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Case Report

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# Patent vitello-intestinal duct with a rare presentation of reverse intussusception with appendicular agenesis: A case report

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**KEYWORDS**

Reverse Intussusception, Vitello-intestinal duct, Pediatric, infant, Congenital anomalies

**ABSTRACT**

Background: Vitello-intestinal duct (VID) is found in approximately 2% of newborns, with patency in about 6%. Failure of obliteration leads to some anomalies, most commonly Meckel's diverticulum. Protrusion of the small bowel is one of the rare presentations.

Case Presentation: A 10-month-old male infant presented with a prolapsed, irreducible polypoid-shaped loop of intestine protruding from the anterior abdomen. On exploratory laparotomy, patent VID was confirmed with appendicular agenesis and reverse intussusception. Gangrenous bowel was resected, followed by end-to-end ileo-ileal anastomosis, with a favorable outcome.

Conclusion: Patent VID with ileal intussusception and prolapse is a rare entity, which can be easily diagnosed clinically. A high index of suspicion is needed for any associated gut anomaly in these cases, like appendicular agenesis.

## INTRODUCTION

The vitello-intestinal duct (VID) gives sustenance to the fetus, until the placenta is developed. At approximately five weeks of gestation, the midgut undergoes rapid enlargement and protrudes into the umbilical cord, where its apex remains connected to both the VID and yolk sac.[1] Rotational error around the tenth week of gestation leads to bowel anomalies, the most frequent being Meckel's Diverticulum, patent VID, and exomphalos.[1]

The VID anomalies occur in approximately 2% of newborns, out of which 6% are patent, through which small bowel intussusception can occur, in around 20% of cases.[2]

Appendicular agenesis is exceptionally uncommon, with an incidence of 1:100,000 cases undergoing surgery for suspected acute appendicitis, diagnosed intraoperatively.[3].

## CASE REPORT

A 10-month-old male infant presented in pediatric emergency with the complaint of a bright red mass emerging from the abdomen, at the place of the umbilicus. According to the mother, after the dehiscence of the umbilical cord, the opening had never healed with subsequent redness of the surrounding area with blood and mucus discharge. At 3 months of age, she noticed a protrusion from the umbilical region, progressively increasing in size with passage of stool and flatus through it. There was a history of multiple episodes of vomiting with no passage of bowel per anus. General physical examination showed that the patient was irritable with excessive crying, dehydration, tachycardia with signs of shock. Abdominal examination revealed a firm and taut abdomen, with a bright red, polypoid shaped loop of intestine protruding from the anterior abdominal wall, at the level of the umbilicus. It was irreducible, bled on contact, and showed signs of impending gangrene. No other significant findings were seen on examination. Patient was resuscitated with intravenous fluids and inotropes were started. The baseline investigations revealed a raised C-reactive protein, low hemoglobin (9gm/dl), raised TLC (20,000/cmm). First line antibiotics were started. No radiological investigations were done. Exploratory laparotomy was performed, with right transverse paraumbilical incision. The terminal ileum had slid into the proximal ileum, giving rise to a reverse intussusception and subsequent gangrene, and thus, giving rise to Ram's horn appearance (Figure 1a and 1b). Additionally, appendix was not found even after thorough search, making it a congenital case of appendicular agenesis. The intraoperative diagnosis

was, therefore, of that of patent VID, with gangrenous reverse (retrograde) ileal intussusception and appendicular agenesis. Reduction of the intussuscepted ileal loop was tried but failed, hence, the gangrenous bowel was resected, followed by an end-to-end ileo-ileal anastomosis. The abdominal wall defect was repaired. The postoperative period was uneventful. On the eighth postoperative day, the patient was discharged in fair health. Subsequent follow up appointments after 1 week, 2 weeks and after 1 month showed no additional concerns or complaints.

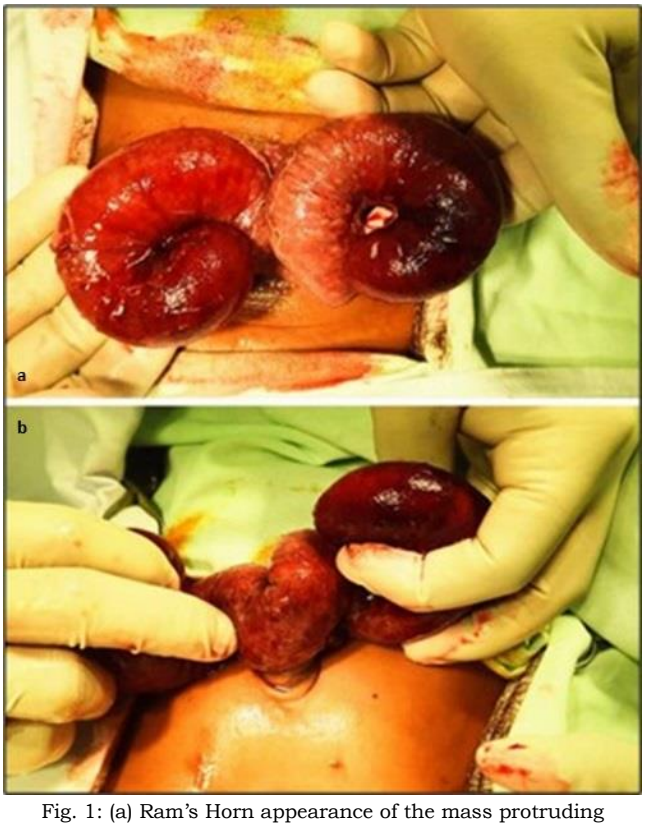


Fig. 1: (a) Ram's Horn appearance of the mass protruding through the umbilical defect. (b) Preoperative picture showing the prolapsed ileal loop with reverse intussusception.

## DISCUSSION

Patent VID, although rare, can lead to various intra-abdominal problems. The protrusion of the VID causes a T-shaped bulge in the intestine through the umbilicus, and in some cases, it may even result in a Ram's horn extending through the anterior abdominal wall. [2]

Ileal intussusception into the patent VID can be due to its large mouth and the shorter distance between the VID and the ileocecal valve in infants.[1] Diagnosis of patent VID is done from history and physical examination and prenatally using serial ultrasonography.[4] Carbimazole and methimazole treatment in mothers is seen to be associated with patent VID as well as choanal atresia.[5]

The diagnosis of a patent VID is typically based on the patient's history of fecal discharge from the umbilicus and findings from the physical examination. Investigations such as fistulograms may be performed when it is important to differentiate between patent VID and patent urachus, but they are not required in situations such as the ones shown here since they will not influence the surgical choice.[6]

Appendicular agenesis, as observed in our case, can arise from intrauterine vascular events, auto amputations caused by fibrous bands or appendicular atresia.[7] The case presented here pertained to type 3 variation of congenital absence of appendix.[8]

The exact incidence of patent VID with reverse intussusception is not known, with very few reported in literature.[1,9-15] Timely identification and surgical reduction represent the preferred treatment approach, although in our case, this was not achievable due to gangrenous bowel necessitating resection and ileo-ileal anastomosis.

To conclude, Patent VID with ileal intussusception and prolapse is a rare entity, but can be easily diagnosed by a thorough history taking and physical examination. The diagnosis of appendicular agenesis is purely intraoperative. Thus, we should keep a high index of suspicion whenever we encounter a case of patent VID for any associated gut anomaly, for timely intervention and good prognosis.

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