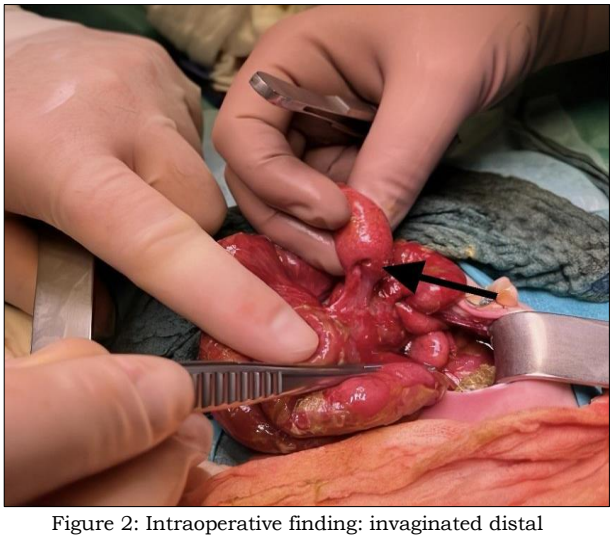

Figure 2: Intraoperative finding: invaginated distal bowel at ileal atresia (arrow)

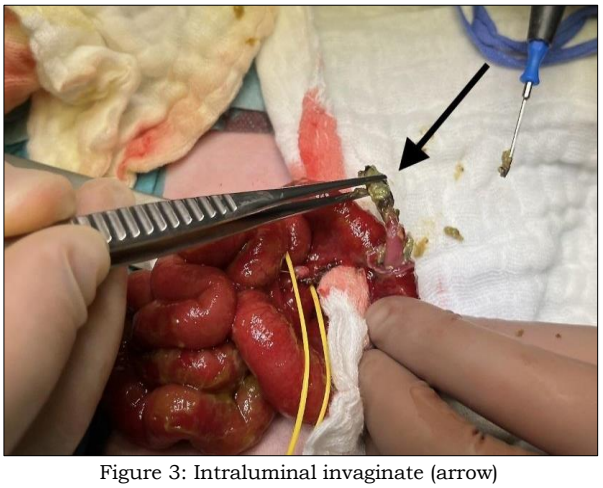

Figure 3: Intraluminal invaginate (arrow)

A short segment of the ileum was resected on both ends and an ileostomy was created; a primary anastomosis was avoided due to the meconium peritonitis and edema. The postoperative period was uneventful. After a contrast study of the distal limb, chyme re-feeding was started and well-tolerated. Cystic fibrosis was suspected due to the hard meconium plugs but could be excluded by the sweat test. The stoma was reversed at three months, without any further complications.

DISCUSSION

Fetal ascites may represent the first and only detected sign of an IUI, however, it is not specific [2]. Unlike other cases, there was no significant bowel dilatation noted both prenatally on ultrasound and also not on the abdominal radiograph after birth in our patient. This suggests that the event likely occurred later in pregnancy. However, the exact etiology and order of the separate pathologies in this case may not be ascertained. Perforation and meconium peritonitis are typical complications of meconium ileus, with the IUI potentially being a concurrent condition. Conversely, apart from the IUI, we did not identify any sites of (occult) perforation.

While there are reports and series documenting intrauterine intussusception (IUI), many cases remain undetected during pregnancy and for several hours or even days after birth until symptoms of bowel obstruction manifest [3]. The most frequently reported subsequent types of ileal atresia are type II and IIIa [4]. Similar cases involving IUI, ileal atresia (IA), and ascites/peritonitis are summarized in Table 1. Despite several reports of IUI and IA, only a few were accompanied by perforation and/or peritonitis. Specifically, there are only five cases documented with prenatal ascites, of which three had transient ascites and two, like our case, had accumulating ascites [2,5]. No cases involving both IUI and meconium ileus (MI) were found. Furthermore, some reports of neonatal intussusception in the presence of a meconium plug have been published, indicating a potential relationship between intraluminal meconium obstruction and the development of intussusception.

IUI is generally considered a rare cause of intestinal atresia, while vascular accidents or a volvulus being more common. The diagnosis of IUI and its sequelae is usually only made at the time of laparotomy [6], although prenatal ultrasound findings like fetal ascites, pseudocysts, calcifications, or dilated bowel loops indicate an intestinal obstruction that may require surgery postnatally. Therefore, parents with these prenatal findings should be counseled to deliver at a center with available pediatric surgery services [7].

This case underscores the association between fetal ascites, intrauterine intussusception (IUI), and meconium peritonitis, while also emphasizing the favorable prognosis similar to that observed in cases of intestinal atresia with prompt management.

Acknowledgements: Nil

Conflict of Interest: None.

Source of Support: Nil

Consent to Publication: Author(s) declared taking informed written consent for the publication of clinical

photographs/material (if any used), from the legal guardian of the patient with an understanding

that every effort will be made to conceal the identity of the patient, however it cannot be guaranteed.

Author Contributions: Author(s) declared to fulfil authorship criteria as devised by ICMJE and approved the final version.

Table 1: Similar cases with IUI and IA. Abbreviations: GA = gestational age, FT = full term, PT = preterm, NPU = no prenatal ultrasound

Report	Number of cases	GA	Prenatal ascites	Prenatal bowel calcifications	Bowel obstruction on day:	Dilated bowel loops on radiograph	Site of IUI	Complete atresia	Perforation / Peritonitis
Todani et al. 1975	2	FT	NPU	NPU	2	+	Ileum	+	-
		PT	NPU	NPU	2	+	Ileum	+	-
Adejuyigbe 1990	1	FT	NPU	NPU	2	+	Ileum	+	-
Kelly, Singh 1991	1	28	NPU	NPU	4	+	Ileum	+	+
Nguyen et al. 1995	2	31	-	+	1	+	Ileum	+	+
		FT	NPU	NPU	2	+	Ileum	+	-
Zehra Gündögü et al. 1996	1	FT	NPU	NPU	3	+	Ileum (Meckel)	+	-
Wang et al. 1998	4	40	NPU	NPU	1	+	Ileum	+	-
		40	NPU	NPU	2	+	Jejunum	+	-
		38	NPU	NPU	2	+	Ileum	-	+
		38	NPU	NPU	1	+	Jejunum	-	-
Rattan et al. 2000	1	FT	NPU	NPU	1	+	Ileum	+	-
Shimotake et al. 2000	1	FT	+	+	1	-	Ileum	+	+
Kilic et al. 2003	1	FT	NPU	NPU	1	+	Ileum (Meckel)	+	-
Mcheik et al. 2003	1	FT	-	+	1	+	Ileum	+	-
Yang et al. 2004	2	39	+	+	1	-	Ileum	+	+
		34	++	-	1	+	Ileum	+	-
Lee et al. 2005	1	28	NPU	NPU	6	+	Ileum	+	-
Huang et al. 2007	1	FT	-	-	2	+	Ileum	-	-
Lin et al. 2007	1	37	+	+	1	+	Ileum (Meckel)	+	+
Pueyo et al. 2009	1	FT	-	-	2	+	Ileum	+	-
Sarin 2010	1	40	NPU	NPU	1	+	Ileum	+	+
Gudi et al. 2011	1	37	-	+	1	+	Ileum	+	+
Kim et al. 2011	1	33	++	-	14	-	Ileum	-	+
Deshmukh et al. 2012	1	FT	-	-	1	+	Jejunum	+	-
Husaric et al. 2012	1	36	NPU	NPU	1	+	IC-junction	+	-
Ohuoba et al. 2013	1	39	-		1	+	Ileum	+	-
Chouik et al. 2014	1	FT	-	-	3	+	IC-junction	+	-
Joshi et al. 2015	1	FT	NPU	NPU	5	+	Jejunum	-	-
Le et al. 2016	1	37	-	+	3	+	Ileum (Meckel)	-	+
Carine et al. 2020	1	38	-	-	1	+	Ileum	+	-
Zenk et al. 2022	1	FT	-	-	5	+	Ileum	+	-
Our report	*1*	*36*	++	-	*1*	-	*Ileum*	+	+

REFERENCES

1. Dalla Vecchia LK, Grosfeld JL, West KW, Rescorla FJ, Scherer LR, Engum SA. Intestinal Atresia and Stenosis: A 25-Year Experience with 277 Cases. Arch Surg.1998;133(5):490–7.
2. Yang JI, Kim HS, Chang KH, Hong J, Joo HJ, Ryu HS. Intrauterine intussusception presenting as fetal ascites at prenatal ultrasonography. Am J Perinatol, 2004;21(04):241-6.
3. Wang NL, Yeh ML, Chang PY, Sheu JC, Chen CC, Lee HC, et al. Prenatal and neonatal intussusception. Ped Surg Int.1998;13:232-6.
4. Chouikh T, Charieg A, Mrad C, Ghorbel S, Saada S, Benkhalifa S, et al. Intestinal atresia caused by intrauterine intussusception: A case report and literature review. J Pediatr Surg Case Rep.2014;2:203-5.
5. Kim MW, Bae HS, Boo YJ, Lee JH, Hong SC, Oh MJ, et al. Meconium Peritonitis Associated with Intrauterine Intussusception: A Case Report. Kor J Ultrasound Obs Gyn. 2011;13(1):23-7.
6. Joshi SB, Kinhal V, Desai M, Choudhari FUR. A rare case of jejunal atresia due to intrauterine intussusception. J Clin Diagn Res. 2015;9(9), PD30.
7. Shyu MK, Shih JC, Lee CN, Hwa HL, Chow SN, Hsieh FJ. Correlation of prenatal ultrasound and postnatal outcome in meconium peritonitis. Fetal Diagn Ther. 2003;18(4):255-61.