Subphrenic Extrathoracic Rupture of the Esophagus: *

First Reported Case

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IT IS the purpose of this communication to report the first recorded instance of subphrenic, extrathoracic rupture of the esophagus. Several instances of esophageal rupture which involved both epiphrenic and subphrenic esophagus, and even stomach, have been reported and were recently summarized by Perkoff and Sensenig.[11] These comprise but a small fraction of all cases of esophageal rupture, however, as this catastrophe is generally confined to the chest cavity.

The case herein reported is of subphrenic, extrathoracic, retroperitoneal esophageal rupture, treated by transabdominal drainage, with survival.

Case Report

R. E., a 53-year-old white man, was admitted to the Valley Forge General Hospital at 4:30 a.m., October 28, 1962, because of severe substernal and epigastric pain. Seven hours prior to admission the patient had a headache after drinking several bottles of beer. He took an aspirin and attempted to wash it down with water but he felt it lodge "in the throat, just before it got to the stomach." He then gulped another glass of water and was suddenly stricken by severe, stabbing, substernal pain. Shortly thereafter he began to vomit bright red blood in small amounts. The pain increased while he was driven for several hours to the hospital. On admission he described the pain as severe and steady, radiating to the back, relieved somewhat by sitting.

The patient stated that for about two years he had had difficulty in swallowing, feeling that food was sticking in his throat, but he said that x-rays showed only that he had an "acid stomach." He denied other gastrointestinal symptoms. He admitted to heavy beer intake and used aspirin frequently for various minor complaints.

Physical examination revealed a stocky, middle-aged man, writhing in abdominal pain and complaining of severe thirst. Temperature was 37.7° C.; pulse was 100/min.; blood pressure was 150/100 mm. Hg.; respirations were 32/min. Examination of the abdomen disclosed spasmodic rectus muscle contractions whenever palpation was attempted. There was exquisite epigastric tenderness, but no definite rebound or percussion tenderness could be elicited. Bowel sounds were hyperactive and of normal pitch. The remainder of the physical examination was nonrevealing.

Laboratory studies were of little aid: white blood count was 9,400 with 80% polymorphonuclear leukocytes; hemoglobin 15.8 Gm./100 ml.; hematocrit 46%; serum amylase 27 mg./100 ml.; blood urea nitrogen 13 mg./100 ml. Serum electrolyte concentration and electrocardiogram were normal.

Admission x-ray films of the chest and abdomen were interpreted by the surgeon as showing only elevated hemidiaphragms.

Shortly after admission intravenous fluids were infused, and morphine was administered with dramatic relief of pain and restlessness. A nasogastric tube was inserted and yielded blood from the esophagus and bile-stained fluid from the stomach. Diagnostic paracentesis produced several drops of hemorrhagic fluid resembling beef broth. The working diagnoses at this time were incomplete esophageal rupture and acute pancreatitis.

Conservative management led to considerable improvement in the patient's condition over a 48-hour period, though abdominal tenderness persisted and progressive fever was noted.

On the third hospital day the initial chest and abdominal x-rays were reviewed. The radiologist's interpretation was abscess in the area of the lesser peritoneal sac. Repeated x-ray films showed to

* Submitted for publication January 29, 1964.

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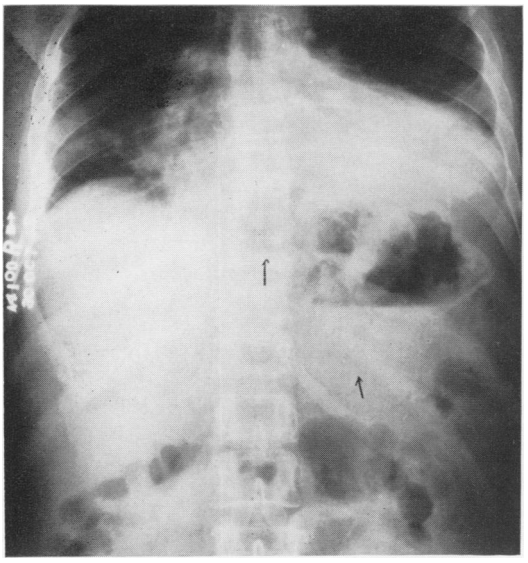

FIG. 1. Upright abdomen. Study done on the third hospital day shows best the air-fluid level (upper arrow) over the epigastrium separate from the displaced gastric fundus. Note elevated left hemidiaphragm and gross parenchymal reaction in the left base, result of adjacent subphrenic inflammatory process. Multiple air bubbles within a soft tissue density (above lower arrow) proved at operation to be part of abscess.

better advantage the same findings, primarily an air-fluid level superimposed over the epigastrium and independent of the gastric air-fluid level which was displaced inferolateral (Fig. 1). In an attempt to better define the nature of this process an aqueous contrast medium (Hypaque) was administered orally. This revealed subphrenic extravasation of the opaque material at a point immediately proximal to the gastro-esophageal junction into a large space in the region of the lesser peritoneal space (Fig. 2).

Operation was performed shortly after the contrast x-ray study. The abdomen was entered through an upper midline incision. On opening the peritoneum, the only immediately apparent abnormality was fibrin deposition between the stomach and the left lobe of the liver in the region of the esophageal hiatus. The lesser peritoneal space was entered by division of the gastrocolic omentum. No purulent exudate was encountered, but the pancreas was approximately twice normal size and hemorrhagic. Cephalad to the pancreas, a soft cystic area, thought to be the abscess, was palpated retroperitoneally near the gastric cardia. Division of the peritoneum overlying this mass released about 200 cc. of bloody, foul-smelling pus, which was cultured. By inserting a finger into the peritoneal rent from which pus was exuding, an extremely friable area in the left posterior esophageal wall immediately proximal to stomach could be palpated, though the edges of the rupture, and its dimensions could not be discerned without risking further esophageal injury. The abscess cavity was evacuated as thoroughly as possible, and the contaminated peritoneal surfaces were irrigated with saline. A Stamm gastrostomy and a Witzel jejunostomy were performed. The area of the abscess cavity and the subphrenic space were generously drained; the wound was closed with wire figure-of-eight sutures to linea alba and peritoneum and with silk sutures to skin.

Postoperatively no oral intake was allowed, and the stomach was kept empty by means of the gastrostomy. The jejunostomy was utilized for feeding. The abscess culture grew out a mixed flora of *Proteus, Clostridia,* and *Streptococci* sensitive to several antibiotic drugs which were utilized in large doses.

For 3 weeks the patient's condition remained critical. Atrial fibrillation, severe bilateral pneumonia, and marked azotemia required careful management. Large extrarenal fluid losses from the gastrostomy brought about fluid and acid-base imbalance. Retroperitoneal hemorrhage, resulting from progression in size of the original abscess with erosion of vascular structures, prompted counter-drainage of the left subphrenic space by a posterior approach. The inevitable esophagocutaneous fistula developed.

Improvement followed, and once the patient *turned the corner,* his progress was rapid and complete.

Since discharge from the hospital, the patient's wounds have completely healed. Barium x-ray study of his esophagus on April 3, 1963, showed complete healing with the only residuum a small cicatricial "diverticulum" at the site of rupture. There is no evidence of stricture clinically or by x-ray. The patient has no dysphagia, heartburn, dyspepsia, regurgitation or food intolerances. He is taking an unrestricted diet and has regained 30 of about 45 pounds lost. Aspirin and beer are avoided by his own choice.

Comments

The clinical course of a patient with esophageal rupture is sufficiently characteristic that if the physician considers the diagnosis, no diagnostic problem exists. It has been said that one can postulate a correct diagnosis in many cases by a telephoned history.[8] Briefly, the patient is likely to be a middle-aged man who may have a

history of gastro-intestinal difficulty. Following a heavy meal or alcohol intake, he suffers the onset of excruciating substernal and epigastric pain, usually after vomiting. The pain is aggravated by breathing, reclining or moving about and may be initiated by a sensation of something giving way. A physician is usually consulted immediately, and the patient is found in extreme pain, in a sitting position, with a rigid upper abdomen. Cyanosis, hypotension, tachycardia and tachypnea are common. Chest findings are abnormal and may include signs of mediastinal air, mediastinal widening, hydrothorax, pneumothorax, or a combination of these. Subcutaneous emphysema may be present in the neck if the patient is seen after several hours of illness. Chest x-ray film is the most reliable confirmatory diagnostic measure, and still more precise diagnosis follows utilization of radiopaque-medium swallow and thoracentesis.

In the case presented here, although the history was characteristic, physical findings and admission plain x-ray films were not definitely confirmative of esophageal rupture. Certainly it would have been foolhardy to undertake thoracotomy on the basis of the admission studies; nevertheless, it seems evident that had we been more aggressive in diagnostic efforts, an accurate diagnosis could have been established within a short time. X-ray study of the upper gastro-intestinal tract by means of appropriate medium would have been the simple method of investigation and would have established the diagnosis. Contrast media study of the esophagus was delayed because of seemingly progressive improvement in the patient's condition, because of a belief that the chief pathologic process was hemorrhagic pancreatitis, and because of the improbability of rupture of the esophagus producing such sparse thoracic findings. Prompt diagnosis in cases of this type, of course, is a prime objective.

Once the diagnosis was established, our course seemed clear. Definitive esophageal repair seemed out of the question because of the elapsed time since rupture. Drainage of the area of rupture was thus elected. The usefulness of the gastrostomy and feeding jejunostomy was demonstrated in the postoperative period.

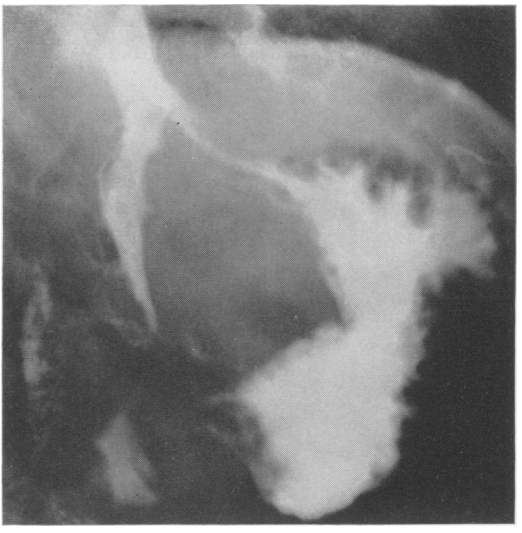

FIG. 2. Fluoroscopic spot of gastroesophageal junction done about 6 weeks following admission shows extravasation of the opaque subphrenically. Note anterior displacement of stomach and increased thickness of hemidiaphragm. Less well-defined streaks of opaque are actually delineating esophagocutaneous fistulous tracts.

The problems in the postoperative period were difficult but, with the exception of the esophagocutaneous fistula, they were those pepuliar to any complicated case involving leakage peritonitis with septic complications. Judicious management of these problems was followed by complete recovery.

Discussion

The theories regarding etiology and pathogenesis of esophageal rupture have been discussed in detail previously.[1, 2, 3, 5]

The unique feature of this case is the subdiaphragmatic position of esophageal rupture. In reviewing the case it is of extreme interest to attempt an explanation of this occurrence. X-ray films of the patient's upper gastro-intestinal tract taken more

than a year prior to rupture revealed a small sliding esophageal hiatal hernia and a badly deformed duodenal bulb without active duodenal ulceration. Although the physiology of the esophagus and the esophagogastric junction are at best incompletely understood, it would appear that a hiatal hernia is the most likely cause of disturbed anatomy and physiology which might explain the unusual position of rupture; further, it is quite possible that the same disturbances may have set the stage for, and precipitated, the rupture as well. Dysphagia, with the sensation of food sticking in the esophagus, is a common manifestation of hiatal hernia and probably results from disturbed mechanics during esophageal food transport as well as from peptic esophagitis and cardiospasm secondary to gastric acid reflux. This may have been the cause of the patient's long history of pre-rupture dysphagia. It is of further interest that antecedent gastrointestinal symptoms are present in over a third of the cases of esophageal rupture.[9] The possible role played by peptic esophagitis in esophageal rupture has been discussed in the literature,[4, 6] and an attempt to evaluate the loss of mucosal integrity was made by Derrick *et al.*,[3] who found that a decided decrease in pressure was required to rupture the esophagi of cadavers following mucosal removal. What part salicylate ingestion may have played in the production of esophagitis in this case is speculative. We postulate that the esophagus ruptured when the patient caused an acute increase in intraluminal esophageal pressure by swallowing water against an acutely obstructed lower esophagus. Though this is not a common mechanism, it does occur.[2, 5] We further postulate that when the lead point of the obstructed esophageal segment (also presumably the point through which rupture occurred) descended during deglutition, unimpeded by a normally snug phrenic crus, subphrenic rupture occurred. Since rupture most commonly occurs posterolaterally on the left,[2, 5] it is not surprising that the rupture was retroperitoneal.

Regardless of the location of rupture, the treatment following an early diagnosis is prompt operative closure of the esophageal rent with appropriate drainage. In the case of subphrenic rupture, the surgeon should avoid contamination of the chest, if possible, by utilizing transabdominal esophageal repair. Transabdominal repair of the ruptured esophagus has been performed successfully on two recorded occasions.[7, 13] Obviously, if the chest has been entered when subphrenic rupture is discovered, chest contamination must be dealt with and septic thoracic complications anticipated. The situation requiring fine judgment would be early subphrenic rupture technically impossible to repair when approached by laparotomy; the choice would lie between 1) foregoing esophageal repair in favor of drainage procedure and 2) producing chest contamination by conversion of the laparotomy incision to a thoracoabdominal approach in order to effect esophageal closure.

When diagnosis has been delayed, a more conservative approach is indicated, and drainage of the area of rupture is the principle objective. Complementary procedures will vary with circumstances.

Some idea of the difference between these two approaches may be gained from Samson's analysis,[12] in which drainage procedures resulted in a mortality of 47 per cent, with a morbidity of from 6 weeks to 1½ years, whereas direct repair resulted in a mortality of 33 per cent, with a morbidity of from 13–22 days. It must be remembered, however, that drainage procedures have been reserved for late cases, and, as mortality rises steeply with delay in initiation of definitive therapy,[10] Samson's figures may not be as indicative as they seem at first glance.

The possibility that esophageal rupture can occur below the diaphragm without involving the chest has always existed. It

seems improbable that this case represents the first instance in which such a rupture did in fact take place, but a thorough search of the literature has failed to reveal another case of this type. We regard it as a rare variant of an uncommon surgical emergency.

Summary

A case of subphrenic extrathoracic rupture of the esophagus has been reported. The following aspects of the case deserve emphasis:

1) The history was classic for esophageal rupture.

2) Prompt utilization of every diagnostic means, including contrast x-ray films of the esophagus, is recommended to establish an early diagnosis.

3) It is felt that the presence of a sliding esophageal hiatal hernia was the dominant factor leading to the unusual location of the esophageal rupture.

4) The general principles of therapy for any esophageal rupture are the same. Subphrenic rupture, however, should be treated by an extrathoracic operative approach if possible.

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Addendum

Since this manuscript was originally prepared for publication, several papers have been published which relate directly to our case.

The collective review of spontaneous esophageal rupture by Tesler and Eisenberg [3] provides comprehensive discussion of the subject and again confirms the absence of a case of subphrenic extrathoracic rupture of the esophagus reported in the literature.

Bruno *et al.*[1] discuss the Mallory-Weiss type of esophagogastric mucosal lacerations and spontaneous esophageal rupture. They propose that, since the clinical implications of the two entities are so disparate, esophageal rupture of the spontaneous type should be known as the "Boerhaave syndrome," commemorating the original report of the entity by Boerhaave. The case presented here would then be a variant of the Boerhaave syndrome.

The recent discussion of the esophagogastric junction from the Mayo Clinic [2] reflects the difference of opinion concerning the physiology and anatomy, as well as pathophysiology, of this critical area. Further, if such an entity as the "hypertensive gastroesophageal sphincter" exists, our patient's prerupture dysphagia may have been related to such a mechanism.

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