FIVE DUODENAL ULCERS IN A SIBSHIP

BY

T. H. ALMOND, M.A., M.B., B.Chir.

Surgical Registrar, Victoria Hospital, Swindon, Wilts

A family tendency to peptic ulceration is not uncommon and much work has been carried out by investigators into the incidence of peptic ulceration occurring in first-degree relationships (Wright *et al.*, 1955). In one survey in North London, the incidence of peptic ulcer occurring in first-degree family relationships was found to be notably raised. The tendency, too, for the same type of ulcer—that is, gastric or duodenal—to occur in these relationships was also increased (Doll and Buch, 1950).

Recent studies have also been made of the incidence of peptic ulceration occurring in twins, although as yet the investigations have not been extensive enough to be of statistical significance (Harvald and Hauge, 1956). In the case of normal brother-and-sister relationship two aetiological factors will obviously be common—that is, genetical and environmental. The former factor will in many cases include an identical blood group, a factor which has already been shown to have some significance in the aetiology of peptic ulceration (Aird *et al.*, 1954). Similarly, the psychosomatic types are likely to be similar in the same family, and this also may be of considerable significance (Hurst, 1924). Such factors as diet, living conditions, smoking and drinking habits, etc., can be regarded as environmental.

An instance of a large family in which four brothers and one sister have all had duodenal ulcers occurred in this area. The mother has a history of many years' indigestion, but has not been in hospital. It is understood that the father is well. There is no blood relationship between father and mother.

History of the Cases

The mother, aged 68, had 15 children, whose ages now range from 20 to 50 years. There were six girls and nine boys. One of the sons died soon after birth, another was killed as a child in a road accident, and a third died on active service during the war, from wounds. Of the six remaining sons two, whose ages are 40 and 47, are alive and well, with no suggestion of peptic ulceration.

Apart from the sister who had a duodenal ulcer, all the others are alive and well, although one sister, aged 28, has periodic asthmatical attacks. None has any ulcer symptoms. Apart from the ulcers, the whole family seems to enjoy extremely good health.

The first member of the family to be treated surgically for a duodenal ulcer, Mr. A. Z., aged 38, had a perforated ulcer in 1944. He had at that time a history of only four months' indigestion. After a repair operation he was symptom-free for a further five years until he had a second perforation. A further repair operation was carried out, but he continued to have attacks of pain, and in 1951 a partial gastrectomy of the Polya type was carried out. At operation a large chronic duodenal ulcer was found. His post-operative course was satisfactory and he has remained well since.

The second member of the family to come under our care was the youngest brother, Mr. B. Z., aged 25. He was admitted to hospital in March, 1958, as an emergency case with severe abdominal pain in the left hypochondrium. He gave a history of several attacks of this pain during the previous six months, none of which had been so severe as the attack which brought him into hospital. The symptoms and signs on admission were not suggestive enough to make

a specific diagnosis, and they subsided under conservative treatment. Subsequently we carried out a barium-meal x-ray examination which showed considerable delay in stomach-emptying and a strong suggestion of a duodenal ulcer. Shortly after the x-ray examination the patient had a severe haematemesis which necessitated a blood transfusion. Three days later he had a further haematemesis. A Polya partial gastrectomy was then carried out and a large duodenal ulcer was found. The patient made a full recovery and has remained well.

The third member of the family with an ulcer, Miss C. Z., aged 28, came under our care a month later. She had a history of epigastric pain extending back for the previous six years, and had been treated medically in hospital on two occasions. In 1957 a barium-meal x-ray examination had shown the presence of an ulcer crater on the posterior wall of the duodenum and, as her condition had not improved for several months, it was decided that partial gastrectomy would be advisable. At operation—a Roux en Y partial gastrectomy—an ulcer was found on the posterior wall of the duodenum. She has remained well since this operation was carried out.

In June, 1958, the eldest of the brothers, Mr. D. Z., aged 49, who gave a history of five years' indigestion, was operated upon. A barium-meal x-ray examination in May had shown some degree of pyloric stenosis and the patient was anxious for surgical treatment. At operation two ulcers were, in fact, found—a large gastric ulcer on the posterior wall of the stomach and a small duodenal ulcer near to the pylorus. A high Polya partial gastrectomy was therefore carried out. His post-operative course was satisfactory and he has remained well.

Yet another brother, Mr. E. Z., aged 33, had been treated medically for indigestion for the previous 10 years. Two years ago his symptoms became more severe and a barium-meal x-ray examination showed the presence of a duodenal ulcer. He continued, however, with medical treatment, but a repeat x-ray examination in September, 1958, showed no improvement. In January, 1959, he was sent to the surgical out-patient department. He was becoming tired of continuous medical treatment and, as his symptoms appeared to be getting worse, it was felt that he too would benefit from gastrectomy. Accordingly he was operated on early in February and a large chronic duodenal ulcer was found. A partial gastrectomy of the Polya type was carried out. It is too soon to assess the results, but his immediate post-operative condition was very satisfactory.

Comment

Intensive efforts have been made to find common factors amongst the family that might be of significance regarding the aetiology of the ulcers. All the affected members had the same blood group, which was A-positive. At some time or other all have been engaged in driving occupations, with some irregularity in their meal-times. One member, D. Z., said they were brought up as a "frying-pan" family, but this was not confirmed by the others. None were heavy smokers, although all did smoke, and their drinking habits were all moderate. None gave the impression of being the hypersthenic, anxious, emotional type suggested by Hurst as being more ulcer-prone. There was nothing of significance in their previous illnesses.

It is interesting to note that with the exception of Mr. A. Z., all the other siblings had their operations carried out within a period of 12 months. Miss C. Z. and Mr. D. Z. had a similar length of history, but B. Z.'s history dates only a few months. However, he was the first of the family to have partial gastrectomy. One is tempted to conclude that the good results obtained from this operation helped to persuade his brothers and sister to have surgical treatment as well.

Another interesting point is the ratio of four males to one female having a duodenal ulcer. Numerous investigations have shown that the sex incidence shows a far greater percentage of duodenal ulcers occurring in the male than in the female, the average being in the region of 80% male.

Summary

A family is described in which duodenal ulcers occurred in several members. Five of the family—four brothers and one sister—had partial gastrectomies. Apart from a direct family relationship, no other common factor has been found which might be significant in the aetiology.

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Medical Memoranda

Carotenaemia Occurring in a Case of Anorexia Nervosa

Carotenaemia occurs when excessive amounts of carotene are ingested, in diabetes mellitus, in hyperlipaemic conditions, and in myxoedema; and Cohen (1958) described a "unique case" which he believed to be due to an inborn error of metabolism. Carotenaemia has not previously been reported to occur in anorexia nervosa, and the following case is described in order to draw attention to the possible association.

CASE HISTORY

An unmarried woman of 28 was seen in March, 1958, complaining of anorexia and amenorrhoea since the age of 16½. She had attended various hospitals in the past, with only temporary increase of weight each time. She weighed 7 stone (44.5 kg.) and was 5 ft. 6½ in. (169 cm.) tall.

On examination there was general emaciation, the breasts were small but developed, and the hair distribution was normal. There was no hypertrichosis. The skin was dry, thin, and inelastic, and acrocyanosis was pronounced. A faint yellow tinge of the skin was noticed, although the cyanosis made this less obvious than it would otherwise have been; it was especially marked over the palms and soles, which were bright orange, and to a less extent around the nose and in the axillary folds. The conjunctivae and mucous membranes were clear; the urine was a pale clear yellow and contained no urobilinogen or bile salts. Direct questioning revealed that she had first noticed the yellowness in February, since when it had fluctuated in intensity but never completely gone. The skin had not itched and she had not bothered about it. For the past 10 years she had eaten large amounts of fruit and cooked green vegetables (but never raw carrots), and there had been no recent change in this habit. She had been mildly constipated all her life, but took no aperients or liquid paraffin. Carotenaemia was diagnosed, and was confirmed by finding a serum carotene level of 240 µg./100 ml. (normal 50–120 µg.). The serum vitamin-A concentration, 182 I.U./100 ml. (normal 80–130), was also higher than normal.

There was no evidence of diabetes, a glucose-tolerance test was normal, and the blood cholesterol was 200 mg./100 ml. Clinically she was euthyroid. The B.M.R. was -1%, and the radioactive iodine uptake and excretion were within normal limits. Liver-function tests, plasma protein values, urine, and blood showed no abnormality. She was treated with strict bed rest, green vegetables were omitted from her high-calorie diet, and fresh fruit was restricted. The yellow pigmentation, although fluctuating, remained; and on discharge, after eight weeks as an in-patient, she weighed 8½ stone (54 kg.), the serum carotene was 180 µg./100 ml., and the vitamin A concentration 140 I.U./100 ml.

COMMENT

Carotene is present in variable amounts in many plant materials and in butter and egg yolk (Harris, 1955). It cannot be synthesized by animals and is utilized by them in the formation of vitamin A. This conversion occurs mainly in the intestinal mucosa and to a less extent in the liver, and in some species there is evidence that the absorption of carotene varies directly with thyroid activity (Wright, 1952). Cohen (1958) states that 4–8 lb. (1.8–3.6 kg.) of raw carrots must be eaten daily for some months before carotenaemia appears clinically and that the pigmentation disappears in two to six weeks when the diet is corrected. It seems unlikely, therefore, that the carotenaemia in this case is due to excessive intake, especially in view of the fact that her dietary habits have not altered recently. Pigmentation, as in other states of cachexia, has been described in anorexia nervosa (Berkman, 1930), but this is a brownish colour and obviously very different from the colour and distribution of carotenaemia. Yellow pigmentation of the nails was described in starving Dutch patients in 1944–5 (*Malnutrition and Starvation in Western Netherlands*, 1948), but the skin was said to be sallow, with dun-brown pigmentation. Metabolism is often depressed in anorexia nervosa, although it was not in this patient, but myxoedema does not occur and the serum cholesterol is usually normal.

Cohen described a case of carotenaemia, for which no cause could be found, in a woman of 38 who had had a yellow tinge to her skin for several years, most pronounced on the palms and soles but also affecting the lips and buccal mucosa. In this case the serum carotene was raised and there was vitamin-A deficiency. In addition the patient had lost weight. The case reported here differs in that the serum vitamin-A concentration is high. But large amounts of vitamin A, sufficient for at least six months' deprivation, are normally stored in the liver, and since the carotenaemia in this case is of only three months' duration, while Cohen's patient had had carotenaemia for several years, this may explain the difference. Alternatively, the abnormality may involve a different process. In any case, the possibility that this abnormality is associated with weight loss is interesting, and seems worthy of further investigation.

P. J. DALLY, M.B., M.R.C.P., D.P.M.,
Research Registrar, Department of Psychological
Medicine, St. Thomas's Hospital, London.

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