Journal of Pediatric Surgery Case Reports 20 (2017) 14–16

Contents lists available at ScienceDirect

Journal of Pediatric Surgery Case Reports

journal homepage: www.jpscasereports.com

Case Report

New-born born with patent vitellointestinal duct with prolapsed (intussusceptions) of proximal and distal ileal loop: A case presentation

Fariha Akil Fazal*, James Muturi Ndungu, Humdun Said, Josephat Njiru, Fred Kambuni

Section of Paediatric Surgery, Department of Surgery, University of Nairobi, Kenya

ARTICLE INFO

Article history:
Received 27 January 2017
Received in revised form
20 February 2017
Accepted 21 February 2017
Available online 27 February 2017

ABSTRACT

Failure of obliteration of vitellointestinal duct (VID) can present with a wide variety of congenital intestinal malformations. Patient can present with the anomaly itself or due to complications secondary to the anomaly like intestinal obstruction due to volvulus, intussusception or adhesions. Prolapsed ilial loops through a patent VID is a rare presentation of the above. To date only fourteen cases of this presentation have been reported in the English medical literature. We are reporting a case of the same in a new-born presenting with it from the time of birth, which to the best of our knowledge has not been reported before and therefore this the youngest reported case of its nature in the current English medical literature.

1. Background

During the fifth week of gestation, midgut enlarges rapidly and as it becomes too large for the abdominal cavity, it herniates through the umbilical cord. The apex of the herniated midgut is continuous with VID and yolk sac. Superior mesenteric artery forms the axis of this herniated midgut. Around tenth week of gestation herniated midgut returns back into the peritoneal cavity [1,2]. During this complex developmental process several anomalies may occur because of the complexity of the process. Examples include bowel atresias and stenoses, abnormalities of the vitellointestinal duct (Meckel's diverticulum, patent vitellointestinal duct (PVID), umbilical fistulas, umbilical sinus tracts, umbilical cysts and umbilical polyps), failure of ceacal descent, malrotation, malfixation, reversed bowel rotation and exomphalos [1–3].

2. Case presentation

An 18 h old male new-born was referred to Kenyatta National Hospital with a mass protruding from the umbilicus since birth. Baby was born at full term via simple vaginal delivery to a parous 3 mother. The mother as well as the referral letter confirmed that the baby was born with the mass. Antenatal scans were reported to be normal and there was no history of polyhydramnios. Birth weight was 2500 g and Apgar scores were 9/1, 10/1, 10/10.

On examination at 18 h, new-born was comfortable and alert. Abdominal examination revealed a bright red 'Y' shaped loop of small intestine was protruding from the umbilical ring. It was fixed to the umbilicus, with easily bleeding mucosa and irreducibility. Rest of the abdomen was not distended nor tender (Figs. 1 and 2). Bowel sounds were absent. There was no bilious aspirate in the orogastric tube. Anal opening was normally placed and patent, however, he had not passed any meconium. Patient had passed clear urine twice and the bladder was not palpable. Rest of the systemic examination was normal.

Laboratory investigation including full hemogram, biochemistry and coagulation profile were normal. A plain abdominal radiograph in supine and lateral decubitus position showed normal bowel pattern with no air fluid levels and air in the pelvis (Fig. 3).

The neo born was started on intravenous fluids, antibiotics intravenous ceftriaxone (100 mg/kg/day) and metronidazole (30 mg/kg/day) and intramuscular vitamin K 5 mg.

Patient was prepared for an emergency laparotomy because of the prolapsed ileal loops. Examination under anaesthesia revealed that prolapsed bowel loops were intussuscepted proximal and distal ileal segment through patent VID. There was no mass palpable underneath to suggest any enteral or urachal cyst.

We made a transumblical circumferential incision in to the

* Corresponding author.
E-mail address: Farihahaleem@hotmail.com (F.A. Fazal).

http://dx.doi.org/10.1016/j.epsc.2017.02.014

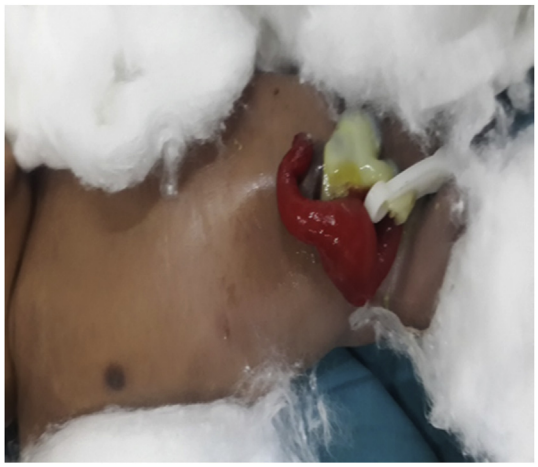

Fig. 1. 'Y' Shaped loop of small intestine protruding from the umbilicus.

peritoneum. The outer surface of the prolapsed loop was firmly adherent to all the layers of the anterior abdominal wall and therefore had to be meticulously dissected of the anterior abdominal wall. Continuity with proximal and distil bowel loop as well as the relationship with the umbilical ring and appendix was confirmed. Then both proximal and distil loops were gently reduced with ease. There was no bowel wall edema and the mucosa was health. After complete reduction defect connecting the small bowel with umbilicus was found to be approximately 2 cm wide. It was resected and anastomosed after ligating the aberrant blood supply to the Meckel's diverticulum (Fig. 4). Umbilicus was closed with a purse string suture (Fig. 5).

Patient passed meconium on first operative day after which the orogastric tube was removed. He was started on breast milk on the second day. Subsequently the patient was discharged on the forth Post-operative day and has been doing well for over six months of follow up.

3. Discussion

Vitellointestinal duct (VID) or Omphalomesenteric duct connecting the primitive gut to the yolk sac usually obliterates around the seventh or eighth week of gestation. Failure to obliterate may lead to variety of congenital anomalies including; Meckel's diverticulum, vitelline cord, umbilical sinus, enteric fistula and or haemorrhagic umbilical mass [1–4].

Double intussusception of the small bowel through a patent VID

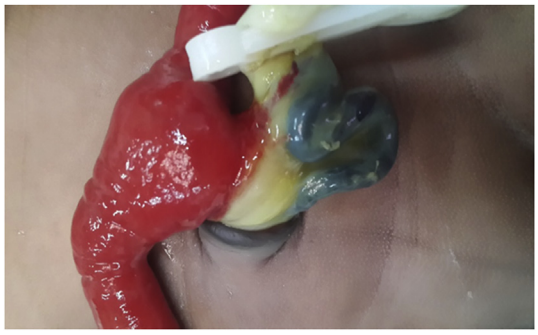

Fig. 2. Closer view of the loop of bowel revealed prolapsed small bowel mucosa excreting bile at one end.

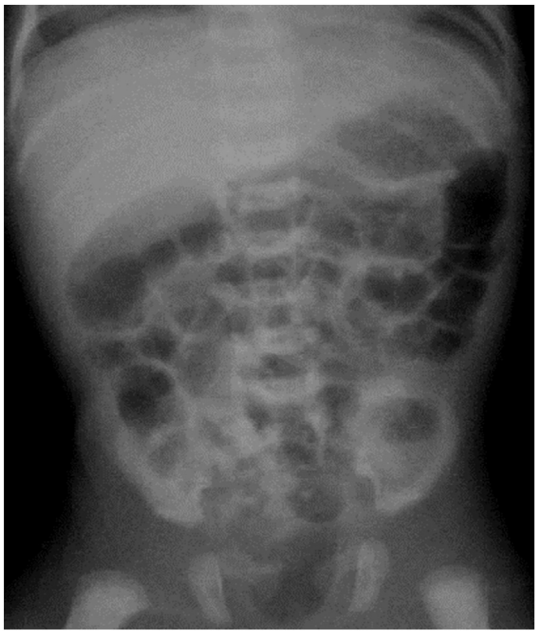

Fig. 3. Preoperative plain abdominal radiograph showing normal bowel pattern.

is very rare entity. To date only 14 cases have been reported in the English medical literature. These include: Elebute and Ransome-Kuti in 1965 [5], Davis and Kehm in 1967 [6], Lal and Dhall in 1976 (reported three cases) [7], Rohatgi and Gorthi in 1984 [8], Gvalani et al., in 1985 [9], Mohite et al., in 2007 [10], Zea et al., in 2009 [11], Agrawal and Memon in 2010 [12], Singh et al. 15 in 2011 [13], Pauleau et al., in 2012 [14], Patel et al., in 2013 [15] and Kadian in 2015 [16]. Moreover, to the best of our knowledge there is no reported case of a new born presenting with patent VID with prolapsed (intussusceptions) of proximal and distal ileal loop.

Antenatal diagnosis of a PVID anomaly can be confused with hernia of the umbilical cord or exomphalos minor which fails to obliterate on serial ultrasonography. Early diagnosis of PVID anomalies mandates surgeons to have a high index of suspicion in suspected cases. This would lead to prompt diagnosis and management of this rarity and save the neonate the morbidity and mortality associated with subsequent intestinal obstruction and ischemia.

As noted in the previous case reports on the topic [1,2,17], outcome is highly dependent on time of presentation, early diagnosis, associated anomalies and the size of defect. When patients present late the approach must be more conservative with formal laparotomy and or ileostomies. However, if patients present early a trans umbilical approach can be taken comfortably with good outcomes. Clinicians especially general practitioners need to be vary of a possible VID malformation in susceptible neonates when they present with intermittent discharge from the umbilicus.

It is the widely patent type of PVID which are more likely to present with either complete or partial prolapse of ileum through the defect. The prolapse itself is probably caused by a sudden increase in intraabdominal pressure associated with straining in neonates with a widely patent VID. Moreover, since the distance between the ileocecal valve and VID is shorter in neonates it leads to higher intraluminal pressure causing double intussusception [16].

Resection and anastomosis is preferable to wedge resection of PVID because of the associated risk of ectopic gastric or pancreatic

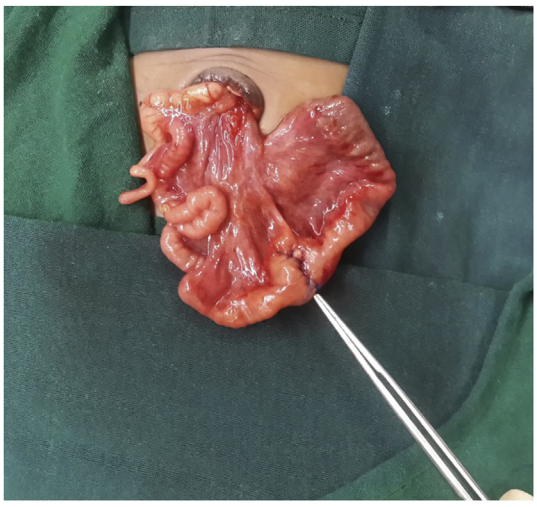

Fig. 4. End to end anastomosis after reduction of intussuceptions and resection of bowel. Anatomic relationship to the ileocecal valve and aberrant blood supply to the Meckel's diverticulum.

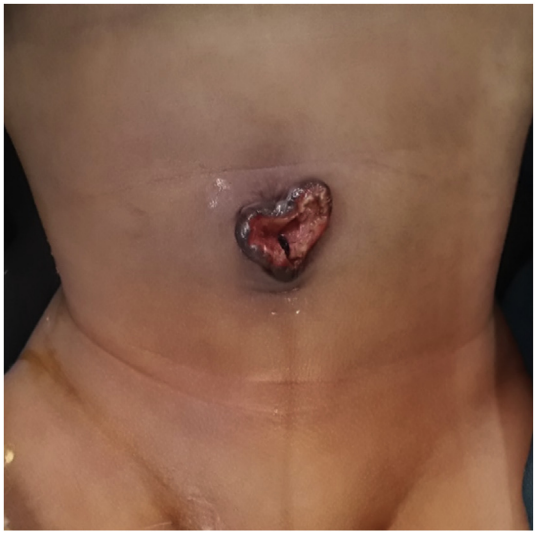

Fig. 5. Umbilicus closed with a purse string suture.

mucosa as well as the associated ischemia secondary to intestinal obstruction and strangulation [17].

We believe that this is a neonatal emergency which must be dealt with urgently due to the associated intestinal obstruction from intussusception, strangulation and gangrene of the prolapsed intestinal loop. In our case, we were able to do a primary closure of the VID following reduction of the prolapse due to the early presentation of the patient. Transumbilical approach was also possible because of the early presentation with good cosmetic outcome.

All reported cases before this presented during late neonatal period or infancy. This patient however, presented with it from the time of birth, which makes us wonder whether perinatal events lead to its earlier presentation or was it a prenatal event. Therefore, clinician must have a very high index of suspicion for diagnosing it timely, though its management will always follow principals of standard neonatal bowel surgery.

4. Conclusion

Patent vitellointestinal duct with prolapsed (intussusceptions) of both proximal and distal ileal loop is a very rare occurrence together with being a neonatal surgical emergency due to the associated morbidity.

Conflict of interest

None.

Acknowledgment

Consent for publication obtained from the parents of the patient.

References

[1] Mohite PN, Bhatnagar AM, Hathila VP, Mistry JH. Patent vitellointestinal duct with prolapse of inverted loop of small intestine: a case report. J Med Case Rep 2007;1:49.
[2] Zea M, Chana R, Anees A, Khan S. Inverted ileal prolapse through patent Vitellointestinal duct: a case report. Internet J Pediatr Neonatol 2008;10(2).
[3] Stone PA, Hofeldt MJ, Campbell JE, Vedula G, DeLuca JA, Flaherty SK. Meckel diverticulum: ten-year experience in adults. South Med J 2004;97:1038–41.
[4] Cullen JJ, Kelly KA, Moir CR, Hodge DO, Zinsmeister AR, Melton 3rd LJ. Surgical management of Meckel's diverticulum. An epidemiologic, population-based study. Ann Surg 1994;220:564–9.
[5] Elebute EA, Ransome-Kuti O. Patent vitello-intestinal duct with ileal prolapse. Arch Surg 1965;91:456–60.
[6] Davis RM, Kehm RW. Omphalocele with patent vitellointestinal duct and ileal prolapse. Am J Surg 1967;113:571–3.
[7] Lal MM, Dhall JC. Ileal prolapse through patent vitello-intestinal duct: a report of 3 cases with review of literature. Indian Pediatr 1976;13:571–3.
[8] Rohatgi M, Gorthi SN. Omphalocele with patent omphalomesenteric duct and ileal prolapse. Indian J Pediatr 1984;51:119–23.
[9] Gvalani AK, Acharya AG, Rao RV. Ileal prolapsed through a patent vitelloin-testinal. Indian Pr 1985;38:629–32.
[10] Mohite PN, Bhatnagar AM, Hathila VP. Patent vitellointestinal duct with prolapse of inverted loop of small intestine: a case report. J Med Case Rep 2007;1:49.
[11] Zea M, Chana R, Anees A, Khan S. Inverted ileal prolapse through patent vitellointestinal duct: a case report. Internet J Pediatr Neonatol 2009;2013:2.
[12] Agrawal S, Memon A. Patent vitellointestinal duct. BMJ Case Rep 2010;2010. pii bcr1220092594.
[13] Singh S, Pandey A, Ahmed I, Rawat JD, Sharma A, Srivastava NK, et al. Prolapse of bowel via patent vitello intestinal duct—a rare occurrence. Hernia 2011;15:567–9.
[14] Pauleau G, Commandeur D, Andro C. Intestinal prolapse through a persistent omphalomesenteric duct causing small-bowel obstruction. S Afr J Surg 2012;50:102–3.
[15] Patel RV, Kumar H, Sinha CK, Patricolo M. Neonatal prolapsed patent vitel-lointestinal duct. BMJ Case Rep 2013;2013.
[16] Kadian YS. Patent vitellointestinal duct with inverted ileal loop prolapse: a rare presentation. Onc Gas Hep Rep 2015;4:95–7.
[17] Pathak Ashish, Agarwal Nitin, Singh Poonam, Dhaneria Mamta. Prolapse of inverted ileal loops through a patent vitellointestinal duct. BMJ Case Rep 2015;2015. bcr2015211563.