

CARCINOMA ARISING IN REDUPLICATION OF THE STOMACH (GASTROGENOUS CYST): A CASE REPORT*

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DUPLICATIONS OF THE alimentary tract are relatively uncommon, and are generally recognized for the first time when obstruction of the involved segment develops. Gross, Holcomb and Farber⁷ reported 68 such cases from the Children's Hospital in Boston, the location of the reduplication in these cases ranging from the finding of large bowel mucosa in the region of the base of the tongue to reduplication of the rectum. The terminal ileum appears to be the most common site of such reduplication, and cysts lined with gastric mucosa in the posterior mediastinum,^{13, 17} or arising in the mediastinum and presenting in the neck, are second in frequency of occurrence. Enterogenous cysts in the pre-sacral area have been reported also.¹²

Reduplications of the stomach (gastrogenous cysts) are not often recorded in the literature. These cysts are presumably congenital in nature, and have the common anatomic characteristics of a wall containing all the normal layers of the stomach (serosa, muscularis, mucosa), except in the region where they are attached to the stomach proper, in which there is no serosa, and the muscularis is common to the cyst and to the stomach itself. Various theories of origin have been proposed, including reversion to earlier phylogenetic forms, and the development of diverticula of the stomach which later become pinched off, resulting in cyst formation. Perhaps the most acceptable theory is that of Bremer, as elaborated by

Gross and co-authors.⁷ This theory takes cognizance of the fact that in the embryo at about six weeks, the lumen of the intestine is occluded by the rapid proliferation of epithelial cells. Subsequently, vacuoles appear in the midst of this profusion of cells, and in the ordinary course of development, these vacuoles coalesce to form a lumen again. It is supposed that the persistence of one or more of these vacuoles separate from the lumen of the main channel may lead to duplication.

Scott and Payne¹⁴ have reported a case of a patient operated on because of pyloric obstruction, in the stomach of whom were found multiple cystic structures in the gastric wall, with an occasional associated abscess. They classified this condition as diffuse congenital cystic hyperplasia of the stomach. Metz, Householder and DePree⁵ reported a case of a congenital cyst produced by the development of two transverse septa in the stomach, resulting in complete pyloric obstruction in a new-born male, and successfully treated by perforation of the septa. Bikoff,³ Ellis⁴ and Ferraro⁶ have each reported single cases of gastric cysts recognized in infancy and treated surgically. In the first two mentioned reports, the cysts were removed with a portion of the gastric wall, the stomach being closed, and the result in each case was successful. In Ferraro's case, the cyst had been previously "drained," and was finally resected with the sinus tract resulting from drainage, as well as with a portion of the gastric wall, but the infant

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FIG. 1. Roentgenogram showing constriction of the distal portion of the gastric antrum.

died as a result of multiple systemic disorders. Berg and Marx² reported a case of a two-year-old white male with pyloric obstruction on the basis of such a cyst, successfully treated by subtotal gastric resection. A similar case was reported by McCutchen and Josey⁹ in a five-month-old

female child, with the added unusual finding of a Meckel's diverticulum adherent to the cyst, through which the cyst communicated with the terminal ileum. This was satisfactorily treated by excision of the cyst and closure of the stomach. Only five reports^{1, 10, 11, 15, 16} of such cysts discovered for the first time in the adult have been found in the literature. One of these¹⁵ was so extensive that total gastrectomy was necessary for its removal, with a satisfactory outcome. Another¹⁰ was satisfactorily treated by partial gastric resection. Two^{11, 16} were treated successfully by local excision of the cyst, and in the fifth case¹ the cyst was so extensive that in order to avoid total gastrectomy, the cyst was partially excised, and the remainder of the wall cauterized with formalin 10 per cent, followed by marsupialization of the cyst remnant. The result in this case was also satisfactory.

The purpose of this communication is to report a case in which a reduplication of the stomach was not recognized until the patient, a white female, developed anemia at the age of 64. Following resection, it was determined that carcinoma had developed in the gastrogenous cyst. To our knowledge,

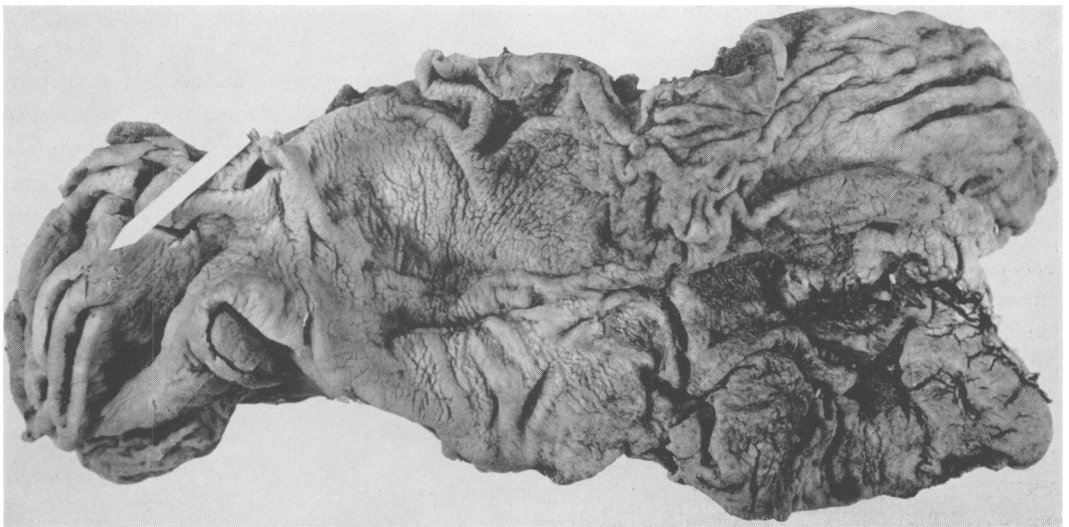


FIG. 2. Stomach opened along its greater curvature. Arrow points to the intact gastric mucosa overlying the intramural cyst.

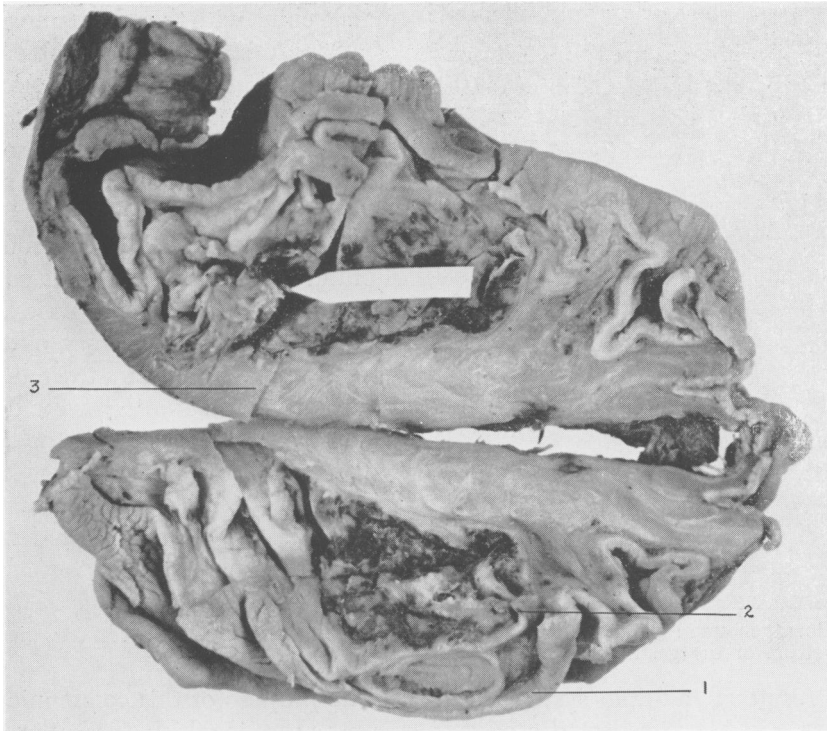


FIG. 3. Gastric cyst bisected and reflected. Arrow designates polypoid growth. Note the layer of gastric mucosa (1) in relation to the lining of the cyst wall (2). Thickened fibro-muscular wall (3) is infiltrated by adenocarcinoma.

this is the first such case reported in the literature.

CASE REPORT

Mrs. A. S. R., a 64-year-old white female housewife, was admitted to the Roper Hospital on January 7, 1954, because of weakness, anorexia and malaise of 3 months' duration. For the preceding 2 months she had noticed some degree of constipation, accompanied by a weight loss of 13 pounds, but without obvious change in the caliber of her stools, and without melena, nausea or vomiting. Two years previously she had noted severe gaseous eructations, which had been relieved by soda without recurrence. Aside from known arthritic symptoms referable to her right shoulder and left knee, her past history and the review of systems was essentially negative.

On physical examination, her temperature was 100°, pulse 74, and blood pressure 170/100. A fluc-



FIG. 4. (x19) Section through stomach wall shows intact gastric mucosa (A) and the lumen of the gastric cyst (B) which lies within the muscular wall of the stomach.

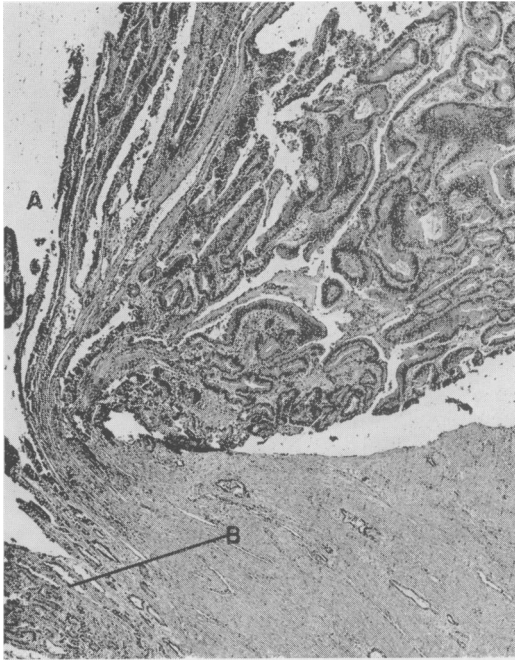


FIG. 5. (x19) Section of the gastric cyst shows the polyp (A) and its relation to the mucosal lining of the cyst (B).

tuant mass 5 cm. in diameter was noted attached to the skin over her left patella, and was interpreted as a sebaceous cyst. Her skin and mucous membranes were pale. Just beneath the right costal margin, a vaguely outlined firm mass, approximately 6 cm. in diameter, was palpable. This mass was slightly movable. The remainder of the physical examination was not significant. A routine urinalysis was essentially negative except for 15 to 20 WBC/hpf. Her erythrocyte count was 3,000,000; hbg., 8 Gm.; leukocyte count, 9,400, with a relatively normal differential count. Marked microcytosis and hypochromia were noted. Her prothrombin activity was 85 per cent of normal, and her bleeding time 3 minutes, and clotting time 4 minutes. The BUN was 24 mg. per cent and the total serum proteins 6 Gm. per cent with 2 Gm. per cent albumin, 1 Gm. per cent alpha globulin, 0.84 Gm. per cent beta globulin, and 1.94 Gm. per cent of gamma globulin. Urine cultures indicated the presence of a pure culture of aerobacter aerogenes. No free hydrochloric acid was recovered from the stomach after the administration of histamine, and the maximum response in total acidity was 14°. An electrocardiogram was essentially normal except for a sinus tachycardia. A roentgenogram after the oral administration of barium indicated the presence of a constant constriction of the distal 3 cm. of

the antrum of the stomach (Fig. 1), with no delay in the passage of the barium. This was interpreted as probable carcinoma of the gastric antrum. Barium enema and cholecystogram showed no abnormality.

With a presumptive diagnosis of carcinoma of the antrum of the stomach, the patient was prepared for operation by the administration of vitamins, blood transfusions, and intravenous protein hydrolysate, and the urinary tract infection was successfully treated with terramycin administered orally, with the result that the urine became clear and her temperature returned to normal.

On January 19, 1954, under general endotracheal anesthesia, the abdomen was explored through an upper curvilinear transverse incision, transecting both rectus muscles. In the distal end of the antrum of the stomach a mass of rubbery consistency, approximately 6 cm. in diameter, was palpable. There was no serosal invasion, and no obvious lymph node enlargement, nor was there any evidence of metastases to the liver, para-aortic nodes, or in the cul-de-sac. The remainder of the exploration was within normal limits. Under the assumption that this represented a relatively early carcinoma of the antrum of the stomach, a radical subtotal gastric resection was carried out, removing the greater omentum and gastro-colic omentum, the greater portion of the gastrohepatic ligament, and the lower three-fourths of the stomach. Gastrointestinal continuity was restored by an ante-colic short loop Hofmeister gastro-jejunostomy. The appendix was removed as an incidental procedure. On completion of the operation, the resected stomach was opened, and it was then noted that the gastric mucosa was intact, and moved fairly freely over a submucosal mass approximately 6 cm. in diameter (Fig. 2). On cutting into this mass, it was apparent that it was a cystic cavity lined at least in part with epithelium, filled with necrotic material and cloudy fluid, and containing several small polypoid structures adherent to its lining (Fig. 3).

Following operation, the patient developed a rather persistent paralytic ileus which required intubation for seven days, but which gradually subsided. On her 6th postoperative day it was noted that she was mildly jaundiced, but in view of the many blood transfusions she had received before and during her operation it was assumed that this was on the basis of hemolysis due to a mild blood transfusion reaction. This assumption was apparently confirmed by the subsidence of the jaundice before her discharge from the hospital on the 19th postoperative day, at which time she was taking a soft diet in five feedings daily, and was ambulatory. For the first few weeks after discharge the patient had a rather difficult time because of recurrent abdominal cramps and mild anemia, which was

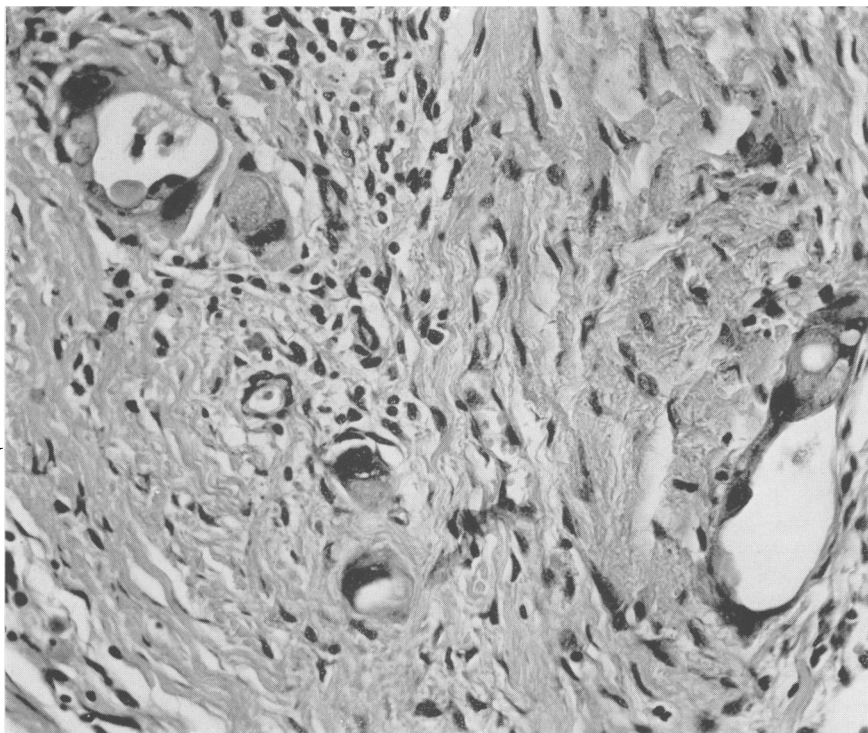


FIG. 6. Sections through the muscular wall of the stomach shows infiltration by adenocarcinoma (x300).

satisfactorily corrected by the administration of iron and liver extract. During this time she lost about 10 pounds in weight, but after the first 3 months her weight stabilized, and there has been no further significant weight loss. When last seen on January 19, 1955, one year after operation, she was getting along well, and examination revealed no evidence of recurrent carcinoma.

Pathologic Examination. The specimen consists of a portion of stomach which measures 23 cm. in length along its greater curvature. When opened, there is a nodular elevation of intact gastric mucosa by a subsurface, 4-cm., slightly fluctuant mass. The gastric mucosa moved freely over this mass, and is apparently not attached to it. A horizontal section through the stomach wall and the mass reveals a cystic structure containing mucoid material. Arising from the wall of the cyst is a friable polypoid structure 1.5 cm. in length. In the attached mesentery there is a firm, 0.7-cm. lymph node. The appendix is also received and is grossly not unusual.

Histological sections through the stomach wall and cystic mass (Figs. 4, 5 and 6) reveal an intact normal gastric mucosa. The intramural cyst is lined by columnar epithelial cells, some of which show secretory activity and which in some areas form

well-differentiated glandular structures simulating normal gastric mucosa. The polypoid structure is likewise comprised of glandular epithelial cells which show frequent mitoses, together with nuclear and cellular variations and infiltration of the underlying muscle coat of the stomach. The lymph node is completely replaced by a similar gland forming malignant epithelial growth.

SUMMARY

A case of reduplication of the stomach (gastrogenous cyst), in which carcinoma developed, is presented. This condition was first recognized as a result of the development of anemia in a 64-year-old white female, and was treated by radical subtotal gastric resection, with a satisfactory immediate result.

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