Gastric-Type Carcinoma Arising in Duplication of the Small Intestine *

THOMAS MICOLONGHI, M.D., GEORGE F. MEISSNER, M.D.**

Providence, R. I.

DUPLICATIONS of the alimentary tract have been described with increasing frequency in the literature of the past 50 years. They manifest themselves generally during the first decade of life by symptoms resulting from intestinal obstruction, melena, or anemia, or may be discovered incidentally at laparotomy or postmortem examination in adults. In this report is presented an unusual case of duplication of the ileum in an adult, containing an adenocarcinoma, which apparently arose from ectopic gastric mucosa lining the duplication.

Case Report

The case is that of a 54-year-old white male admitted to the Rhode Island Hospital with complaints of intermittent abdominal pain and weight loss of about one month's duration. This was accompanied by a marked diminution in appetite. The pain appeared to localize in the left lower quadrant at the time of admission. The patient noticed streaks of blood in his stools, attributed to hemorrhoids. The patient was moderately undernourished on physical examination. There was moderate tenderness and a suggestion of a mass in the left lower quadrant. Bowel peristalsis was apparently normal. Laboratory studies showed a hemoglobin level of 9.9 grams, white count of 12,500 (73 neutrophiles, 20

lymphocytes, 4 monocytes, 3 eosinophiles), total protein of 5.5 grams and albumin of 3.4 grams per 100 cc. The urinalysis was negative. A barium enema performed shortly after admission showed displacement of the sigmoid and lower descending colon by a left lower quadrant mass. There was no evidence of polyp, diverticula, or filling defects in the colon. The clinical differential diagnosis lay between carcinoma of the sigmoid colon or diverticulitis with perforation and abscess formation. At laparotomy there was a mass in the ileum, which appeared to extend into the adjacent mesentery and was adherent to a portion of the sigmoid colon. A wide resection of ileum and attached sigmoid colon was performed, followed by end-to-end anastomosis. Examination of the remainder of the abdomen showed small nodules of apparent metastatic tumor throughout the peritoneal cavity. The clinical postoperative diagnosis was: obstructing adenocarcinoma of ileum with metastases. The patient was discharged from the hospital on the fifteenth postoperative day. He died at home three months later. Permission for autopsy was not granted.

On gross examination, the specimen consisted of 42 cm. of small bowel with attached mesentery (Fig. 1). The small bowel was lined by normal ileal mucosa, but at the midportion of the specimen on the mesenteric side, there was an ulceration measuring 2 cm. in diameter, which had sharply depressed firm edges with an irregular jagged contour. The center of the ulcer led into a tubular structure situated

^{*} Submitted for publication March 25, 1957.

^{••} Institute of Pathology, Rhode Island Hospital, Providence, Rhode Island. The authors are indebted to Dr. A. H. Eckstein for permission to report the case.

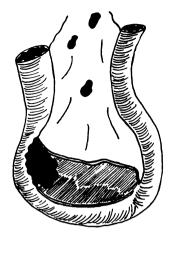


Fig. 1. Diagram of a segment of small intestine containing a tubular duplication. There is a carcinoma arising at the left end of the duplication. The mesenteric lymph nodes are replaced by metastatic tumor.

within the leaves of the mesentery and lying parallel to the long axis of the small bowel along its mesenteric border for a length of 15 cm. This tubular structure terminated in a circular opening, .8 cm. in diameter, thus re-establishing communication with the normal lumen of the small intestine. Apart from the ulcerated mass at one orifice the lining of the tubular structure was thick and velvety, resembling that of stomach mucosa. The remainder of the wall showed well-developed muscular coats similar to stomach or intestinal wall. It appeared to be fused with the adjacent small intestine along one border. Cross sections through the ulcerated mass showed yellowish white, firm, slightly gelatinous tissue with areas of necrosis extending throughout the wall into the adjacent mesentery. It appeared grossly as if the tumor arose from the duplication and extended slightly into the ileum at one stoma. mass was irregular in outline and measured 5 cm. in diameter. The ulcerated area measured 2 cm. The adjacent mesentery

contained many lymph nodes, which were enlarged and firm, measuring on the average 2 cm. in diameter. A segment of attached sigmoid colon showed a few small, firm serosal nodules. Its mucosa and wall were not remarkable.

Histologic examination of the duplication showed it to be lined by typical gastric mucosa with fundic and pyloric glands, as well as parietal acid-secreting cells (Fig. 2). Both muscular coats were present. Sections from the ulcerated tumor mass showed a margin of gastric-type mucosa with focal atypicality bordering an ulceration which was partly composed of tumor and partly of necrotic debris with acute and chronic inflammatory cells resembling a peptic ulcer (Fig. 3). The tumor was composed of small nests and columns of undifferentiated cells invading a dense fibrous tissue stroma. The tumor cells rarely formed abortive tubular or gland-like structures. In many in-

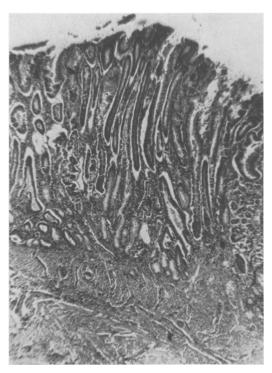


Fig. 2. Duplication. The intact lining consists of gastric mucosa. (Hematoxylin-Eosin × 20.)

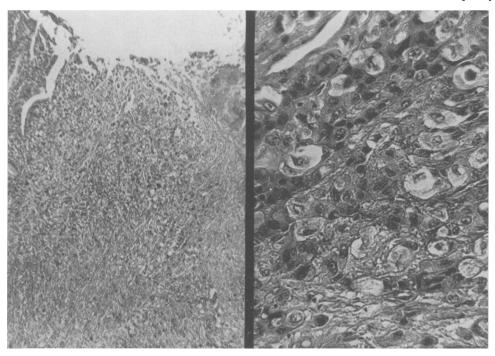


Fig. 3. Duplication. There is tumor invading its wall. On the left there are remnants of intact mucosa and on the extreme right there is "peptic" ulceration. (Hematoxylin-Eosin \times 120.)

Fig. 4. Duplication. This is a high-power view of the carcinoma. It consists of poorly differentiated tumor cells, many of which produce mucin. (Hematoxylin-Eosin \times 800.)

stances, distinct mucin production (signetring cells) was noted in the cytoplasm, which was pale-blue and vacuolated (Fig. 4). There were occasional atypical mitoses. The appearance was that of an infiltrating poorly differentiated and mucus-secreting adenocarcinoma consistent with an origin from gastric mucosa. Other sections showed invasion of the ileum by the above-described mass. The remainder of the sections of uninvolved ileum and attached sigmoid colon were not remarkable. Sections from the mesenteric lymph nodes contained metastatic carcinoma.

Discussion

Duplications of the alimentary tract occur most commonly in the ileum according to a review by Lo Presti, Kaufman, and Hawfield,³ who found the ileum involved in 77 per cent of 150 cases. The term has been extended to include such conditions. previously known as enteric cysts, giant diverticula, and enterogenous cysts. The duplicated portion may be either a spherical or an elongated tubular structure. It may communicate with the main bowel lumen at one or both ends, or there may be no demonstrable communication. Duplications in contrast to diverticula are situated on the mesenteric border within the leaves of the mesentery.1 The blood supply in most cases is derived from that of the adjacent intestine. The lining of the duplication may be identical with the neighboring bowel mucosa; or else it may imitate any other segment of the alimentary or respiratory tract.6 Ectopic gastric mucosa is very common and the finding of true gastric juice, containing enzymes and free hydrochloric acid has been reported in duplications of the ileum.7

No report has been encountered in the American or foreign literature describing a carcinoma originating in the duplication of the small intestine. A case of carcinoma arising from a duplication of the stomach has been described recently by Mayo, Mc-Kee and Anderson.4 Malignant neoplasms have been known to arise in Meckel's diverticula, which, however, differ from intestinal duplications both embryologically and morphologically.⁵ Lookwood,² in 1882, described a duplication of the large bowel and states that one end of the duplicate bowel was lost "in a mass of malignant disease." However, no histologic confirmation of tumor is mentioned in the article.

The case observed in this hospital has many features in common with previously recorded instances of duplication, such as its site in the small intestine, and its lining by ectopic gastric mucosa. It occurred in a middle-aged man and manifested itself clinically by symptoms of obstruction. The occurrence of a carcinoma from ectopic gastric mucosa with its striking histological resemblance to ordinary gastric neoplasm is an unusual feature and has apparently not been reported previously.

Summary

A case is described of tubular duplication of the small intestine complicated by carcinoma, which occurred in an adult male. The carcinoma appeared to have arisen in ectopic gastric mucosa lining the duplication.

The relevant literature is reviewed.

References

- Bremer, J. L.: Diverticula and Duplications of the Intestinal Tract. Arch. Path., 38:132, 1944.
- Lockwood, C. B.: On Abnormalities of the Caecum and Colon with Reference to Development. Brit. Med. J., Sept. 23, 1882.
- Lo Presti, J. M., P. Kaufman and H. H. Hawfield: Duplication of the Alimentary Tract. Clin. Proc. Children's Hospital, Washington, 6:357, 1950.
- Mayo, H. W., Jr., E. E. McKee and R. M. Anderson: Carcinoma Arising in Reduplication of the Stomach (Gastrogenous Cyst): A Case Report. Ann. Surg., 141:550, 1955.
- Moore, R. M. and W. H. Parsons: Carcinoma of Meckel's Diverticulum. Arch. Surg., 67: 939, 1953.
- Nolan, J. J. and J. G. Lee: Duplication of the Alimentary Tract in Adults with a Report of Three Cases. Ann. Surg., 137:342, 1953.
- Ripstein, C. B.: Duplication of the Small Intestine. Am. J. Surg., 78:847, 1949.

Editorial . . .

Secondary Operations and the Biliary Tract

EACH YEAR for the past decade there has been an increase in the number of patients who previously had had an operation upon the biliary tract admitted to The New York Hospital for study and evaluation. Almost all of them gave a history of having had a cholecystectomy. In general it may be said that following operation there was either a persistence or a recurrence of symptoms within a few weeks or months. A small proportion developed new complaints and a few were admitted within a matter of days or weeks because of jaundice or a draining biliary sinus. After critical consideration of

the history, physical findings and information derived from laboratory examinations together with the clinical course it was concluded that another operation was indicated for the majority. At the second operation, with few exceptions, findings were demonstrated which in view of the subsequent clinical course were considered to be the cause of the patient's symptoms. The conditions encountered are listed under three headings. First those within the biliary tract: 1) Common duct calculi. 2) Remnants of the gall bladder and cystic duct containing calculi. 3) Distortion of,