

Case Report

Jejunal atresia (type 3A) with jejunoileal fistula: a case report of jejunal atresia with rare presentation

Surishta K. Rana*, Sargam Verma

Department of Surgery, Sri Guru Ram Dass Institute of Medical Sciences and Research (SGRDIMSR), Amritsar, Punjab, India

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***Correspondence:**

Dr. Surishta K. Rana,

E-mail: drsrishtyrana66@gmail.com

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ABSTRACT

A six-month-old girl presented to pediatrics emergency with features suggestive of subacute presentation of intestinal obstruction. Laparotomy revealed type 3A jejunal atresia with jejunoileal fistula. The proximal jejunal end having narrow pinpoint connection with distal segment at 10 cm distal to atretic site. A mesenteric defect noted in between two segments. A long thick fibrovascular band found arising at the mesenteric defect site and distally attached to splenic flexure.

Keywords: Jejunoileal atresia, Entero-enteric fistula, Small bowel atresia

INTRODUCTION

Intestinal atresia is a congenital abnormality where there is significant stenosis or complete absence of a portion of the intestine. Ileal and jejunal atresia are usually described together as jejunoileal atresia (JIA). JIA is a common cause of intestinal obstruction in neonates. It is seen in 1 in 5000 to 1 in 14 000 live births.¹ About 33% of the affected children are born prematurely, and incidence is distributed equally in both sexes.^{2,3} Less than 10% of cases of JIA are seen with extra-abdominal organ abnormalities, and this has been attributed to the late occurrence of localized vascular compromise in-utero. Very few cases of delayed presentation of Type IIIa jejunal atresia beyond the neonatal period have been reported. This case has presented after five and half month of age.

CASE REPORT

A 5-month-22-day-old female baby presented to our center with the chief complaint of vomiting, irritability, decreased feeds and inability to pass stool for 2 days. The vomitus was described to be yellow greenish color,

multiple episodes, gradual onset, non-projectile and non-blood stained. There was no history of pain abdomen. Similar episodes of such abdominal complaints were seen in the patient at 1.5 months and 3 months age for which he was hospitalized by nearby physician for 3-4 days and managed with medicines but complete evaluation was not there. The baby was delivered by LSCS at term with the birth weight of 3900 grams.

She cried immediately after birth and was breastfed within the first hour after delivery. The mother started antenatal clinic at 12 weeks of gestation and made 5 visits and received supplements according to the national guidelines. She was tested negative for retroviral serology and syphilis. Mother was normotensive with normal blood sugar levels. Baby has normal course during perinatal period with no asphyxia, no infection and no hospital admission required. Baby passed meconium normally on day 1 of life. There is history of hospitalization at 6th day of birth for neonatal jaundice. Developmental milestones were achieved normally by the child. Patient was started for weaning 20 days back by semisolid food. On examination the baby was conscious, active and his vitals were stable. Body weight on

admission was 3100 gm that was inappropriate for age. Height was also below standard deviation suggesting of poor growth. Abdominal examination showed symmetrically distended abdomen, with no guarding or rigidity was elicited on palpation and had hyper tympanic percussion note with exaggerated bowel sounds. The rectum had little soft fecal matter. Isolated cleft palate was also noticed on examination in this baby. Subacute intestinal obstruction was suspected and abdominal ultrasound and erect abdominal X-ray were done.

Abdominal X-ray showed dilated bowels with multiple air-fluid levels, abdominal ultrasound showed prominent bowel loops, 2.5 mm in caliber and multiple lymph nodes along the mesentery and left sided hydronephrosis. Contrast enhanced CT scan whole abdomen (Figure 1) was already done prior referral and was suggestive of small intestinal obstruction with transition point in the distal jejunal loops, mesenteric lymphadenopathy and dysmorphic small sized spleen. She was kept nil orally, intravenous fluids administered and scheduled for an emergency laparotomy after examining and thoroughly studying reports.

Management

Patient underwent exploratory laparotomy. During exploration bowel operative findings noticed were- Proximal grossly dilated jejunal loop of 30 cm length from duodenojejunal flexure was present. Adhesions with fibrous band were present at the culmination of this dilated loop. After dissection gently, there was a narrow pinpoint end to side communication towards the mesenteric side of distal collapsed loop, 10 cm distal from the distal blind atretic end. 10 cm of this initial portion of distal segment proximal to the fistula was collapsed with intact lumen inside and was lying free within the adjoining adhesions. Small V-shaped mesenteric defect was noted in between the proximal and distal atretic ends. A long fibrovascular band arising from the mesenteric defect site dissected during and adhesiolysis and distal end was traced attached to the splenic flexure of colon and was excised. Figure 2 and 3 shows end to side fistula with atretic distal free end and long fibrous band respectively. Distal small bowel was 70 cm in length till ileocecal junction and the bowel discrepancy noticed was 4:1 (proximal: distal). Free lying segment of distal bowel, till junction of fistulous communication (10 cm) was resected. Proximal and distal bowel ends freshened and fecal decompression of dilated segment was done.

Distal patency was confirmed by saline irrigation into distal segment and looking for small and large bowel filling. Caecum and proximal ascending colon were found free floating in this patient with no attachment to posterior parietal wall. Rest of the bowel examined was healthy and no other abnormality observed. Cheatle maneuver was used on distal bowel segment to address for the bowel discrepancy, following which the bowel

anastomosis was performed in two layers. Postoperatively, child kept in pediatric intensive care unit for observation. Strict nasogastric (NG) aspiration, blood /plasma transfused, intravenous antibiotics and fluids continued. After minimal NG secretions, and good bowel sounds, feed was started on postoperative day 5 and child passed stools on postoperative day 7. NG removed, full oral feeds were achieved by day 8 and child discharged in satisfactory condition on postoperative Day 10. On subsequent close follow ups for 5 months, patient found doing well.

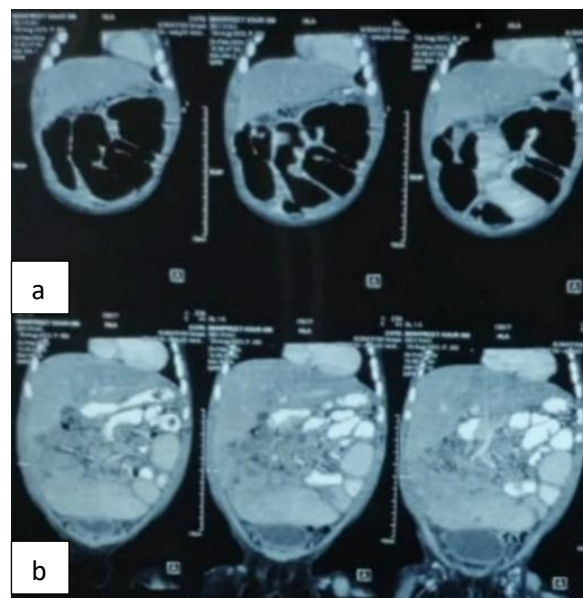


Figure 1 (a and b): CECT abdomen showing small intestinal obstruction with transition point in the distal jejunal loops, mesenteric lymphadenopathy and dysmorphic small sized spleen.

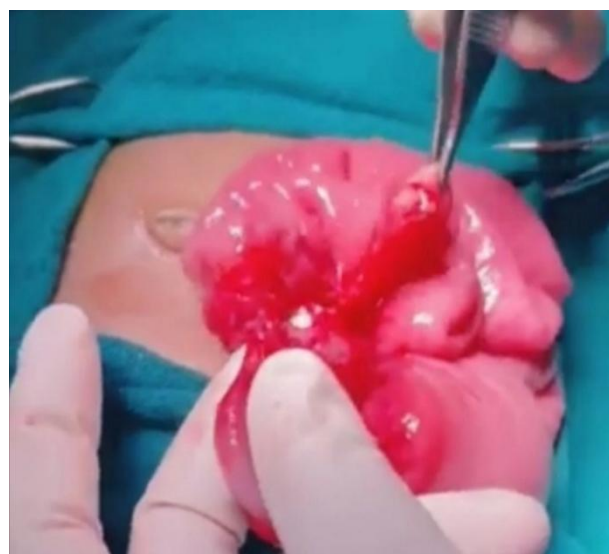


Figure 2: Dilated proximal jejunal loop outlined in green, collapsed distal free atretic end marked with yellow arrow and end to side enteroenteric fistula (jejunoileal) marked using blue arrow.

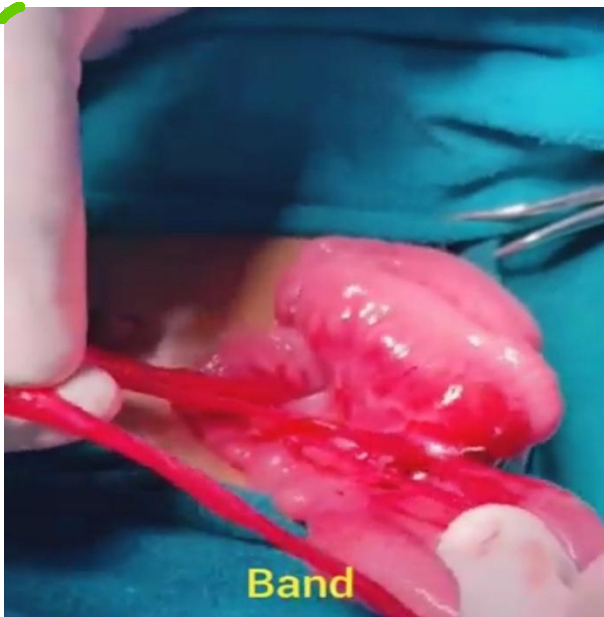


Figure 3: Long vascular band (marked using green arrow) arising from adhesions at mesenteric defect site and distal end attached to splenic flexure and also showing fistula site marked with yellow arrow.

DISCUSSION

Intestinal atresia is the most common congenital anomaly of small intestine with an incidence of 1.3–2.9 cases per 10,000 live births. Different theories have been described regarding the etiology. The most accepted etiology for jejunoileal atresias was proposed by Louw and Barnard which relates to late intrauterine mesenteric vascular accidents including volvulus, intussusceptions, snaring at umbilical ring, kinks, and bands.³

The patient was born at full term by LSCS having a birth weight of 3.6 kg. She was started on breastfeed on day 1 of life which she tolerated well and passed meconium on the same day. The child was not completely diagnosed till 6 months of age despite two short admissions and hospital stays. The surgical diagnosis was missed for this longer duration due to the less severity of previous clinical symptoms and got settled by conservative management by pediatrician in initial two visits. Also, another reason probably could be severe clinical symptoms for this underlying pathology could have manifested in response to weaning that was started recently. To the best of our knowledge, the communication of blind loop of intestinal atresia through entero-enteric fistula (EEF) has barely been documented previously.

Children with intestinal atresia typically present by 1-3 days of life with abdominal distention, bilious vomiting, and failure to pass meconium. Cases of intestinal atresia with delayed presentation have also been reported.^{2,3} Delayed presentation is seen mostly in Type I atresia with a defect in the diaphragm. The degree of obstruction in

Type I jejunal atresia depends on the size of the defect. Smaller the defect size, earlier is the presentation.⁴ Delayed presentation of intestinal atresia has also been seen when it is caused by intrauterine intussusception. Usually, these cases present within 48 hours, because of complete intestinal obstruction. Differentiation from the sequelae necrotizing enterocolitis (NEC) may be difficult. Uncertainty in diagnosis may lead to a delay in confirming the diagnosis. Considering that NEC sometimes merits conservative management while intussusception with intestinal atresia will need surgical intervention, a delay in diagnosis may affect the prognosis.⁵ Type 3A atresia show V-shaped mesenteric defect with missing communication between proximal and distal atretic ends, there might be presence of fibrous band connecting the end between the defect. This case is a type 3A atresia variation with jejunoileal fistula communication 10 cm beyond distal atretic end that is rarely documented. The cause of congenital atresia with such manifestation and clinical scenario, may be due to any of the late intrauterine mesenteric vascular accidents including volvulus, intussusceptions, snaring at umbilical ring, kinks, and bands.³ There was obvious presence of a long thick band in our index case that was joining adhesion site to the splenic flexure.

Cases of acquired intestinal atresia in NEC with spontaneous reconstitution of intestinal continuity through fistulization have been described. Ischemia and bowel necrosis can stir up inflammatory response which may result in affected bowel to adhere to adjacent bowel and eventually lead to fistula formations.⁶ The other cause acquired atresia described is adhesive band entrapping the mesentery and bowel segment but without any EEF.⁷ Our case had no abdominal symptoms during neonatal that rules out NEC as the underlying cause for this clinical manifestation. The cause for EEF in our patient might be antenatal perforation leading to peritonitis. The intrauterine inflammation between the bowel loops might have eventually leads to EEF.

CONCLUSION

The findings in the index case and the underlying etiologic mechanisms are likely to have been antenatal and may have important implications in understanding the pathophysiology of intestinal atresia and the antenatal healing processes. The rarity of such clinical case presentation of jejunoileal Type 3A with enteroenteric end to side fistula at 6 months age, makes this case report worth writing.

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