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Sigmoid Colon Perforation in Neonate Having Low Anorectal Malformation with Anocutaneous Fistula- A Rare Presentation

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Summary

Occurrence of bowel perforation in newborn with low Anorectal Malformation (ARM) is very rare, as thorough clinical examination of perineum in newborn is usually sufficient to make diagnosis of anorectal malformations. However, in 21-32% cases of ARM, there can be delay in diagnosis resulting in inadequate treatment. This diagnostic and therapeutic delay may lead to bowel perforation. We report a case of 2.6 Kg male neonate with low ARM who had presented to us with pneumoperitoneum. On exploratory laparotomy, a sigmoid colon perforation was found which was repaired with proximal diverging colostomy. Anoplasty was also done at the same time.

Background

As we know, sigmoid colon perforation in low anorectal malformation is a very rare ocurrence and with anocutaneous fistula it's even rarer. Therefore reporting of this case is very important. Timely diagnosis, early referral to tertiary care centre, prompt and precise intervention are the key to successful management.

Case Presentation

A 2.6 kg male baby presented to our department at day 6 of life with complaints of progressive abdominal distension, respiratory difficulty, vomiting, inability to pass meconium and absent anal opening since birth. The baby was delivered by full-term vaginal delivery at home under observation of local dai. Baby cried soon after birth and passed urine within 24 hours. However, he had not passed meconium till day 5 of life resulting in progressive abdominal distension. He was taken to a local physician who had noticed an absent anal opening and referred the patient to our department. There was no history of meconuria. On physical examination, baby was dehydrated, with heart rate of 148/min and respiratory rate of 34/min. Abdomen was shiny, tensed and grossly distended. Perineal examination revealed absent anal opening and a very narrow pinpoint fistula with meconium staining at the anterior margin of anal dimple (Figure-1). X-ray abdomen was suggestive of gas under bilateral dome of diaphragm indicating pneumoperitoneum (Figure-2). Other blood investigations were within normal limits. Baby was resuscitated with intravenous fluids and antibiotics. After adequate resuscitation, exploratory laparotomy was performed using left transverse sub umbilical incision. Gush of air came as soon as the peritoneum was incised and whole bowel was found stained with meconium and fibrinous flakes. A 5x5 mm perforation was identified in low sigmoid colon at antimesenteric border (Figure-3). Meconium along with air bubbles was seen coming through it after gentle squeeze of descending colon. Rest of the bowel was healthy with no evidence of concurrent presence of necrotizing enterocolitis. After thorough peritoneal lavage,

perforation was repaired along with proximal diverging high sigmoid loop colostomy. Abdominal wound was closed in layers. Cutback Anoplasty was also done. Postoperative period was uneventful. Patient was discharged on 5th postoperative day with advice for regular neoanus dilatation in follow-up.

Investigations

Complete blood count, serum electrolytes, kidney and liver function tests, viral markers (HIV,HbsAg, HCV), PT, INR, Xray abdomen AP erect, 2D echo, USG whole abdomen.

Differential Diagnosis

Not relevant

Treatment

Exploratory laparotomy with primary repair of sigmoid colon perforation with proximal diverging colostomy with Anoplasty.

Outcome and Follow-Up

Postoperative period was uneventful. Patient was discharged on 5th post-operative day with stoma functioning well. Patient is now on serial anal dilatation and planned for stoma closure.

Discussion

Anorectal Malformations are common congenital anomalies seen in neonates, that are usually diagnosed soon after birth with careful clinical examination of perineum and is treated appropriately with reasonably good prognosis. However, lack of awareness and inadequate clinical examination of perineum can result in delay in diagnosis and prompt treatment, with overall incidence of delayed diagnosis reported to be as high as 21-32%.[1] Delay in diagnosis of ARM is usually defined as diagnosis made after the first 48 hours or after 3 months in case of low ARM in female.[1] This delay in diagnosis is because of failure to receive comprehensive neonatal examination within 48 hours with further delay in treatment by social factors such as poverty, illiteracy, poor transport facilities, and scarcity of specialists especially in developing countries like India.[12] Delay in diagnosis and treatment can lead to high chances of bowel perforation, sepsis, aspiration, respiratory embarrassment, electrolyte imbalance and even death.[1,2] Bowel perforation is a serious and uncommon complication of delayed diagnosis of ARM in neonate that, if not diagnosed and treated promptly, can be life threatening with neonatal mortality ranging 3% to 23%.[4,11]

Exact occurrence of bowel perforation in ARM is unknown. In his study, Turowski et al had reported overall 2% incidence of bowel perforation in ARM that increased to 9.5% in ARMs with delayed presentation,[1] whereas Chalapathi et al reported 1.6 % incidence of perforations in 125 ARM cases.[5] In his study, Raveenthiran V found 5.9% incidence of colonic perforation[2] whereas a Japanese study reported 2 perforations (5.6%) in 36 cases of ARM.[6]

Occurrence of BP in ARM has been usually attributed to delay in diagnosis. However, not all BP are due to delayed diagnosis or treatment of ARM, because there are few reports of BP occurring during intrauterine life.[7]

The mechanism of BP in neonates with ARM could be due to combination of factors. Lower gastro-intestinal tract obstruction is the main cause, which leads to increased intra-abdominal pressure, which decreases trans-luminal perfusion. Prolongation of this insult may lead to ischemic gangrene of the least vascularized section like antimesenteric border of sigmoid colon. This mechanism explains occurrence of BP in high ARM without or with narrow fistula and low ARM without fistula. Occurrence of perforation in low ARM with fistula is rare because there is no downstream obstruction; however, if there occur occlusion of tiny fistula by inspissated meconium, there can be chances of BP if not diagnosed timely; as we

had found in our case who had very narrow pinpoint perineal fistula occluded with thick meconium. As ARM is a developmental field defect, the tail end of the gut can be expected to have deficiency of musculature as evident from primary rectal ectasia and congenital pouch colon that explains occurrence of BP in these cases.[8]

One should suspect BP in neonate with ARM presenting with features of sepsis and peritonitis such as shiny, tense distended abdomen with parietal wall edema and erythema.[9,10] Early diagnosis and prompt intervention should be taken. A further confirmation of pneumoperitoneum can be done with abdominal X-ray. Management should be done in form of resuscitation and early surgical intervention. Exteriorization of the perforation as a stoma or its primary closure with a proximal diverting stoma, if perforation is located down in colon, is usually preferred. Primary closure of the perforation may be attempted in selective cases.[10] In low ARM, Anoplasty can be done at same time as diverging colostomy has already been done, as in our case.

Learning Points/Take Home Massages

- Low anorectal malformations can be life thretening if not treated early.
- Must be referred to expert and higher centre for early diagnosis and management.
- High sigmoid diversing loop colostomy with primary repair of sigmoid colon perforation can be done in these patients.
- Anoplasty can be done in same sitting as there is covering colostomy is performed.

Regular anal dilatation must be advised before colostomy closure.

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Figure Presentation

Figure-1

Showing absent anal opening with ano-cutaneous fistula at perineo-scrotal junction. Thick meconium seen at fistula site.

Figure-2

Showing large gas under both domes of diaphragm suggestive of Pneumoperitonium.

Figure-3

Showing 5x5mm perforation in sigmoid colon.