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ORIGINAL ARTICLE

Spontaneous Gastric Perforation in Neonates: A Tertiary Pediatric Surgical Center Experience

Abdelghany EM Abdelgawad,[1,2] Ahmed A Darwish,[1,3]* Emely Hughes,[1] Eleri Cusick[1]

1.Department of Pediatric Surgery, Bristol Royal Hospital for Children, Bristol, UK

2.Deparetment of Pediatric Surgery, Tanta University Hospital, Tanta, Egypt

3.Departement of Pediatric Surgery, Ain Shams University, Cairo, Egypt

ABSTRACT

Objective: To report our experience with spontaneous neonatal gastric perforation (SNGP) in a tertiary hospital over the last 9 years.

Methods: Retrospective review of neonates with SNGP treated in Bristol Royal Hospital for Children between January 2010 and December 2018 was performed. Data retrieved included demography, perinatal clinical details, operative data, and outcome. Cases with esophageal atresia, necrotizing enterocolitis or distal obstruction were excluded.

Results: Of 13 cases, 4 were males and 9 females. Two neonates were full term; while 11 were preterm with median gestational age of 28 weeks. Abdominal distension and metabolic acidosis were mostly found in these patients. Patent ductus arteriosus was encountered in 53.8% of the patients. The most common site of perforation was the greater curvature (38.4%) followed by posterior wall (30.8%) of the stomach. Primary repair was performed in all cases. One neonate developed recurrent perforation in the early postoperative period. Five preterm (38.5%) neonates succumbed in our series.

Conclusion: SNGP is a rare condition increasingly diagnosed in preterm neonates. Greater curvature and posterior wall of stomach are commonly involved. Despite recent advances in intensive care, the prognosis is still poor in preterm babies with multiple associated problems.

Key words: Neonate; Gastric Perforation; Spontaneous; Preterm

INTRODUCTION

Despite an increase in prevalence, neonatal gastric perforation (NGP) remains a relatively uncommon life-threatening entity.[1] The reported incidence of NGP is 1:5,000 live births and represents only 7% of all neonatal gastrointestinal perforations.[2] The first case of NGP was reported in 1825 by Siebold.[1] Proposed etiologies include spontaneous or secondary to either necrotizing enterocolitis (NEC) or distal obstruction but the exact mechanism remains unclear.[1] Recent reports show improvement in survival mainly attributed to the improvement in neonatal intensive care.[3] We herein report our experience with 13 cases of spontaneous neonatal gastric perforation (SNGP) over a period of 9 years.

MATERIALS AND METHODS

A retrospective review of neonates with SNGP treated in Bristol Royal Hospital for Children, Bristol, UK (tertiary care hospital) between January 2010 and December 2018 was carried out. Both electronic and written medical records were reviewed and the data collected included gestational age, birth weight, antenatal course and scans, associated anomalies, clinical presentation, age at perforation, operative findings, treatment and outcome. Data were reported as percentage or median (range). NGP

Correspondence*: Ahmed A Darwish, Department of Pediatric Surgery, Bristol Royal Hospital for Children, Bristol, UK
E mail aadarwish37@yahoo.co.uk

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secondary to esophageal atresia, NEC or distal obstruction were excluded from this study.

RESULTS

13 cases treated for SNGP in our center, 4 males and 9 females. Two neonates were full term (birth weights of 3100 and 3400gm), while 11 were preterm with median gestational age (GA) of 28weeks (range 24-34weeks); and median birth weight of 1185gm (range 566-2530gm). Three neonates were born as twins (one had twin-to-twin transfusion); whereas one as triplet. One neonate was born as a result of in-vitro fertilization. Cardiac anomalies were detected on antenatal scans in 2 patients, and one of these also had hydrops. Maternal pre-eclampsia was noted in two cases.

Laboratory data showed low platelet count in 10 patients, low WBCs in 3 patients (1.2, 4.8, and 4.5 x 103), abnormal clotting profile in 4 patients (one patient had DIC), and low albumin in 4 patients. The retrieved Data showed C-Reactive protein (CRP) <1 in 5 patients (Normal value: <6) while seven had CRP values ranged between 6 and 26 (median 13). Blood cultures showed no growth in 7 patients (5 received antibiotics), two patients had a positive culture with coagulase negative staphylococcus epidermidis and gram-negative bacilli. Another patient had a candida growth on peritoneal dialysis (PD) catheter related peritoneal samples. The latter presented with persistent lactic acidosis and required early ligation of a significant patent ductus arteriosus (PDA). Three cases had no documented culture results.

Abdominal distension was the main presentation with free air detected on x-rays in all cases. Three patients had lactic acidosis that was difficult to manage (one died later). Acidosis was noted before gastric perforation in a patient with large PDA and renal impairment. Of the associated anomalies, PDA was encountered in 7 patients (53.8%); only two cases had chromosomal anomalies (Table 1). Early intubation with ventilation was required in 10 patients mainly due to respiratory distress syndrome (9/10) and abdominal distension (1/10).

NGP secondary to steroid use and traumatic injury by nasogastric tube were suspected in two of our cases. In the later, this was based on history of recent nasogastric tube insertion with X ray findings of free air and abnormal position of the tube (Figure 1).

All patients underwent surgery. The age at perforation ranged between 2-34 days after birth; with 11 patients (84.6%) developing perforation between the 2-9 days of life. The most common sites of perforation were the greater curvature (n=5, 38.4%), posterior wall (n=4, 30.8%), anterior wall (n=2, 15.4%) and fundus (n=2, 15.4%). Primary repair with absorbable sutures in two layers was performed in all cases and a gastrostomy was created in one patient. Only two of our cases had gastric wall tissue samples sent and histology results showed nonspecific inflammation with focal degeneration. Early morbidity included recurrent perforation in one case that required redo-laparotomy with gastric repair and gastrostomy. Late complication was recorded in one case who underwent a laparotomy for adhesive bowel obstruction, at the age of one year.

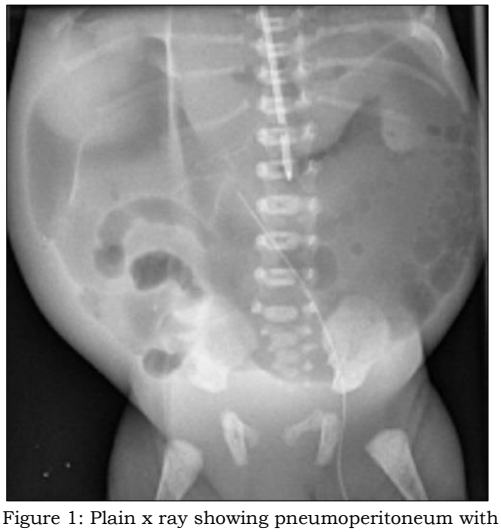

Figure 1: Plain x ray showing pneumoperitoneum with suspected gastric perforation related to the nasogastric tube.

Upper gastrointestinal Contrast study was done routinely a week after the repair of the perforation. This showed minor leak in two cases that were managed conservatively. Repeat contrast after a week, showed a normal repeat contrast with no evidence of leakage (both patients survived). Five preterm neonates succumbed postoperatively (overall mortality rate 38.5%) including one intraoperative death from massive pulmonary hemorrhage. A preterm neonate (650gm) developed recurrent perforation 2 days after the initial surgery, also succumbed 6 days after reoperation. Other causes of death included extreme prematurity, sepsis, and cardiomyopathy related heart failure.

DISCUSSION

SNGP is a rare entity in neonates. Three mechanisms have been proposed for NGP; traumatic, ischemic and spontaneous. Leone et al, suggested that NGP is not spontaneous and most patients have accompanying anomalies including tracheoesophageal fistula or duodenal obstruction.[4] Yang et al, observed an association with duodenal web, hiatus hernia, and malrotation in the affected neonates. Increased intragastric pressure secondary to mechanical obstruction or aerophagia has been postulated as an etiology of gastric perfora-

tion.[5] Although, NGP secondary to traumatic injury by nasogastric tube, and steroid use were suspected in two of our cases, it was obvious that these were only suspicions rather than a definite finding. Cases with obvious predisposing factors such as gastric outlet obstruction or NEC were excluded from our study leaving the etiology in most of our cases undefined. A notable finding in our series is the high prevalence of cardiac abnormalities mainly PDA (7 cases, 53. 8%), a higher number of twins (3/13) and one triplet. whether PDA or its treatment play a role in the mysterious etiology of NGP is totally unknown.

Table 1: Summary of the study patients

Case	Sex	BW(g)	GA(W+D)	Associated anomalies	Age at Perforation	Site of perforation	Outcome
1	F	820	25 +4	PDA	9D	Greater Curvature	Survived
2 (Twin)	F	1920	33	Pulmonary stenosis	4D	Posterior wall	Survived
3	M	970	25+6	PDA	5D	Greater Curvature	*Intra-operative pulmonary Hemorrhage
4 (Twin)	F	1060	28+5		2D	Greater curvature	Survived
5	F	3400	39	chromosomal anomalies, VSD, ASD, PFO, significant PDA(ligated)	34D	Posterior wall	Survived
6	F	1990	29+3	Cardiomyopathy, Hydrops, PDA, Biventricular, hypertrophy and poor function	9D	Fundus	*Heart Failure
7	F	708	25+1		7D	Greater curvature	Survived
8 (IVF)	M	600	25	PDA	5D	Greater curvature	*Sepsis
9	F	636	27+6	PDA	5D	Posterior wall	*Sepsis
10	M	1240	33+3		6D	Anterior wall	Survived
11	F	3100	39	10q deletion. PDA (ligated)	33D	Posterior wall	Survived
12 (Twin)	F	2530	34+5		2D	Fundus	*Sepsis
13 (Triplet)	M	566	24		19D	Anterior wall	Survived

F: female, M: male, GA: gestational age, BW: birth weight, W: weeks, D: days, PDA: patent ductus arteriosus, IVF: in-vitro fertilization, *: Death, VSD: ventricular septal defect, ASD: atrial septal defect, PFO: Patent foramen oval.

Male predominance was noted in many series.[1,6,7] In this study, females outnumbered males (F:M, 2.25:1). SNGP has previously been reported more commonly in full term babies. [6] Recently, prematurity was reported in a substantial proportion of patients with NGP. Lin and colleagues in a report of 15 cases, found a higher incidence of SNGP among premature and low birth weight babies (LBW), and suggested that premature infants are more prone to develop a spontaneous gastric perforation due to the immaturity of the gastric tissue.[1] In the survey from Sato et al, 33.3% (3/9) of patients with SNGP were preterm.[7] Eleven out of 42 neonates with gastric perforation were extremely low birth weight (ELBW) in another study by Byun et al.[8] Prematurity, LBW and ELBW were evident in our series as well where 11/13(85%) patients were preterm. Irrespective of the etiology, SNGP mostly occurs between the 2nd and 7th day of life. [3] This correlates well with our finding where 11 patients (84.6%) developed NGP between 2nd and 9th day of life.

The greater curvature is thought to be the most common site of perforation. Chen et al, in a systematic review of 168 cases with SNGP found a significantly higher predilection for involvement of the greater curvature (73.8%), followed by lesser curvature (n=16; 13.1%), anterior wall (9.0%), and posterior wall (4. 1%).[9] In our series, NGP commonly encountered at greater curvature of stomach followed by posterior wall, anterior wall, and finally the fundus of the stomach.

Urgent surgical repair is the definitive management of gastric perforation and early intervention is crucial to improve survival.[7] Primary closure of the perforation after refreshing the margins with or without gastrostomy is performed as we did in our patients. Rarely, partial or sleeve gastrectomy might be performed in case of massive gastric disruption or multiple perforations.[10] A thorough evaluation of the stomach and the entire gastrointestinal tract for concomitant perforations or abnormalities should be carried out. Postoperative supportive

measures, antibiotics and vasoactive support are basics of postoperative care. Few authors mentioned needle decompression of the abdomen with a large bore intravenous catheter prior to surgery to help improve respiratory and circulatory functions. [11] The latter approach was not practiced in any of our cases. All our patients had primary repair with only one initial gastrostomy. A further gastrostomy was formed in a case of recurrent perforation. We routinely perform contrast study a week after repair in all cases before starting oral feeds, this detected a minor leak in two of our cases.

The prognosis is generally poor with reported mortality rates of 30%-63%. Preterm and LBW neonates have an even higher risk of mortality. Lin et al, reported an overall mortality of 47% that increased to 83% in premature and LBW babies.[1] Chung et al, found male gender and metabolic acidosis to be associated with poor prognosis.[12] Our overall mortality was 38.5% which included preterm neonates. Other main factors that contributed in mortality are sepsis, recurrent perforation and cardiomyopathy related heart failure.

CONCLUSION

The rarity of the condition itself as well as the retrospective nature pose obvious limitations on this study. However, SNGP is extremely rare with all reports in the literature are either case reports or small series. Our findings showed a higher prevalence of SNGP in preterm, LBW and ELBW infants. A potential association with cardiac anomalies especially PDA was also demonstrated. Nevertheless, our survival rate is still promising.

Consent: Authors declared that they have taken informed written consent, for publication clinical photographs/material, from the legal guardian of the patient with an under-standing that every effort will be made to conceal the identity of the patient however it cannot be guaranteed.

Author Contributions: All the authors contributed fully in concept, literature review, and drafting of the manuscript and approved the final version of this manuscript.

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