CASE REPORT

NEONATAL SPONTANEOUS COLONIC PERFORATION: A RARE ENTITY

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Colonic perforation presenting as acute abdomen in the early neonatal period is a rarely reported entity in the literature. Mostly reported cases are secondary to some mechanical obstruction or underlying enteric pathology. Despite advances in neonatal care, it still carries a significantly high mortality. Abdominal X-ray demonstrating free air is sufficient to confirm the diagnosis, mandating early surgical intervention. We present a rare case of spontaneous transverse colonic perforation in a one-day-old neonate presenting with sudden-onset gross abdominal distension and respiratory distress managed by exploration and primary repair.

Keywords: Colonic perforation; Idiopathic; Spontaneous

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INTRODUCTION

Gastrointestinal perforations in the neonatal period are a rare cause of acute presentation. Such perforations can occur at any location in gastrointestinal tract but most commonly reported site is small intestine.[1] Risk factors for enteric perforation in neonates are prematurity, low birth weight and birth asphyxia[1]. Necrotizing enterocolitis is the reported leading cause of neonatal enteric perforations.[2] Other causes include atresia, Hirschsprung disease, anorectal malformation meconium ileus and plug syndrome.[3] If perforation occurs in the absence of any such demonstrable etiology, it is labelled as idiopathic or spontaneous perforation.[4] Spontaneous colonic perforation in a term neonate is a rare entity with very few cases reported in past.[5] Presence of pneumoperitoneum on plain abdomen x-ray is a surgical emergency that mandates emergent exploration following optimization to reduce post operative mortality and morbidity resulting from perforated hollow viscera and peritonitis. Index case presented in early neonatal period with abdominal distension leading to respiratory distress. Plain x-ray abdomen demonstrated gross pneumoperitoneum with classical signs. Exploration revealed mid transverse colonic perforation with normal small and large bowel which was repaired primarily confirming the presence of ganglion cells on frozen section biopsy.

CASE REPORT

Paediatric surgery review was requested in intensive neonatal unit for a one-day old baby boy having abdominal distension and respiratory distress. Child was born at 37 weeks of gestation having birth weight of 2.6 kg to a multigravida by uncomplicated spontaneous vaginal delivery with unremarkable antenatal history. Child cried spontaneously following birth and had normal APGAR score. Neonatal examination did not reveal any abnormality and baby was handed over to mother for breast feeding. Twenty hours following birth, baby was brought back by parents in paediatric emergency stating that baby is not tolerating feeds and also, they had noticed that his abdomen is getting swollen as the time was passing. Child had not passed meconium and had not tolerated breast feeding which he had vomited out after 15 to 20 minutes every time he was fed. Examination revealed lethargic child with pulse of 148 beats/minutes, blood pressure of 68/42mmHg, respiratory rate of 64/minute with normal temperature. Child capillary refill was increased to 3 seconds and skin turgor was reduced though fontanelle were not depressed. Abdominal examination revealed soft distended abdomen without any palpable mass or overlying skin changes. Percussion note was resonant with loss of liver dullness and bowel sounds were not audible. Anal opening was patent and was normally located. Baby was admitted in neonatal intensive unit and all baseline investigations were ordered. Baby was kept nil per oral and appropriate size nasogastric tube was place which showed yellowish aspirate. Abdomen x ray was ordered and paediatric surgery review was sought by initiating emergent call. Meanwhile, baby abdominal distension continued to increase gradually leading to tense distended abdomen and tachypnoea with respiratory rate of 78/minute. Portable abdominal x ray was obtained which shows gross

pneumoperitoneum as Football sign and Rigler sign as shown in Figure -1.

Child was planned for emergency exploration following optimization and after obtaining informed written consent. Exploration was done through supraumbilical transverse incision. Contained meconial peritonitis was found with 3x2 cm perforation in mid transverse colon (Figure-2).

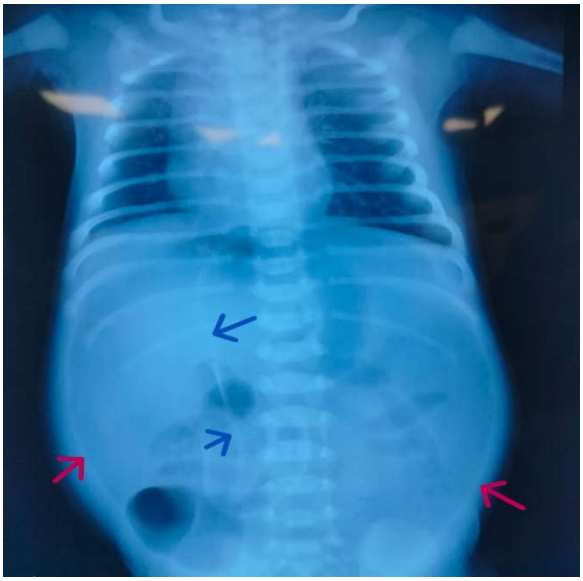

Figure-1: Plain x-ray abdomen showing pneumoperitoneum with Football sign (red arrow) and Rigler sign (blue arrow).

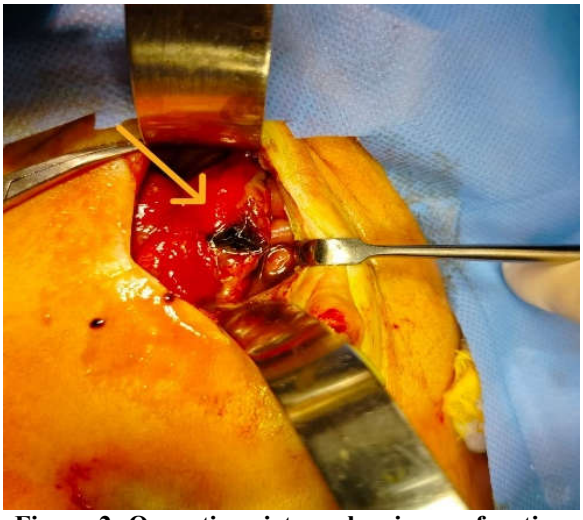

Figure-2: Operative picture showing perforation in mid transverse colon with attached omentum (yellow arrow).

Keeping in mind the possible association of Hirschsprung disease and inability to find definite transition zone in early neonatal period, full thickness frozen section biopsy was sent for ganglion cells. While waiting for frozen section report, entire small gut and large gut was examined for any associated anomaly and through peritoneal lavage was done using luke warm water. As frozen section showed presence of ganglion cells, primary repair with polyglactin 4-0 was done. Postoperative recovery was uneventful. Child was started on feeds on 4th postoperative day which were gradually increased to the required daily amount. Child was discharged on 8th postoperative day. Follow up at 1 and 3 months showed a thriving infant with no gastrointestinal issues.

DISCUSSION

Neonatal spontaneous colonic perforation is an extremely rare cause of emergency exploration with reported mortality of more than 50% if either diagnosis or treatment is delayed. Localized vascular accident during embryogenesis or during perinatal stress causing hypoxia and local ischemia is the most accepted theory. intestinal wall invasion by coliform bacteria causing local endotoxin release and Schwartzman reaction is another possible theory for spontaneous perforation[6]. As spontaneous perforation is supposed to be without any demonstrable mechanical cause therefore, we were unable to find any features suggesting of pathologies like atresia, meconium ileus or plug, small left colon and anorectal malformation both in preoperative and intraoperative findings. As obvious transition zone and colonic dilation in ganglionated segment is absent in early neonatal period as was in our case, full thickness frozen section biopsy from perforation site to rule out Hirschsprung disease by demonstrating presence of ganglion cells is mandatory as was performed in our case. Our index case presented in early neonatal period after 24 hours of birth with progressive abdominal distension and respiratory distress explained by diaphragmatic splintage from massive pneumoperitoneum. Spontaneous colonic perforations typically present in early neonatal period of up to two weeks as compared to perforations associated with necrotizing colitis, a finding reported by Demir N and colleagues.[6] In the presence of massive pneumoperitoneum on x-ray abdomen in a neonate, colonic or gastric perforation should be kept in mind as site of perforation, later being resulting from aggressive bag mask resuscitation for respiratory distress in early neonatal period.[7] No such history was present in our case. Plain x ray abdomen demonstrated classical Football and Rigler signs of massive pneumoperitoneum in our case. These typical x-ray findings obviate the need of further investigations to confirm the diagnosis. Early exploration is the key to prevent mortality from septicaemia, hemodynamic instability

and organ failure. Surgical options include primary repair, exteriorization or resection with primary anastomosis. Factors dictating among these options are site, size and number of perforations, degree of peritoneal contamination and hemodynamic status of patient.[8] Primary repair is the preferred method of dealing in cases of solitary small-size perforation with minimal contamination in healthy bowel and hemodynamic stable patient as was the case in our scenario. One such case was reported in a 21 days old neonate who presented with solitary transverse colon spontaneous perforation which was successfully managed by primary repair.[9] Early detection and intervention are the key to manage such cases as delay in either diagnosis or treatment has inverse relation with survival as shown by Abo-Halawa.[10] Tiwari C reported a case of spontaneous colonic perforation in a term, normal birth weight male presented at 26th day of natal life which was repaired primarily without frozen section biopsy as there was no evidence of Hirschsprung disease in the form of developed transition zone.[11] As transition zone is not evident in early neonatal period, frozen section intraoperative confirmation of ganglion cells remains the most valid tool to exclude Hirschsprung disease before primary repair of perforation as was in our case.[12]

Spontaneous colonic perforation is rare in the neonatal population, and massive pneumoperitoneum should raise suspicion in preoperative workup. Detailed intraoperative bowel examination is a must to exclude other causes before embarking on repair.

Conflict of Interest: None.

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AUTHORS CONTRIBUTION

MMS: Study design, Study conduct, Literature search, Write up. Pervaiz M: Write up, Proof reading.

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