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Six-year retrospective analysis of colonic perforation in neonates and infants: Single centre experience

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ABSTRACT

Background: Developing countries at tertiary referral centre. This study analysed the aetiology of colonic perforation (CP) in neonates and infants. **Materials and Methods:** Retrospective analyses of 60 CP cases (presented from May 2005 to May 2011) were done. **Results:** The mean age at presentation was 8.33 ± 0.11 days (range, 2-110 days). The aetiology were Hirschsprung's disease (HD), necrotising enterocolitis (NEC) and idiopathic perforation in 78.33% (47/60), 6.67% (4/60) and 15% (9/60), respectively. There were 210 patients with histopathologically proven HD; 22.38% (47/210) cases of HD had CP. Most common site of perforation was mid-transverse colon (74%, 35/47) in HD patients. All HD-associated mid-transverse colonic, caecal, appendicular and ascending colon perforations (except one caecal perforation) had aganglionic recto-sigmoid region and ganglionic perforation site. Features of enterocolitis were not found in any HD patients. Two patients (3.33%) died due to sepsis. **Conclusions:** There was a high rate of primary HD-associated colonic perforation in this study. Colonic perforation may the initial presenting condition in HD disease. We advocate colonic biopsy to rule out HD in any neonate presenting with primary colonic perforation.

Key words: Colonic perforation, Hirschsprung's disease, necrotising enterocolitis

INTRODUCTION

The most common site of perforation in neonates is the small intestine.[1] Although the perforation of the colon in neonates is rare, it is associated with more than 50% mortality in high-risk patients.[1] Colonic perforation (CP) may be caused by various conditions such as necrotising enterocolitis (NEC), Hirschsprung's disease (HD), mechanical obstructions (anorectal malformation [ARM], colonic atresia), meconium plug syndrome, small left colon syndrome, idiopathic perforations, isolated intestinal perforation, stercoral perforations and rarely cystic fibrosis.[1,2] We evaluated the aetiology of CP in neonates and infants, and present our experience in this report.

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MATERIALS AND METHODS

All neonates and infants, who had CP diagnosed on laparotomy, were included in the study and data analysed retrospectively. The study was performed from May 2005 to May 2011. Preterm babies and cases, where there was a demonstrable cause of obstructive perforation like volvulus, internal or external hernias, intussusceptions, congenital bands or intestinal atresia, ARM, huge fecolith (stercoral perforation), etc., were excluded from the study.

The babies were adequately resuscitated by nasogastric tube placement for gastric decompression and fluid replacement. Broad-spectrum intravenous antibiotics were administered for sepsis prophylaxis and control. In case of gross abdominal distension, under local anaesthesia, intraperitoneal drain placement was done as an emergency procedure. The investigations included blood counts; serum electrolytes, renal function test, blood culture and C-reactive protein (in suspected cases of NEC) were done.

After stabilisation of the patients, laparotomy was performed. Because of limited facilities of frozen section biopsy, we had taken the biopsy of colon wall adjacent to the perforation (in all cases), and distal to the transition zone (TZ), if it was appreciated [Figure 1] on empirical basis. If we were not able to appreciate the TZ, the biopsy was taken from the recto-sigmoid region. In case of perforation located on the ascending colon or

beyond that, it was exteriorised as loop colostomy, while in caecal perforation, primary closure of the perforation and diversion ileostomy, along with biopsy from multiple colonic sites and perforation site was taken.

The clinical data included age, sex, birth weight, gestation age, and associated anomalies, location of CP, histopathology findings, morbidity and mortality were analysed.

RESULTS

During the study period of six years, A total of 350 neonates and infants underwent exploratory laparotomy for pneumoperitoneum. Out of these, 60 patients had CP. The male to female ratio was 53 : 7 [Table 1]. The mean age at presentation was 8.33 ± 0.11 days. The mean gestation age was 36.39 ± 0.19 weeks. The mean birth weight of neonates was 3.1 ± 0.17 kg. Ten neonates had associated anomalies (congenital heart diseases in six, Down's syndrome in four). The history of non-steroidal anti-inflammatory drugs intake was present in only three patients.

The erect abdominal X-ray demonstrated pneumoperitoneum in (57/60) 95% patients. Histopathological examination of biopsied specimen in 78% (47/60) patients. NEC in full term was found in 6.6% (4/60) and localised idiopathic perforation in 15% (9/60) patients.

In 70% (33/47) of histopathologically proven Hirschsprung's patients, classical history of HD were present, while in 30% (14/47), bowel perforation were the primary presentation (no constipation or no history of delayed passage of meconium). On the other hand, irrespective of clinical presentation, we had treated 210 patients of HD at our institute over the same duration. Out of these 210 patients of histopathologically proven HD cases, 22.38% (47/210) patients had CP. In 33 patients of CP, parents gave previous history of constipation, but 14 patients were absolutely normal. In other words, in our study group 6.6 % (14/210) of patients of HD, the presentation was CP without any other complaints (no constipation or no history of delayed passage of meconium).



Figure 1: Sigmoid colonic perforation

In 74% (35/47) of HD patients, the perforation was in mid-transverse colon, followed by sigmoid colon (15%, 7/47) and splenic flexor (11%, 5/47). In only 26% (12/47) of patients, per-operative spastic zone was identified (recto-sigmoid in 8 and sigmoid colon in 4). In the remaining 25 patients, biopsy from recto-sigmoid region was taken on empirical basis, which was later found to be aganglionic in 80% (20/25). All mid-transverse CP had aganglionic recto-sigmoid region and adequate ganglion at perforation site. In caecal, appendicular and ascending CP, multiple biopsies from colon were taken. All these HD patients underwent Duhamel procedure as definitive procedure later on.

Focal CP due to NEC of full term occurred in four neonates (one at splenic flexor and three at sigmoid colon). The histology showed adequate ganglion cells, besides sign of inflammation anddiffuse micro-thrombi.

Table 1: Patient's characteristics

Characteristics	Results
M: F ratio	53:7
Mean age at diagnosis (days)	8.33 ± 0.11 (ranged, 2 to 110 days)
Mean gestation age (weeks)	36.39 ± 0.19 (35 to 40 weeks)
Mean weight (kg)	4.52 ± 0.21 (3.0 to 8.4 kg)
Associated anomalies	Congenital heart disease = 4 Down syndrome = 6
Maternal/foetal complications	None
Usage of medications	3
Sites of perforation	Mid-transverse colon = 35 (58.33%) Splenic flexor =12 (2.0%) Sigmoid = 9 (15.0%) Caecum = 2 (3.33%) Ascending colon =1 (1.67%) Appendix=1 (1.67%)
Aetiology	Hirschsprung's = 47 (78.33%) Localised/idiopathic perforation = 9 (15%) Full-term NEC = 4 (6.67%) Small left colon = 0 Meconieum ileus = 0
Complications	Sepsis =10 (16.6%) Wound dehiscence = 5 (8.83%) Superficial wound infection = 4 (6.66%) Colostomy prolapsed = 2 (33.3%)
Outcome	2 expired (3.33%)

These patients presented within first week of their life. In these patients, colostomy closure were done 2 months later. These cases had no postoperative complication.

In 9 patients, the cause was localised isolated/idiopathic perforation as there was no evidence of NEC or any identifiable mechanical cause. The histology in these patients showed non-specific changes (oedema, inflammation, haemorrhage and necrosis with occasional micro-thrombi), and no microorganism on blood culture. In these patients also, colostomy closure were done 2 months later without any postoperative complications.

Postoperatively, superficial wound dehiscence developed in 5 patients (8.83%). All these patients were malnourished infants, who had classical history of HD. The dehiscence was managed conservatively. Ten patients had features of sepsis, out of which 2 died because of severe sepsis on the 2nd and 5th postoperative days. Two patients had distal stoma colostomy prolapse that needed revision double barrel colostomy.

DISCUSSION

In preterm babies, pneumoperitoneum is mostly due to NEC; however, in full-term babies and early infancy, the aetiology is quite different.[3-6] In our study, we noticed that in the absence of obvious mechanical cause, CP occurred most commonly due to HD. The bowel perforation usually occurs in 3.2% to 4.4% cases of HD, mostly in the neonatal period.[7-9] However, we noticed that it was also quite common in early infancy.

We had higher rate of CP (6.6%) in HD patients probably due to delayed medical advice sought by the parents. These 6.6% patients presented to our department only for the sufferings from perforation peritonitis. These babies had not had any history of delayed passage of meconium or constipation. This is unique, because to our best of knowledge there is no study in literature which emphasised on the HD initial manifestation as CP.

Some authors advocate the use of water-soluble contrast studies in perforated colon to identify the transitional zone exactly, assuming it is difficult to identify the TZ in the unused colon after colostomy.[7] We never use these studies in any of our patients because it can be more hazardous in sick patients with perforated bowel; instead, we recommend exploration and direct visualisation of TZ marked by non-absorbable sutures with documentation on discharge more beneficial and safe.[7] We did not go for the transanal frozen section biopsy at the first laparotomy for identification of TZ, because in neonates, there is a physiological aganglionic zone, which can mature past the neonatal period.[10]

Contrary to the common sites of CP (proximal colon and appendix) mentioned in literature, we noticed that transverse colon was the most common site of perforation.[8,9,11] There were no features of enterocolitis in any of the HD-associated perforations, which supports the fact that perforation in HD occurs due to increased intraluminal pressure rather than inflammatory process.[4,12] Most of the caecal or proximal CPs occur in TCA.[5,6,12] However, we found only one patient (cecal perforation) of TCA out of four proximal CP. In rest proximal CP (1 appendix, 1 ascending colon, and 1 caecum) patients, the TZ was at recto-sigmoid region. Thus, our study also showed that it is not unusual to have proximal CP in short segment HD. Forceful diagnostic or therapeutic enema given by untrained practitioners (occurred in our 2 cases) for delayed or infrequent passage of stools can cause perforation because of upstream occlusion by non-functional spastic segment.[1] In both these cases, spastic zone were at the sigmoid colon and perforation is distal to TZ in the spastic segment. We found that perforation was proximal to the transitional zone in short or intermediate type HD, and in aganglionic colon in patients of TCA.[7]

NEC in full-term infants accounts for 10% of all cases of NEC, and predominantly involves the colon.[13,14] Although the risk factors in full-term are congenital heart diseases, undiagnosed syndrome, birth asphyxia, intrauterine growth retardation and congenital hypothyroidism, but apparently, normal full-term infants may also develop NEC.[13,14]

Localised or idiopathic perforation was described by Breslau in 1863 as “the perforation for which no other cause or NEC is found.”[15] Usually, it occurs in low or very low birth-weight infants in whom traumatic delivery, congenital defects of the bowel with diverticulum formation, localised vascular insufficiency and intrauterine or post-partum infection have occurred.[15] Localised or idiopathic perforation accounts for one-third of all CP, but we found it in only 9 (15%) of patients. [15] The probable explanation is that our sample population included only full term neonate and infants, while idiopathic perforation occure mostly in low birth-weight babies.

We would suggest biopsy in all perforations in early

part of life to differentiate the NEC of full-term, and idiopathic perforation from HD, as the former two conditions only need stoma closure in later life. Furthermore, both the entities have relatively good prognosis compared to NEC of prematurity.[2,13-15]

To conclude, HD is not only the most common cause of large bowel obstruction in the young infant, but also the most common cause of perforation. Every neonate and infant should be evaluated for HD for CP, whether history and per-operative findings (identifiable TZ) are suggestive of it or not.

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