

BRIEF REPORT

SPONTANEOUS PRE-PYLORIC PERFORATION IN CHILDREN

Kaushal Kulkarni, Charu Tiwari, Nilesh Nagdeve, Rajendra Saoji.

Department of Pediatric Surgery, Government Medical College and Hospital, Nagpur, Maharashtra, India.

ABSTRACT

Background: Gastro-intestinal (GI) perforation is less common in pediatric age-group. Spontaneous pre-pyloric/gastric perforation is rarer. We present 7 cases of spontaneous pre-pyloric perforation.

Materials and Methods: All patients under 18 years of age operated for spontaneous pre-pyloric perforation were included in the study and were analyzed with respect to age at presentation, gender, clinical presentation, management, post-operative recovery, and follow-up. All patients were admitted, and detailed history and vitals were recorded. After confirming the diagnosis on erect X-ray abdomen, they were either taken for exploratory laparotomy if stable or initial external peritoneal drainage followed by exploration in unstable patients.

Results: There were total seven patients with spontaneous pre-pyloric perforation. The mean age at presentation was 4.5 years. There were 5 males and 2 females. The mean duration of symptoms was 3 days. There were three neonates-two were pre-term and low birth weight and underwent initial peritoneal drainage followed by exploration after stabilization. The third neonate was full-term, adequate for gestational age and had associated Tetralogy of Fallot. There were two pre-school children, one school-going child, and an adolescent. At exploration, all had a pre-pyloric perforation which was primarily repaired with omental patch. All patients except one neonate were discharged by 7-10 days. One neonate died within a month of discharge from the hospital.

Conclusion: Though rare, spontaneous pre-pyloric perforation in a pediatric age group may carry high morbidity and mortality. Exploration after stabilization remains the treatment of choice.

Introduction

Pneumoperitoneum on an erect X-ray abdomen suggests gastrointestinal (GI) perforation and is a surgical emergency. Although a benign condition, an undiagnosed or inappropriately managed perforation can carry high morbidity and mortality. Pre-pyloric perforation secondary to peptic ulcer disease and infection by *Helicobacter pylori* and chronic use of non-steroidal anti-inflammatory agents (NSAIDs) is common in adults.¹ However, GI perforation is less common in pediatric age-group with relatively few identified causes and vague and variable clinical presentation. Spontaneous pre-pyloric/gastric perforation is even rarer in this age group. Most reported cases of spontaneous gastric perforation are in neonates with only a few reports of such cases in pre-school children and adolescents published in literature.^{2,3,4} It is a serious condition which can be potentially fatal.⁵ The etiology still remains unclear, although several theories have been proposed.⁴ We present 7 cases of spontaneous

pre-pyloric perforation managed at our institute: three neonates, two pre-school children, one school going child and an adolescent.

Methods & Materials

All patients under 18 years of age operated for spontaneous pre-pyloric perforation over a period of one year from January 2018 to December 2018 were retrospectively studied and included in the study. Institutional ethical committee approval was taken. During this period, a total of 71 patients with pre-pyloric perforation were operated; out of these, 7 patients were less than 18 years of age. Patients presenting to the emergency surgical department with abdominal symptoms and signs suggestive of perforation were admitted and their vitals were recorded; thorough general and local examination were done, and complete history taken. The stomach was decompressed by Ryles' tube insertion and intravenous access was secured for all of them and intravenous fluids, antibiotics (third-generation cephalosporin, aminoglycoside, and metronidazole) and H2 blockers were started as per the weight of the patients. Routine blood investigations and erect x-ray abdomen were done for all. The patients were taken for the operative procedures once stable for surgery. External peritoneal drainage was preferred for unstable patients. Such patients were later taken for exploration after initial stabilization. The patients with

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CONTACT Charu Tiwari

Email: drcharusharma18@gmail.com

Address for Correspondence: Dr Charu Tiwari, Department of Pediatric Surgery, Government Medical College and Hospital, Nagpur, Maharashtra, India.

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pre-pyloric perforation with no antecedent cause were included in the study. Those children with a perforation at other part of GIT and/or having any positive history for pre-pyloric perforation were excluded from the study. The patients were analyzed with respect to age at presentation, gender, clinical presentation, management, post-operative recovery, and follow-up.

Results

Seven children with spontaneous pre-pyloric perforation included in this study out of a total of 71 patients (adults + children) with pre-pyloric perforations who presented to us. The mean age at presentation was 4.5 years; with the range being 2 days to 18 years. There were 5 males and 2 females. All patients presented to the emergency surgical department with a history of abdominal pain, distention, bilious vomiting and constipation. Two of them were febrile. The mean duration of symptoms was 3 days; the range is 1 day to 4 days. There were three neonates out of which two were preterm and low birth weight (LBW). They presented with features of septicemia with hypothermia, delayed capillary refill time (CRT), abdominal distention along with erythema of anterior abdominal wall and thrombocytopenia. One of these had a history of home delivery and the other had a history of premature rupture of membranes

(PROM) leading to prolonged labor. After confirming pneumoperitoneum on erect X-ray, both these neonates underwent initial peritoneal drainage followed by exploration after few days of stabilization and control of septicemia. The third neonate was full-term, adequate for gestational age and had associated Tetralogy of Fallot. After confirming the diagnosis by erect X-ray abdomen, all patients were taken for exploration. All had a pre-pyloric perforation which was primarily repaired with omental patch. The biopsies taken from the site of perforation suggested only inflammation. The neonatal perforation biopsies did not suggest neonatal enterocolitis (NEC) and Hirschsprung disease. The post-operative period was uneventful in 6 patients and they were discharged by 7-10 days. One patient had a stormy post-operative course of events and had a hospital stay of 3 weeks. The pre-term low birth weight septicemic neonate with a history of home delivery died within a month of discharge from the hospital. Gastrin levels were done for 3 patients and were normal for all of them. Widal test was also negative in 4 children; both these tests were not done in neonates. There was no history in any of these patients which could suggest any cause for perforation. There was no history of the use of NSAIDs in any of them. All 6 patients are doing well on follow-up (table 1).

Table 1. Details of each patient

	Patient 1	Patient 2	Patient 3	Patient 4	Patient 5	Patient 6	Patient 7
Age	18 years	4 years	8 years	2 years	3 years	6 years	6 years
Weight	50 kgs	14 kgs	2.2 kgs	1.5 kgs	12 kgs	18 kgs	1.6 kgs
Preterm/Full term	Full-term	Full-term	Full-term	Pre-term	Full-term	Full-term	Pre-term
Birth weight	2.5 kgs	2.3 kgs	2.2 kgs	1.5 kgs	2.8 kgs	3 kgs	1.6 kgs
Type of delivery	Vaginal Delivery	Vaginal Delivery	Vaginal Delivery	Vaginal Delivery (Home)	Vaginal Delivery	Vaginal Delivery	Vaginal Delivery
Gender	Male	Male	Male	Male	Male	Female	Female
Abdominal Pain	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Distension of abdomen	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Vomiting	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Constipation	Yes	Yes	-	-	Yes	Yes	Yes
Duration of symptoms (days)	3	2	4	2	1	5	4
Associated anomaly	Nil	Nil	Tetralogy of Fallot	Nil	Nil	Nil	Nil
Hemoglobin (gm%)	10.2	9.8	9.5	9	10.8	11	8.9
WBC count (cells/cumm)	16,100	15,400	16,500	19,800	14,600	15,900	18,900
Platelets (cells/cumm)	1,80,000	2,20,000	90,000	1,00,000	1,60,000	2,40,000	1,20,000
Antibiotic duration (days)	5	5	12	7	18	5	14
Days to recovery	7	7	14	9	21	7	17
Outcome	Recovered	Recovered	Recovered	Died at home after one month	Recovered	Recovered	Recovered
Gastrin levels	Normal	Normal	-	-	Normal	Normal	-
Widal test	-	Negative	-	-	Negative	Negative	-

Discussion

Spontaneous pre-pyloric perforation is a rare entity with most of the cases reported in neonates.⁴ Gastric perforation in a neonate was first described by Siebold in 1825.^{2,6} There are reports stating spontaneous gastric perforations in otherwise healthy neonates, with clinical presentation between the first 2 and 7 days of life.^{2,7} The etiology is unclear; several possibilities being congenital gastric wall defects, mechanical disruption, stress ulceration secondary to neurogenic disorders, and ischemia of the gastric wall secondary to vascular shunting, diving reflex and intestinal obstruction.⁴ Prematurity, gastric volvulus, foreign body ingestion, trauma or iatrogenic injuries and nasal ventilation have also been cited as proposed causes.^{4,5,8,9,10,11,12} Cases of spontaneous intestinal perforation have been reported in preterm neonates with very low birth weight (VLBW) and extremely low birth weight (ELBW).¹³ Two of the three neonates described above were premature and low-birth-weight. The third neonate was a full-term baby with an associated cardiac anomaly. Reports of gastric perforation in 3 to 5-year-olds are also uncommon where it is usually due to a casual event such as peptic ulceration, trauma, aerophagia, or drug intake including corticosteroids.⁴ It has also been reported in conjunction with Rett syndrome, Burkitt lymphoma, and heterotaxy.^{4,14} Elevated intraluminal pressure and ischemia of the bowel wall are also cited as possible causes.⁴ However, no such causes could be identified in any of our patients. Iatrogenic causes of gastric perforation include blunt trauma due to nasogastric tube placement or over-distension of the stomach due to positive pressure ventilation. According to Drumm B et al it was noted that the most common etiologies inpatient less than 10 years of age were attributed to medications like NSAIDs, lymphoma, gastroenteritis, meningitis and malaria (secondary causes).¹⁵ While for the patients above 10 years, it was found that recurrences were common. However, there were no such causes of perforation seen in our cases. According to published cases in the literature, peptic ulcerations are more common among male adolescents. A study done by Hua et al reviewed 52 cases of perforated duodenal ulcers in the pediatric population. More than 80% were male and 90% were adolescents between the ages of 14 to 18.¹⁶ Though uncommon, peptic perforation can be seen in young patients in association with H. pylori infection, secondary to medications like NSAIDs, corticosteroids, rarely due to Zollinger Ellison syndrome, related to stress, post burns or head trauma.^{17,18,19,20} No such history or causes could be identified in our patients and gastrin levels are done in three of our patients were also normal.

Spontaneous gastric perforation is occasionally seen in adults in conjunction with malignancy or peptic ulcer.^{4,21,22} There is female preponderance and the majority of the ruptures occur around the lesser curvature.^{4,21,22} Spontaneous gastric rupture has also been reported in pregnancy caused by an abrupt rise in abdominal pressure in an already distended stomach; the proposed mechanism being impaired perfusion and/or obstruction in venous drainage due to high intraluminal pressure led to necrosis of the gastric wall.^{4,23}

Radiological imaging suggests free gas under the diaphragm on an erect X-Ray abdomen. Surgical management remains the cornerstone in perforation.^{18,24,25} The traditional open surgical method is usually undertaken. Simple closure with or without omentum is one of the most commonly used technique.²⁴ It is the easiest, quickest and safest operation, and can be applied to all situations by every surgeon. Time should not be wasted to operate after clinical diagnosis and resuscitation as it can lead to adverse results. In contrast to adults partial gastrectomy is seldom advised in the pediatric age group. More recently, laparoscopic gastric or duodenal perforation closure is being advocated and gaining popularity due to less morbidity, lesser pain and reduced hospital stay.^{8,25} However laparoscopic suturing techniques take much longer time, require surgeon expertise and it can be tough on surrounding inflamed tissues as in perforation due to its friability.

Conclusion

Gastric perforation is a surgical emergency and should be included in the differential diagnosis of neonates and children presenting with abdominal distention, vomiting, poor activity, and respiratory distress (25). Pneumoperitoneum on plain x-ray confirms the diagnosis. Treatment is the surgical repair of the gastric defect with or without an omental patch after adequate stabilization. Attempts should be made to investigate the underlying cause; in the absence of which the perforation is deemed spontaneous.

Compliance with Ethical Standards

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Conflict of Interest: None

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