Gastroduodenal fistula in the Zollinger-Ellison syndrome

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Summary

A case of Zollinger-Ellison syndrome complicated by a gastroduodenal fistula is described in a 42-year-old man. So far as the authors know, this is the first report of this complication in gastrinoma.

Introduction

Gastroduodenal fistula is an unusual complication of peptic ulceration. There have, however, been a number of recent case reports referring to this problem, sometimes termed 'the double pylorus' (Gould, 1961; Bennike and Hegedüs, 1976; Hunt, Day and Jewell, 1978; Rappoport, 1978). These reports may reflect the greatly increased use of diagnostic upper gastro-intestinal endoscopy.

This complication in a 42-year-old man with gastric and duodenal ulceration who, on further investigation, proved to have the Zollinger-Ellison syndrome is now described.

Case report

A 42-year-old man, of West Indian origin, presented with a one-month history of continuous, severe, epigastric pain which radiated to the back. His symptoms were worse after food and only partially eased by milk and antacids. His appetite had decreased and his weight had fallen by 7 kg. There was no history of vomiting or diarrhoea and no relevant family history. He admitted drinking up to one half bottle of sherry/day and smoking 15 cigarettes daily. Examination was unremarkable apart from epigastric tenderness.

Investigations

Barium meal examination revealed a large volume of food debris in the stomach and an enormous lesser curve gastric ulcer. A fistulous tract connected the gastric ulcer with an ulcer in a deformed duodenal cap (Fig. 1). The pylorus was narrowed and deformed.

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Haemoglobin concentration was 12.8 g/dl, WBC $7.5 \times 10^9/l$, MCV 100 fl, ESR 59 mm/hr, fasting plasma gastrin level was 288 pmol/l (normal range <30 pmol/l). Serum iron, total iron binding capacity, serum vitamin B₁₂, red cell folate, blood urea, creatinine and electrolytes, liver function, calcium and phosphate levels were all normal.

A secretin infusion test (Thompson *et al.*, 1972), revealed a transient sharp rise in plasma gastrin, characteristic of the Zollinger-Ellison syndrome. A selective coeliac axis arteriogram was performed and a slight blush was demonstrated adjacent to the pancreaticoduodenal arcade, suggestive of a tumour in the head of the pancreas.

Treatment with cimetidine 200 mg thrice daily and 400 mg at night was commenced with symptomatic relief, but in view of the diagnosis of gastrinoma, laparotomy was performed. The findings were of a lesser curve gastric ulcer with a fistulous tract connecting its base with a superior duodenal ulcer. A small tumour was present in the head of the pancreas. Total gastrectomy was performed, with formation of a Roux-en-Y anastomosis. It was not possible to remove the tumour.

Postoperative recovery was uneventful and histology confirmed the benign nature of the ulcers.

Discussion

Gastroduodenal fistula may occur in one of 2 positions (Gould, 1961). The more commonly encountered fistula joins the lesser curvature in its middle or lower third to the first part of the duodenum. Less commonly, the fistula is from the posterior gastric wall to the third or fourth part of the duodenum. Rokitsky (1861) suggested that gastroduodenal fistula was caused by 2 ulcers, one in the stomach near the pylorus, and a second adjacent in the duodenum, eroding towards each other. This would seem a likely mechanism in the present case.

Since the original description of the Zollinger-Ellison syndrome (Zollinger and Ellison, 1955) there

FIG. 1. Barium meal showing a fistulous tract connecting the base of a gastric ulcer with a duodenal ulcer, and a narrowed pylorus.

have been many reports describing the radiological changes in this condition (Amberg *et al.*, 1964) including evidence of increased secretions, mucosal oedema and hyperplasia, dilatation of the stomach and duodenum, and jejunal hypermotility in addition to peptic ulceration.

Total gastrectomy with removal of the primary tumour, where possible, has long been the accepted treatment for the Zollinger–Ellison syndrome. Recently, however, it has been suggested that long-term treatment with cimetidine might provide an alternative to total gastrectomy (McCarthy, 1978). Such an approach would obviate the need for operation in patients with proved metastases, when conventional chemotherapy can be used in conjunction with cimetidine and, in patients without metastases, permit more radical pancreatic surgery to remove the gastrinoma. In the present patient, however, it was felt unlikely that he would comply with long-term cimetidine therapy and in view of the unusual peptic ulcer complication, surgery was considered appropriate.

With the increasing availability of serum gastrin assays, Zollinger–Ellison syndrome is being recognized at a much earlier stage (Thompson *et al.*, 1975). The diagnosis should be considered not only in patients with recurrent or multiple peptic ulcers, but also in the presence of unusual ulcer complications, such as gastroduodenal fistula.

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