A CHRONIC CICATRIZING LESION OF THE STOMACH — PROBABLY SYPHILITIC IN ORIGIN

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Thirty-three years ago Rutherford Morison[1] pointed out that the chronic inflammatory swellings produced by syphilis and tuberculosis are not infrequently mistaken for malignant disease. He stated: "It has often occurred to me in palliative operations for 'hopeless cancer', when they have been successful beyond the most optimistic expectations, that other surgeons must have had similar experiences. Gastro-enterostomy for 'hopeless cance rof the pylorus', entero-anastomosis for 'inoperable cancer' of the small gut, and colostomy for 'irremovable malignant obstruction of the colon', have all been followed by entire relief of the obstruction for which the operation was performed, and at the end of 10-20 years, a few of these patients are alive and well. The probable explanation is that tubercle or syphilis . . . misled us because some have developed other tuberculous lesions and others have been cured by a course of iodide and mercury." The following case is of considerable interest, especially because of the difficulty in establishing a diagnosis on the clinical, radiological and histological evidence.

CASE REPORT

An 11-year-old Coloured female was admitted to the Red Cross War Memorial Children's Hospital, Rondebosch, Cape, on 4 March 1957. She complained of a steady loss of weight associated with anorexia for one year before her admission to hospital. For 3 weeks before admission she had been vomiting food recently eaten and had experienced attacks of postprandial epigastric pain which was relieved by induced vomiting. There were no other relevant features on special interrogation of the patient.

The patient is an only child. Apart from 2 abortions 5 and 8 years after the birth of the patient, her mother had had no other pregnancies or abortions.

On examination the patient weighed 35¼ lb. (The normal weight for an 11-year-old child is 88·4 lb. S.D. 17·4 lb.) There was marked loss of subcutaneous fat and muscle substance. The patient had a dry skin with associated follicular hyperkeratosis over the extensor aspects of the upper arms and legs. In the left groin there was a scar of a healed apocrine gland infection. There were 2 perforations (Fig. 1) in the soft palate communicating with the nasopharynx. No abdominal mass could be palpated. The hymen was intact. There were no other positive findings on general examination.

Fig. 1. Perforations in the soft palate.

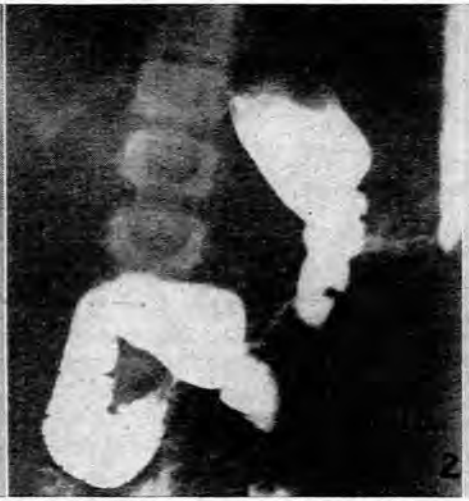


Fig. 2. A barium-meal examination showing marked narrowing of a segment of the pyloric antrum.

Fig. 3. A specimen of the stomach and omentum.

Special Investigations

Bedside and side-room. The urinalysis was normal. The haemoglobin was 13·5 g. per 100 ml. and the sedimentation rate 87 mm. in 1 hour (Westergren). The Mantoux test ($\frac{1}{1000}$ old tuberculin) was negative.

Radiological (Dr. E. van der Burg). A barium-meal examination was carried out on 7 March 1957 (Fig. 2). There is marked narrowing of a considerable segment of the pyloric antrum. The narrowing appears to have occurred at the expense of both the greater and lesser curvature aspects. Although throughout this examination the fundus of the stomach appeared to be distensible and not rigid, the body of the stomach on the other hand showed a constant serated appearance on the greater curvature aspect. On the lesser curvature of the stomach at about the region of incisura there was a small rather flat projection suggestive of ulceration.

Examination of the skull and long bones failed to show any evidence of bone or joint abnormality.

Bacteriological. Kirschner culture for *M. tuberculosis* in the gastric lavage was negative. Blood Wassermann and Kahn tests were both positive. The mother's blood Wassermann and Kahn tests were both positive; the father's blood Wassermann and Berger tests were both positive, while the Kahn test yielded a doubtful positive result.

Biochemical. The blood urea was 60 mg. per 100 ml.; serum albumin 3·1 g. per 100 ml.; serum globulin 4·4 g. per 100 ml.; zinc turbidity 40·1 units; thymol turbidity 12·4 units; thymol flocculation +++; total bilirubin 0·790 mg. per 100 ml.

Course before Operation

From the day of admission the patient was put on a high-calorie, low-residue diet but it was found necessary to supplement her oral intake of fluids with intravenous dextrose and serum. During this time the patient would vomit 150-250 ml. of fluid on certain days, but sometimes she would go for 2 days without vomiting.

Treatment with procaine penicillin 600,000 units daily was commenced on 11 March 1957. Six days after the administration of penicillin therapy, the patient vomited 765 ml. of fluid. The next day she vomited 720 ml. whereas she had taken only 480 ml. of fluid by mouth. As the patient's condition appeared to be deteriorating in spite of intravenous therapy, it was decided to perform a laparotomy the next day.

Operation and Subsequent Course

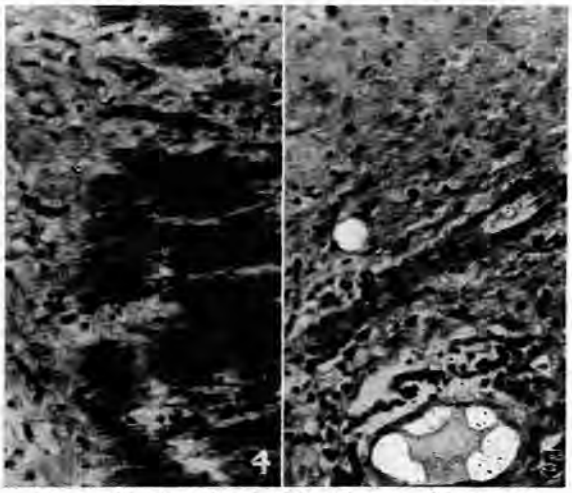
On 19 March 1957 a laparotomy (Prof. J. H. Louw) was performed. The stomach was small, with considerable thickening of the walls throughout its whole length. This thickening commenced abruptly at the oesophago-gastric junction and terminated likewise at the pylorus. The walls were firm and smooth and encroached considerably on the lumen, especially of the pyloric antrum.

There were several enlarged glands along the greater curvature and also in the left gastric and subpyloric groups. These were firm and appeared hyperplastic. Further exploration showed no other abnormalities.

The nature of the pathology was not clear. In some respects it resembled a diffuse infiltrating carcinoma or localized lymphoma. In others it resembled a chronic inflammatory process. On account of the possibility of malignancy a total gastrectomy was performed. Continuity was re-established by an antecolic oesophago-jejunal anastomosis.

Pathology Report (Dr. D. McKenzie). 'The specimen (Fig. 3) consists of a stomach and omentum. The stomach measures 7 cm. on the lesser curvature. There are small glands along the greater curvature up to 0·5 cm. in diameter. The pyloric canal just admits a small probe. The muscle in this area is hypertrophied and is glistening in appearance and firm in consistency. The submucosa and mucosa in this area do not appear to be affected. The wall of the stomach appears thickened throughout its whole length but the glistening appearance of the muscle terminates at the pylorus distally and approximately in the region of the mid-gastric sphincter proximally. The cut surface of the lymph nodes show no obvious abnormality.

Histology (Figs. 4 and 5) shows dense fibrosis of the submucosa; the process extends to the proximal end of the stomach but is slightly less dense in this area. Fibrosis extends into the muscle



Figs. 4 *and* 5. Dense fibrosis of the submucosa of the stomach.

layer and there is quite considerable replacement of muscle by fibrous tissue. Distally the fibrotic process ceases abruptly at the pylorus and the duodenum is normal. The gastric mucosa is lost in several areas and where it is present just adjacent to the epithelium there is considerable vascularization of the tissue. There is patchy filtration of chronic inflammatory cells maximal just below the mucosa—these cells are mainly lymphocytes and occasional plasma cells. There are no endarteritic changes in the vessels. Spirochetes could not be demonstrated in any of the sections stained by the Warthan-Starry method. The lymph nodes are normal. This is a chronic cicatrizing gastric fibrosis which histologically gives no indication of the aetiology of the process. Any attempt at specific diagnosis would have to rest on the sum total of other evidence.'

The day after operation, the patient showed signs of congestive cardiac failure. She was successfully treated with intravenous digoxin. The remainder of the patient's postoperative course was uneventful. Bowel sounds were heard on the 2nd postoperative day and intravenous fluids were discontinued after the 4th postoperative day. The stitches in the abdominal wound were removed on the 11th postoperative day. Penicillin, together with streptomycin 0·25 g. twice a day, was continued until the 13th postoperative day. When the patient was discharged on the 36th postoperative day she weighed 52 lb.

Second Admission

On 26 June 1958 the patient was readmitted for examination. During the preceding 14 months she had been in good health. She was able to eat 3 meals a day without discomfort, and had not had more than 3 bowel actions a day. The stools were well formed. She had been attending school regularly and her scholastic achievements were equivalent to those of a White child of the same age.

She weighed 73 lb. (normal weight for a 12-year-old child is 100·4 lb. S.D. 18·8 lb.)—a weight increase of 37 lb. since her first admission. No abnormal features apart from the perforation in the soft palate and healed apocrine-gland infection, which were present on her first admission, were noted.

The fact that this child has been virtually free of post-gastrectomy symptoms merits comment. The ability of a few patients subjected to total gastrectomy to maintain normal body weight, is thought to be due to true reservoir function developing in the upper small intestine.[9]

Special Investigations

Bedside and side-room. The haemoglobin was 10·3 g. per 100 ml.; the haematocrit was 33; the sedimentation rate was

15 mm. in 1 hour (Westergren). On examination of a peripheral blood smear the erythrocytes were normochromic and normocytic.

Radiological. A barium-meal examination performed on this admission showed no hold up of barium at the lower end of the oesophagus. The afferent and efferent loops of the jejunum were normal.

Biochemical. Blood urea 18·5 mg. per 100 ml.; serum albumin 4·5 g. per 100 ml.; serum globulin 2·56 g. per 100 ml.; zinc turbidity 27·2; thymol turbidity 5·53; thymol flocculation 0.

Bacteriological. The blood Wassermann and Kahn tests were still both positive.

DISCUSSION

In this patient the presence of a chronic cicatrizing fibrotic lesion involving the stomach together with a positive Wassermann reaction suggests a diagnosis of tertiary syphilis of the stomach. The age of the patient together with positive serological evidence of syphilis in the parents is in favour of congenital infection.

Syphilis of the stomach is unquestionably a rare lesion. Estimates of incidence of gastric syphilis are very difficult to make and it is often impossible to judge the validity of the reported cases.[2] Congenital syphilis of the stomach is rarer still. In a recent review of world literature, Mendl *et al.*[2] were able to find only 10 cases.

In 1918, Caster and Mathias[3] held the view that every case of gastric ulcer was due to either acquired or congenital syphilis. Four years later, Crookshank[4] stated that in the diagnosis of syphilis of the stomach one implied the association of (1) syphilitic infection, (2) a gastric syndrome, and (3) a specific (syphilitic) lesion of the stomach. The diagnosis was unjustifiable unless (1) and (3) were joined to (2).

In the past, attempts have been made at diagnosing gastric syphilis on the basis of certain clinical findings.[5] Today few clinicians, if any, would venture such a diagnosis on the basis of suggestive clinical findings even if the Wassermann reaction is positive, which it is in the vast majority of reported cases.

If a patient who is under 45 years of age presents with symptoms suggestive of a gastric carcinoma, but the symptoms are of considerable duration and the Wassermann reaction is positive, then a diagnosis of gastric syphilis should be considered. Achlorhydria is invariably present in gastric syphilis but the presence of achlorhydria in the adult is of no value in the differential diagnosis of syphilis and carcinoma of the stomach. In children, gastric anacidity is rare[6] and its presence is therefore of more value. The radiographic findings usually suggest a malignant lesion of the stomach.

In a suspected case of gastric syphilis, the therapeutic response to penicillin therapy is of diagnostic value. Each case must be judged on its own merits before embarking on a therapeutic test. It would be wrong to treat a positive Wassermann reaction while the patient is subject to a gastric carcinoma which requires urgent surgery. Even a favourable therapeutic response to antisyphilitic therapy may not in itself be regarded as proof of the syphilitic nature of the lesion. Rafsky *et al.*[7] have pointed out that non-syphilitic lesions may not heal until the associated syphilis is treated.

Harris and Youmans[8] state that when the syphilitic lesion is located near the pylorus, obstruction may be worse after antisyphilitic treatment and may require surgical intervention. This is a possible explanation of the increased frequency of vomiting in our patient 6 days after commencement of penicillin therapy.

The histological lesion in gastric syphilis is not pathognomonic and the only microscopic proof is the demonstration of spirochaetes which have the morphological characteristics of *T. pallida*, in the lesion.[10] If one adopts the latter criterion syphilis of the stomach has been histologically proved on only 2 occasions.[11, 12]

Mendl *et al.*[2] have given a classification in 4 stages of the radiological appearances in gastric syphilis. Their 4 stages correspond to a pathological classification described by Davicovic,[13] who describes 4 different evolutional stages of gastric syphilis. These stages are: a local plaque-like submucous infiltrate; an overgrowth of this infiltrate to produce a so-called syphiloma; a local annular fibrotic lesion; and, finally, a diffuse leather-bottle infiltration of the stomach.

While the classification described by Mendl *et al.*[2] is of value in reminding us that syphilis of the stomach may mimic other gastric lesions radiologically, none of the radiological appearances described is diagnostic of syphilis. This is understandable because classification of syphilis of the stomach is based on pathological criteria in which the stages described may be produced by gastric lesions other than syphilis.

We feel that any attempt which is made at describing syphilis of the stomach in terms of stages must inevitably lead to the false conclusion that syphilis of the stomach exists as a morphological entity. Syphilitic catarrh and chronic syphilitic gastritis[14] are nebulous terms which belong to the past. Aird[14] states that these terms, together with terms such as syphilitic round ulcer, diffuse syphilitic, local syphilitic fibrosis, and local pyloric stenosis, describe gastric lesions which in most cases are not syphilitic at all.

Differential Diagnosis

In our patient a total gastrectomy was performed in the belief that the patient had a primary gastric lymphoma. In this condition pain, dyspepsia, anorexia and loss of weight are the main symptoms.[15] A mass may be palpable.[16, 17] Achlorhydria is not a feature of gastric lymphoma.[18]

Since 1949, when Ross[19] first described a case of Crohn's disease of the stomach, there have been 5 further reports[20-24] of involvement of the stomach in Crohn's disease. The subject of non-specific granulomatous disease of the proximal gastro-intestinal tract has been reviewed by Zeifer.[25] He states that the positive pre-operative diagnosis of non-specific granulomatous disease of the proximal gastro-intestinal tract is virtually impossible, since it will depend on exclusion of diseases with granuloma-producing qualities. Despite the most painstaking efforts to exclude these processes, a precise diagnosis will require surgical exploration in most cases. It is pertinent to mention, however, that none of the cases reviewed by Zeifer demonstrated a positive Wassermann reaction. The fact that there is often involvement of the gastro-intestinal tract other than the stomach suggests a diagnosis of Crohn's disease. The histology of the stomach in our patient was of no assistance in distinguishing this condition from syphilis of the stomach.

Tuberculosis, Boeck's sarcoid and amyloidosis of the stomach may also produce a clinical picture similar to that found in our patient but these conditions were not seriously considered in the differential diagnosis.

We must conclude that the diagnosis of syphilis of the stomach is difficult. The diagnosis in our patient was made on collateral evidence.

In 1931 Eusterman[5] described 93 cases of syphilis of the stomach, 'the rarity of which had in the past been stubbornly maintained by most pathologists and by some clinicians and surgeons'. He made a plea for the more frequent recognition of syphilis of the stomach. Today, because of a marked reduction in the incidence of syphilis and the emphasis on accuracy in diagnosis of upper gastro-intestinal lesions, the pendulum has swung in a direction opposite to that hoped for by Eusterman.

SUMMARY

1. The clinical, radiological and histological features of a chronic cicatrizing fibrotic lesion of the stomach in an 11-year-old Coloured female are described.

2. Difficulties in diagnosis of syphilis of the stomach are discussed.

3. It is concluded that the lesion in the stomach is a manifestation of late congenital syphilis.

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