Spontaneous Rupture of the Gastro-enteric Tract in the Newborn: *

A Report of 13 Cases and Description of a Characteristic X-ray Finding

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SPONTANEOUS rupture of the stomach in the newborn was first reported by Siebold in 1825.[21] Twenty-five years later, Leger[10] reported the first successful operation for this condition. Since 1950 a number of papers on this subject have appeared in the literature (Table 1).

Although *spontaneous* perforation of the gastro-intestinal tract may occur after the newborn period, use of this term should be confined to cases occurring in newborns if accurate analysis of cases is to be made. Perforations associated with distal obstruction should not be classified as *spontaneous,* since obstruction may produce proximal perforation of the gastro-enteric tract.

Various theories of the cause of spontaneous perforations have been entertained including congestion of the bowel wall secondary to aspyhxia, or septicemia,[22] trauma coincident with delivery,[1] excessive gastric acidity, direct or indirect mechanical injury from gavage tubes or resuscitation efforts,[22] congenital mural defects of the gastro-enteric tract, meconium stasis, ruptured diverticulum, and coincident central nervous system abnormalities.[4, 8]

Whether any or all of these mechanisms are responsible is difficult to confirm from careful study of these patients. Recognition of specific etiology from autopsy examination is difficult in many instances, due to postmortem autolysis, and pathologic changes secondary to generalized peritonitis, frequently present in these cases. Anatomic location and gross appearance of the lesion may be of value in determining etiology (Table 2). By far the most common reported type of perforation is a rather long, linear, defect located at or near the greater curvature of the stomach.[3, 11, 16, 20, 22] Because of the anatomic relationship between muscle wall and vascular

TABLE 1. *Spontaneous Perforation of the Gastro-enteric Tract Per Cent Mortality—Reported Series—1950–1962*

Author	Year	Total Cases	Deaths	% Mortality
Legar	1950	1	0	0
Ross	1951	2	1	50
Kellogg	1951	1	0	0
Beattie	1952	1	0	0
Braunstein	1954	5	4	80
Northway	1954	1	0	0
Arnold	1955	1	0	0
Mann	1955	1	0	0
Vargus	1955	11	9	81
Musser	1955	3	3	100
Whittico	1956	2	0	0
Meyer	1957	2	2	100
Moore & Chan	1957	2	1	50
Castleton & Hatch	1958	3	3	66
Linkner & Benson	1959	13	7	56
Hamrick	1959	3	2	66
Cammack	1960	5	4	80
Ogilvy & Owen	1960	1	1	100
Purcell	1962	1	0	0
Present series	1962	13	10	80
Totals		72	46	63

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supply in this location, it is the most logical region in which perforations secondary to congenital muscle defects might occur. In fact, it is the only location in which histologic evidence of deficiency, or absence of muscular layers has been reported.[3, 17, 20, 22]

The next most common type of perforation is a *punched out* lesion of the anterior or posterior wall of the stomach. It is difficult to conceive that congenital muscle defects could produce well circumscribed punched out lesions. Some of these perforations have been documented as having been produced by catheters, while others are clearly not traumatic in origin. The latter may be due to acute peptic ulceration, necrosis secondary to septic emboli, or perhaps related to associated central nervous system disorders.

Microscopic examination of these *punched out* perforations, which on gross examination are not unlike acute perforated peptic ulcers, shows little more than mild inflammatory reaction in contrast to the intense inflammatory reaction in acute peptic perforations in older patients. These perforations may in fact be secondary to peptic digestion for the typical histologic findings of peptic ulcer seen in older individuals may well be less evident in newborn infants. Acute onset, immaturity of cellular defense mechanisms, and lack of chronicity might explain the absence of typical pathologic findings of peptic ulceration in the newborn.

The third most commonly encountered type of perforation is a short, linear, rent, high on the lesser curvature of the stomach, at or near the cardia. Three out of four of these perforations are reported to

TABLE 2. *Spontaneous Perforations of Gastro-enteric Tract Location and Type—Collected Series*

Author	Year	Case Reports	Stomach	Linear	"Punched out"	Duodenum Small Bowel and Colon
Legar	1950	1	1	1		
Ross	1951	2	2	2		
Kellogg	1951	1	1		1	
Beattie	1952	1	1		1	
Braunstein	1954	5	5	5		
Northway	1954	1	1	1		
Arnold	1955	1	1	1		
Mann	1955	1	1	1		
Vargus	1955	11	11	7	4	
Musser	1956	3	3	3		
Whittico	1956	2	1	1		1
Meyer	1957	2	2	2		
Moore & Chan	1957	2	2	2		
Castleton & Hatch	1958	3	3	2	1	
Linkner & Benson	1959	13	13	6	7	
Hamrick*	1959	3	2	1	1	
Cammack**	1960	5	1	1		2
Ogilby & Owen	1960	1	1	1		
Purcell	1962	1	1	1		
Present series	1962	13	8	5	3	5
Totals	12 yrs	72	61	43	18	8

* Transverse tear of pylorus.
** Unknown location.

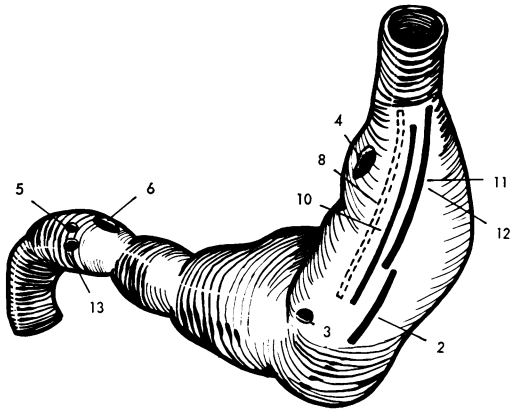

FIG. 1. Location and type of spontaneous perforations of the stomach in this series. Numbers refer to cases listed in Table 3.

have followed *tracheal catheterization,* or use of mechanical resuscitators without appropriate reducing valves and adequate pressure control.

Cronin,[6] in a review of the literature, found that overdistention of the stomach was the commonest cause of spontaneous rupture in children and adults. He also reported that these perforations were usually linear tears along the lesser curvature of the stomach. Since this is the most rigid and least distensible region of the stomach, perforation from overdistention would seem logical in these cases.

Spontaneous perforation of the small and large bowel in newborns has been infrequently reported. Cammack *et al.*[4] reported two cases and three cases are included in this report.

Although speculation concerning the precise etiology of these perforations is intriguing, it is of far less importance than early recognition of the condition, with prompt institution of appropriate treatment. Survival following spontaneous perforation of the gastro-intestinal tract in the newborn requires early recognition, and rapid definitive management. The effect of early diagnosis in reduction of mortality is clearly demonstrated by Linkner and Benson,[11] who reported 13 cases with six survivors, compared to the high mortality rate evident in collected reports (Table 1).

Material

From 1953 to 1962, 13 cases of spontaneous perforation of the gastro-enteric tract were seen at the John Gaston Hospital. Ten were located in the stomach or duodenum and three occurred elsewhere in the enteric tract (Fig 1). Three of these cases have been previously reported by McCormick,[12] and one case was reported by Hand.[7]

Five patients had linear defects and three had *punched out* lesions of the stomach. Two patients had *punched out* defects of the duodenum. Linear tears were located on the greater curvature of the stomach in four patients. None had received any form of resuscitation or intubation prior to the onset of symptoms. Of the five patients with *punched out* lesions of the stomach or duodenum, three had been intubated by catheter at some time prior to onset of abdominal findings.

One of the two remaining cases with punched out perforations expired eight days after admission without benefit of operation. Autopsy revealed a circular perforation 5 mm. in diameter of the anterior duodenal wall and marked generalized peritonitis. Microscopic examination proved the lesion to be perforated duodenal ulcer. The other expired 16 hours after closure of a 5 mm. circular perforation of the anterior wall of the duodenum from the effects of generalized peritonitis. Autopsy failed to establish the etiology of perforation.

One case with a linear tear of the lesser curvature of the stomach was encountered. Resuscitation by positive pressure oxygen given by mask, and both tracheal as well as gastric intubation by catheters had been carried out prior to onset of symptoms. Trauma was thought to be the etiology in this case. Operative closure of the perforation was well tolerated and the patient was

without signs of any abnormality when last seen, two years postoperatively.

Three cases with perforations elsewhere in the enteric tract were encountered. Two patients were thought to have perforations secondary to inflammation. One, with perforation of the cecum, was associated with *pseudomonas* septicemia. Autopsy in this case demonstrated multiple ulcers of the cecum, right colon, ileum and stomach. *Pseudomonas aeruginosa* was obtained in pure culture from the blood postmortem. The other, a perforation of the ileum, at the site of an inflamed Peyer's patch, lived for 18 days after surgery when death from overwhelming sepsis occurred. The third patient with a nongastric enteric perforation had a 2 cm. linear tear of the antimesenteric border of the ileum, without evidence of trauma, surrounding inflammation, or distal obstruction. Successful surgical closure of the perforation was done, and there has been no evidence of any abnormality during the 24-month period of observation following this procedure.

Nine patients were operated upon with three survivors. All of the survivors had laparotomy less than six hours following the onset of abdominal distention, contrasted to an average of 10.4 hours for nonsurvivors. These findings emphasize the necessity of early diagnosis and prompt institution of treatment in patients with spontaneous perforation of the gastrointestinal tract.

Overwhelming sepsis (secondary to peritonitis) was the primary cause of death in eight of nine patients who failed to survive. One patient with minimal localized peritonitis died in acute pulmonary edema which, in retrospect, was apparently related to excessive fluid administration.

Increasing awareness of this entity leading to more prompt diagnosis, should significantly improve results. Prior to 1959 all

TABLE 3. *Spontaneous Perforations of the Gastro-enteric Tract in the Newborn Type-Etiology-Treatment & Results—J.G.H., 1953–1962—13 Cases*

Case	Year	Resuscitative Procedures	Tracheal of Gastric Intubation	Operation Time Post Onset (Hr.)	Type and Location of Perforation	Outcome and Casue of Death
1	1953	No	No	No	Punched out—cecum	Died 8 days post admission—peritonitis
2	1954	No	No	Yes 17	Linear—greater curvature stomach	Died 11 hours postop.—stomach
3	1955	Yes	Yes	No	Punched out—ant. wall stomach	Died 4 days post admission—peritonitis
4	1955	Yes	Yes	Yes 5	Punched out—ant. wall stomach	Died 48 hours postop.—pul. edema, peritonitis
5	1956	No	No	No	Punched out—ant. wall 1st portion duod.	Died 8 days post admission—peritonitis
6	1957	Yes	Yes	No	Punched out—ant. wall pylorus	Died 3 days post admission—peritonitis
7	1958	No	No	Yes 24	Punched out—distal ileum	Died 18 days postop.—disseminated moniliasis
8	1960	Yes	Yes (Both)	Yes 5	Linear—lesser curvature from cardia to incisura	Living and well 2 years postop.
9	1960	No	No	Yes 2	Linear—distal ileum	Living and well 18 months postop.
10	1961	No	No	Yes 8.5	Linear—greater curvature stomach	Died 3 hours postop.—peritonitis
11	1961	No	No	Yes 3	Linear—greater curvature stomach	Died 6 hours postop.—peritonitis
12	1961	No	No	Yes 3.5	Linear—greater curvature stomach	Living and well 1 year postop.
13	1962	No	No	Yes 5	Punched out—ant. wall 1st portion duod.	Died 16 hours postop.—peritonitis

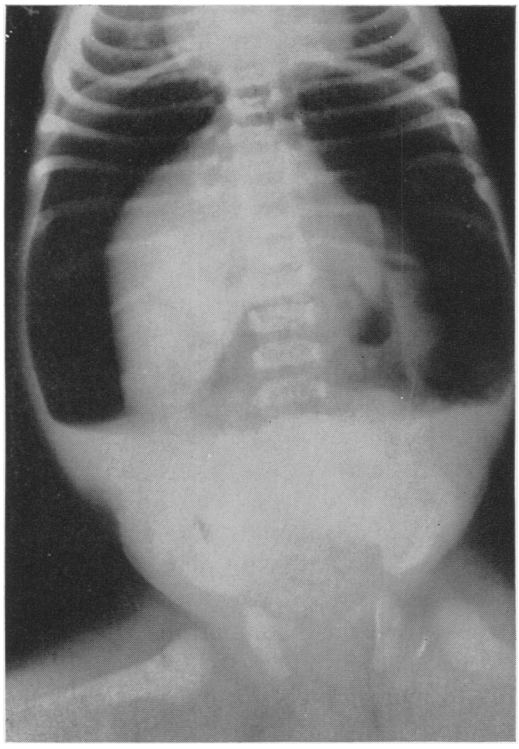

FIG. 2. Upright film of the abdomen, 48 hours postpartem in Case 8.

of seven patients with spontaneous perforation of the gastro-enteric tract in this series died of their disease. Only three of these patients were operated upon. Two of the three operative deaths were related to delay in diagnosis (average 15 hours from perforation to laparotomy). The third death following operative treatment (previously mentioned) was possibly related to overhydration (Table 3).

Since 1960, renewed interest, and awareness of this clinical entity has permitted earlier diagnosis and institution of treatment. All of the six patients seen since 1959 were operated upon with closure of the perforation. Three of these patients survived. The three deaths were attributed to generalized peritonitis. Despite efforts to establish prompt diagnosis, prolonged delay of operative treatment was considered to be partially responsible for these deaths.

Diagnosis

Early diagnosis is the most important single factor in recovery. The following case report details the classical findings nearly always seen in this condition.

Case Report

A full term, six pound, five ounce, colored male was born on 6-16-61 following uncomplicated gestation. A normal spontaneous delivery. Respirations began without difficulty at birth and no form of resuscitation was required. Normal meconium stools were noted 24 hours following delivery. Two days postpartum, acute onset of vomiting (a small amount of green stained material), massive abdominal distention, and rapid shallow respirations developed. An upright film of the abdomen revealed massive pneumoperitoneum (Fig. 2). Parcentesis by needle, inserted into the right upper quadrant of the abdomen, yielded a large amount of air and 50 cc. of bile stained fluid. His marked respiratory difficulty was immediately relieved by this procedure and laparotomy was carried out three and a half hours after onset of symptoms. At operation, under general endotracheal anesthesia, a 5 cm. linear tear of the greater curvature of the stomach extending downward from the cardia was found. There was no active bleeding from the apparently necrotic edges of the perforation. The rent was repaired in two layers and a Stamm gastrostomy was performed. His postoperative course was uneventful. The patient was last seen one year postoperatively, when he was without evidence of further difficulty.

The clinical picture as illustrated by this case is so characteristic of newborns with spontaneous perforation of the stomach that it is difficult to understand why early diagnosis is not the rule rather than the exception in these cases.

Nearly all patients with spontaneous perforation of the gastro-enteric tract exhibit acute onset of massive abdominal distention associated with respiratory difficulty; these findings can be considered as presumptive evidence of spontaneous perforation of the gastro-enteric tract. If, in addition, any form of resuscitation or intubation was performed, the diagnosis of traumatic perforation of the stomach is strongly suggested. Absent liver dullness to percussion

is almost always found on physical examination and when this diagnostic sign of pneumoperitoneum can not be demonstrated, diagnosis can be quickly confirmed by an upright x-ray film of the abdomen. All cases in this series demonstrated massive pneumoperitoneum on x-ray examination of the abdomen. The pneumoperitoneum often has a characteristic conformation which resembles the outline of pack-horse saddle bags. We have chosen to call this x-ray picture the *saddle-bag* sign. The saddle bag configuration corresponds to the area of radiolucency produced by large amounts of intraperitoneal air displacing the liver and spleen downward and toward the midline (Fig. 3).

Abdominal distention usually presents within 24 to 36 hours following delivery and resuscitative efforts in cases of traumatic perforation. Patients in our series, with linear perforations of the greater curvature of the stomach unassociated with intubations, had satisfactory progress for several days before the onset of acute abdominal distention. Vomiting in perforations of the stomach and duodenum was infrequent and of minimal amount. All 13 patients had normal meconium stools, which is an important diagnostic aid in ruling out intestinal obstruction as the cause of abdominal distention. Pneumoperitoneum of *saddle-bag* configuration on upright x-ray films of the abdomen confirms the diagnosis.

Treatment

Respiratory difficulty should be relieved as soon as possible by needle aspiration of the peritoneal cavity. The patient should then be prepared for laparotomy without further delay. When perforation is recognized early, general endotracheal anesthesia is satisfactory and safe. If the diagnosis has been delayed, local infiltration anesthesia is the method of choice.

An upper midline incision is the most useful approach, since the majority of cases will have perforations located high on the greater curvature of the stomach.

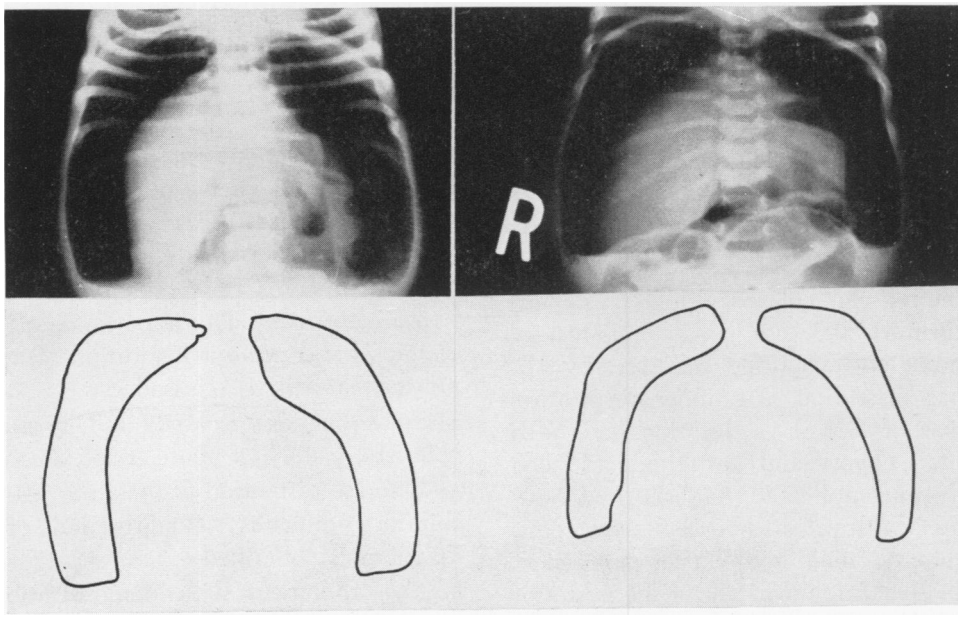

FIG. 3. Characteristic *saddle-bag* configuration of pneumoperitoneum in spontaneous rupture of the stomach in the newborn.

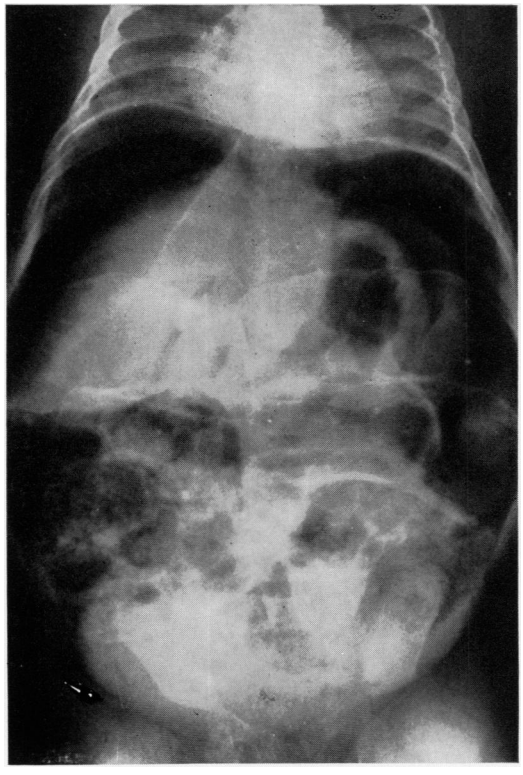

FIG. 4. Upright film of the abdomen on admission of a five-month-old infant with a punched out perforation of the anterior wall of the stomach.

A Stamm gastrostomy is useful in postoperative care of these infants.

Gastric Perforations in Infancy

Two cases of gastric perforation occurring after the newborn period, not included in analysis of cases of perforations in the newborn in this report are of interest. An eight-month-old colored female was admitted on 11-30-53, with a 24 hour history of nausea and vomiting followed by abdominal distention. An upper respiratory infection of two days duration had been present. Delivery and gestation had been entirely uncomplicated, and she had been in good health prior to onset of symptoms. Laparotomy, nine hours after admission, revealed a 1 cm. linear tear of the posterior wall of the greater curvature of the stomach near the cardia, and generalized peritonitis. Death occurred four hours following repair of the perforation. At autopsy the suture line was found to be intact and positive findings were acute inflammation of the submucosa with necrosis in the area of perforation.

The second case occurring in infancy was a five-month-old colored female, admitted 3-16-62, with a history of normal spontaneous delivery, but with subsequent intermittent episodes of upper respiratory infections and frequent vomiting since birth. She had been treated with antibiotics during the week prior to admission for respiratory infection. One day before admission, fever followed by marked abdominal distention developed. Admission upright x-ray films of the abdomen revealed extensive pneumoperitoneum (Fig. 4). At laparotomy, three and one-half hours later a 1 cm. punched out lesion of the anterior wall of the pars media of the stomach with localized peritonitis was found. Her postoperative course was uneventful. Extensive postoperative workup, including barium studies of the gastroenteric tract was entirely normal. The patient was well with no reported further difficulty when last seen five months postoperatively.

Summary

The term *spontaneous perforation* of the gastro-intestinal tract is defined, and a plea to confine its use to cases occurring in the newborn period is made.

Thirteen cases of spontaneous perforation are reported. The importance of early diagnosis and prompt institution of appropriate treatment is stressed.

Early diagnosis is facilitated by a characteristic history of acute onset of massive abdominal distention associated with respiratory difficulty. Confirmation of the diagnosis is obtained by an upright film of the abdomen, with demonstration of pneumoperitoneum of *saddle-bag* configuration. Two cases of *spontaneous* perfora-

tion of the stomach in infants past the newborn period are presented.

Prompt diagnosis and surgical treatment is the only hope for survival in these infants.

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