Case Report

Access this article online
Website: www.afrjpaedsurg.org
DOI: 10.4103/0189-6725.160411
Quick Response Code:

Spontaneous ileal perforation complicating low anorectal malformation

TiJesuni Olatunji, Matthias Igoche, Pascal Anyanwu, Emmanuel A. Ameh

ABSTRACT

Anorectal malformation is a common anomaly in neonates. Although colorectal perforations have been reported as a complication, ileal perforation is rarely encountered. This is a report of a 2-day-old boy presenting with a low anorectal malformation, complicated with ileal perforation, necessitating laparotomy and ileal repair. Anoplasty was done for the low anomaly. Early presentation and prompt treatment of anorectal malformations is important to prevent such potential life threatening complication.

Key words: Ileal perforation, low anorectal anomaly, spontaneous

INTRODUCTION

Gastrointestinal perforations in neonates are uncommon. Although physical examination of the perineum is often sufficient to diagnose anorectal malformation, delay in diagnosis is not uncommon[1,2] and this may lead to complications. Spontaneous intestinal perforation is a serious complication in infants with anorectal malformation and has been reported to occur in the colon and rectum, but perforation in the ileum has not been reported. The aim of this case report is to raise awareness to this complication and highlight the importance of early diagnosis in avoiding such complication.

CASE REPORT

A 2.5-day-old male newborn was referred from a peripheral hospital with progressive abdominal distension, respiratory difficulty, and inability to pass meconium since birth. The infant was product of term gestation, labour and delivery were uneventful. Physical examination showed severe dehydration and respiratory distress but no cyanosis. Respiratory rate was 56/min, and oxygen saturation was 65% in room air. The lung fields were clear. The abdomen was markedly distended, and bowel sounds were absent. Anal opening was absent, and there was a dimple in the region of the anus but no meconium bulge or track.

Cross table lateral X-ray showed a low anomaly. Abdominal and chest radiographs showed large pneumoperitoneum [Figures 1 and 2]. Serum electrolytes showed hypokalaemia of 2.9 mmol/l, hyponatraemia of 114 mmol/l and elevated creatinine of 209 umol/l. Dehydration, hypokalaemia and hyponatremia were corrected. Oxygen was administered by nasal catheter. Respiratory distress continued to worsen rapidly. To relieve respiratory distress, pneumoperitoneum was vented percutaneously using 24 Fr cannula inserted into the right hypochondrium, below the liver, with significant improvement in respiration. While preparations for surgery were made, re-examination of the perineum showed a pinhole fistula in the anal dimple, through which a size 14 Fr cannula was inserted (after gentle progressive dilatation) into the rectum

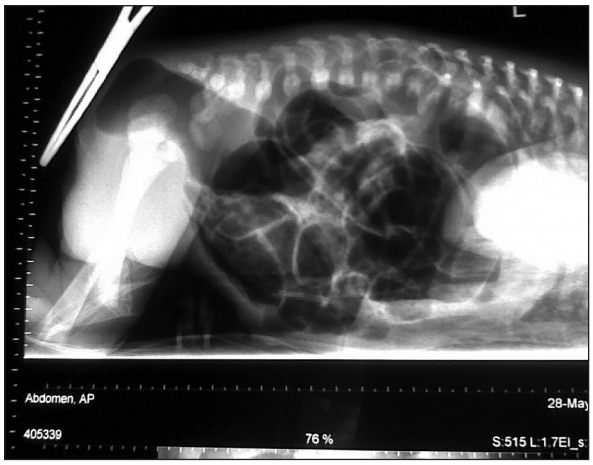

Figure 1: Cross table lateral X-ray showing low anorectal anomaly

Department of Surgery, Division of Paediatric Surgery, National Hospital, Abuja, Nigeria

Address for correspondence:
Dr. TiJesuni Olatunji,
Department of Surgery, Division of Paediatric Surgery,
National Hospital, Abuja, Nigeria.
E-mail: garvouis@yahoo.com

and large amounts of meconium aspirated resulting in further reduction in abdominal distension and improvement in respiration.

At laparotomy, the findings were: Perforated ileum on the antimesenteric border at 2.0 cm from the ileocaecal junction and peritoneal soilage with meconium. There was no evidence of enterocolitis. The ileal perforation was debrided and repaired, and peritoneal cavity thoroughly cleansed with warm normal saline and abdomen closed. An anoplasty was done for the low anorectal anomaly.

Postoperatively, the patient developed superficial surgical site infection in the abdominal wound but this was controlled by local wound care. Anoplasty was dilated daily as from 10th day postoperatively. He did well and was discharged on the 11th postoperative day to continue anal dilatation as outpatient. He remained well at 2nd clinic follow-up visit a month after discharge but was subsequently lost to follow up.

DISCUSSION

Spontaneous intestinal perforation is a serious complication in infants with anorectal malformation and has been reported to occur in 2% of the patients, with late presentation increasing the perforation rate to 9.5%.[3,4] Intrauterine perforation has also been reported.[5] Most reported spontaneous perforations are in the colon,[2,3,6,7] and perforation in the ileum has not been reported. Our patient had ileal perforation. Two theories put forward to explain colorectal perforations in these patients,[4] include.

Vascular aetiology

Downstream obstruction leads to increased intraluminal pressure that impairs the transmural perfusion, resulting in focal ischaemia. A competent ileocaecal valve further contributes to very high intraluminal pressure and perforation.

Congenital aetiology

Congenital muscular deficiency in the region of the tail gut, making the rectum susceptible to perforation in the presence of distal obstruction.

Ileal perforation in our patient was probably caused by increased intraluminal pressure in the presence of an incompetent ileocaecal valve, more so that the wall of the ileum is relatively thin and unlikely to withstand high intraluminal pressures as the colon.

Clinical evidence of intestinal perforation is reported to be absent in 88% of these patients.[4] Presence of anterior abdominal wall oedema,[8] should raise the suspicion of perforation. Nonetheless, a high index of suspicion is important to make a clinical diagnosis, particularly in patients presenting after 48 h of age. Pneuomoperitoneum be present may on plain upright abdominal radiograph, as in our patient, [Figure 2] and such radiographs should be requested in infants with anorectal malformation presenting after 48 h. It may be possible to recognise pneumoperitoneum on cross table lateral X-ray.[4] However, the absence of pneumoperitoneum does not exclude intestinal perforation.

Our patient required percutaneous venting of the large pneumoperitoneum to improve respiration and oxygenation before surgery. In one report of a 1-day-old infant,[6] peritoneal drain was inserted under local anaesthetic due to poor general condition of the infant, to release meconium and air before laparotomy after 24 h. Abdominal distension of often marked in these patients, compromising respiration and any effort to achieve decompression is helpful. Ileal perforation was repaired in our patient. Definitive options for such ileal perforation could include simple repair, resection and anastomosis or exteriorisation of the perforation as a stoma, depending on the quality of intestine adjacent to the perforation. A low malformation can be treated at the same operation as in our patient. A colostomy would be required in high malformations, and the malformation can be treated at a later date.

Mortality for neonatal intestinal perforation generally can be high,[8] and a high mortality from perforation has also been recorded in infants with anorectal malformation.[4] Prompt diagnosis and treatment

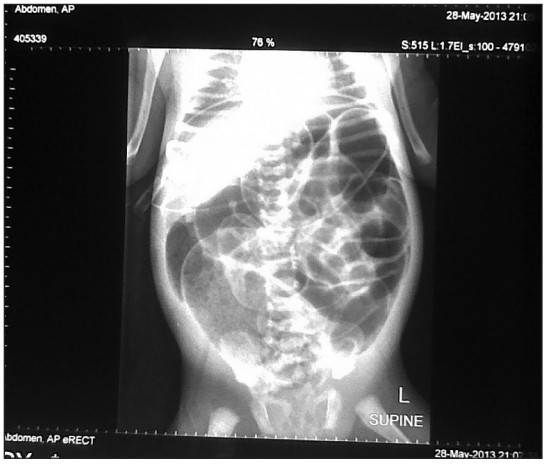

Figure 2: Erect chest/abdominal radiograph and cross table lateral X-ray showing a low anorectal anomaly and pneumoperitoneum

should minimise morbidity and mortality. It is hoped that earlier presentation would prevent spontaneous intestinal perforation in infants with anorectal malformation.

REFERENCES

1. Adejuyigbe O, Abubakar AM, Sowande OA, Olayinka OS, Uba AF. Experience with anorectal malformations in Ile-Ife, Nigeria. Pediatr Surg Int 2004;20:855-8.
2. Lukong CS, Ameh EA, Mshelbwala PM, Jabo BA, Gomna A, Akiniyi OT, *et al*. Management of anorectal malformation: Changing trend over two decades in Zaria, Nigeria. Afr J Paediatr Surg 2011;8:19-22.
3. Tong WD, Ludwig KA. Neonatal colon perforation due to anorectal malformations: Can it be avoided? World J Gastroenterol 2013;19:3915-7.
4. Raveenthiran V. Spontaneous perforation of the colon and rectum complicating anorectal malformation in neonates. Pediatr Surg Int 2012;47:720-6.
5. Tongsong T, Chanprapaph P. Prenatal diagnosis of isolated anorectal atresia with colonic perforation. J Obstet Gynaecol Res 2001;27:241-4.
6. Sandlas G, Kothari P, Sarda D, Karkera P. Spontaneous bowel perforation in a neonate with anorectal malformation. Saudi J Gastroenterol 2011;17:72-3.
7. Chan KW, Lee KH, Tsui SY, Wong YS, Pang KY, Mou JW, *et al*. Bowel perforation in newborn with anorectal malformation and no fistula at presentation. J Pediatr Surg 2014;49:390-4.
8. Chirdan LB, Ameh EA. Neonatal intestinal perforation in a developing country. Ann Trop Paediatr 2001;21:26-8.

Cite this article as: Olatunji T, Igoche M, Anyanwu P, Ameh EA. Spontaneous ileal perforation complicating low anorectal malformation. Afr J Paediatr Surg 2015;12:152-4.

Source of Support: Nil. **Conflict of Interest:** None declared.