Medical Memoranda

Recurrent Benign Ileal Ulcer Occurring with the Coeliac Syndrome

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Primary benign ulcers of the small intestine occurring beyond the duodenum are rare. They were first described by Baillie in 1805, but most of the older reports can be discarded, as they were not well documented and may have been due to regional enteritis or the Zollinger–Ellison syndrome, conditions not recognized at that time. Recently the disease has become more common (Jordan, 1968), and this may well be due to enteric-coated potassium tablets (Baker *et al.*, 1964 ; Räf, 1967). This report describes the case of a man with coeliac syndrome and recurrent benign ulceration of the ileum.

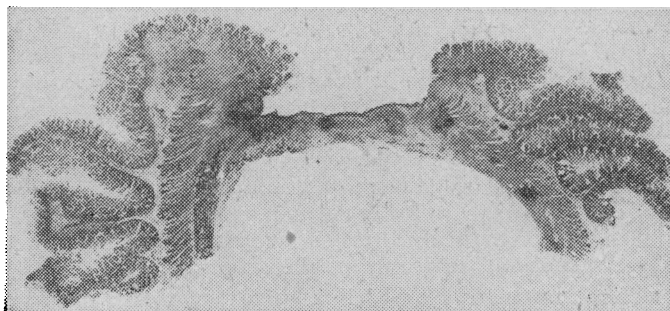
Case Report

In October 1963 a man aged 40 was admitted to hospital as an abdominal emergency, suspected of having a perforated peptic ulcer. Examination showed signs of peritonitis, and an abdominal *x*-ray investigation supported the diagnosis of a perforated abdominal viscus. At laparotomy, however, the perforation was shown to be in the ileum, 14 cm. from the ileocaecal valve. The ulcer was resected with an end-to-end anastomosis. Histology showed it to be a simple non-specific ulcer with no evidence of Crohn's disease, enteritis, or malignancy.

A previous history of chronic iron-deficiency anaemia and intermittent diarrhoea raised the possibility of a pre-existing malabsorptive state, and when fully recovered from his operation he was placed under investigation. He was found to have a faecal fat output of 26·4 g. over three days, a xylose excretion of 3·7 g., and a flat glucose-tolerance test. A jejunal biopsy showed almost complete atrophy of the villi. He was thought to have the coeliac syndrome, and was therefore discharged on a gluten-free diet.

He made a remarkable recovery, but unfortunately by 1965 had stopped his gluten-free diet for socioeconomic reasons and was therefore treated with prednisolone and vitamin supplements. Over the next three years he had only occasional attacks of diarrhoea ; otherwise he remained symptom-free. At no time was he taking enteric-coated tablets.

In January 1969 his diarrhoea increased, he became anorexic, lost weight, and had increasing abdominal pains and vomiting. Malignant change in the long-standing coeliac syndrome was thought to have occurred. Laparotomy (L. R. Celestin) showed an ulcer 30 cm. from the ileocaecal valve. This was resected, and the patient made an uneventful recovery, being discharged symptom-free on the fourteenth postoperative day.



Simple chronic ulcer of small intestine. (H. and E. ×3.)

The resected ileal segment was 6 cm. long with a chronic ulcer crater 1·5 by 0·7 cm. by 0·6 cm. deep. Histologically this was a simple chronic ulcer of the small intestine (see Fig.) which had penetrated the full thickness of the wall, its floor being formed by thickened peritoneum and subperitoneal connective tissue.

Comment

The coeliac syndrome is known to be associated with reticulosis (Gough *et al.*, 1962 ; Harris *et al.*, 1967 ; Brunt *et al.*, 1969), and duodenal ulceration has recently been recorded with the coeliac syndrome (Finlayson *et al.*, 1968). There are, however, only 16 recorded cases of small-bowel ulceration occurring with the coeliac syndrome (Bayless *et al.*, 1967), and in 12 of these death resulted from perforation. Usually small-intestinal ulcers present with obstruction (Wayte and Helwig, 1968) ; they may, however, occur with perforation or haemorrhage. The present case is unusual in that the ulcer recurred, and this has been recorded only once previously in over 200 cases. When there is no history of digestion of enteric-coated tablets the aetiology of these ulcers becomes obscure, yet in association with the coeliac syndrome it is possible that the prolonged alteration in epithelial cell turnover could alter mucosal healing, creating areas of regenerative failure and ulceration (Bayless *et al.*, 1967).

The main clinical pointers to malignant change in the coeliac syndrome are pain, profound loss of weight, and anorexia (Brunt *et al.*, 1969). An exploratory laparotomy may be necessary to determine the diagnosis. As the clinical features of benign ulcers, which may also be a complication of the coeliac syndrome, are similar to those of malignant change a laparotomy is mandatory, for the prognosis of benign ulcers after surgery is excellent.

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