Summary

The chronic inflammatory changes of the skin known as erythema ab igne, due to prolonged sitting over a fire, are common in this country but rare in the United States.

Similar cutaneous changes due to the heat from the *kangri* bowl in Kashmir and the *kang* bed in China have been shown to produce cancers.

Four cases are described in which the patient has developed a squamous-cell carcinoma on an area of erythema ab igne, and one case in which there were premalignant changes. Two of the four patients had been treated for, and cured of, cancer of other organs some years previously; the other two showed the cutaneous changes known as poikiloderma atrophicum vasculare, sometimes found in association with malignant disease.

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PNEUMATOSIS CYSTOIDES INTESTINALIS

REPORT OF A CASE

BY

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[WITH SPECIAL PLATE]

Cases of gastro-intestinal pneumatosis have been reported in the literature from time to time. The condition was known to John Hunter, who described its occurrence in swine. There is a specimen in the Hunterian Collection of the Royal College of Surgeons: this was sent to John Hunter by Jenner and the gas from the cysts was analysed by Cavendish; a description of the specimen is to be found in Hunter's *Works* (Nitch, 1923). The first reference to the condition in man is in the catalogue of the Anatomical Museum of Amsterdam in 1737 (quoted by Lamont, 1929), where it describes "a portion of jejunum of a man showing a tumour which arose from wind." Bang described the condition in 1876 and Mayer in 1825, the latter calling it emphysema bullosa. Since then there have been many descriptions and much discussion in the literature as to the possible aetiology of these gaseous cysts in the abdomen. Jackson (1940) collected 171 cases from the literature and described one personal case. There have been over 17 further cases reported (Somerville, 1951).

The case now described is of interest because of the confusion caused by air cysts around the terminal ileum, which had become displaced and lay in a suprahepatic position under the right diaphragm.

Aetiology

The association of pneumatosis cystoides with pyloric stenosis is well known. Berglund (1939) states that in 60% of the cases described in the literature pyloric stenosis has been present. Lamont (1929) claims that 50% of cases are definitely associated with duodenal or gastric ulcer.

The aetiology was first fully reviewed by Nitch in his presidential address to the Section of Surgery of the Royal Society of Medicine (1923). He collected 85 cases from the literature, analysing the aetiological factors, and described two cases of his own.

Many theories have been put forward. Neoplastic, bacterial, and mechanical factors have been considered. Eveleth and Biester (1938) discovered that in hogs fed on polished rice cysts developed on the serosa of the intestine. This led many observers to suspect malnutrition as being the main causative factor in this disease. Schorr *et al.* (1951) drew attention to the fact that patients with this condition were usually undernourished, owing mainly to the long history of associated peptic ulceration and subsequent pyloric stenosis. If the pyloric stenosis is relieved the cysts tend to regress immediately, and have been observed to do so by Berglund (1939) both radiographically and by laparotomy. The disease affects infants, and Moore (1929) reported four cases of pneumatosis in infants who died of enteritis with ulceration. Botsford and Krakower (1938) also reported six cases occurring in children in association with diarrhoea or vomiting; no mucosal defects were found on necropsy in these cases. Morison (1952), in discussing the pathology of this condition in infancy, postulated that some of the gas might have been produced by micro-organisms invading the tissues before or after death.

Various authors have suggested that gas is driven into the lacteals through mucosal ulcers, and that the raised intra-abdominal pressure from repeated vomiting might lead to gaseous distension of lacteals.

Case Report

A man aged 51 was brought to St. Bartholomew's Hospital after having collapsed on the platform of a railway station. He complained of severe upper abdominal pain, worse on the right side. He was shocked, dehydrated, and vomiting blood. He gave a 20-year history of dyspepsia, having had a course of medical treatment in hospital 19 years ago for peptic ulcer. Four years ago he was operated on at another hospital for perforated gastric ulcer; the records show that no other abnormality was found in the abdominal cavity. Since this time he had developed two distinct types of pain, one coming on 1½ hours after meals and situated in the epigastrium, and the other in the right hypochondrium which radiated to the back and right shoulder and was not associated with meals. There was a weight loss of 4 stones (25.4 kg.) and he had been unemployed for four years (since his operation for perforation) on account of his disability. Recently repeated vomiting had developed and he had noticed melaena stools on several occasions. Since his operation he had suffered from very severe bronchitis, which had become worse in recent months.

Three days before admission he experienced sudden epigastric pain which persisted; 48 hours later the pain became worse and colicky. He vomited altered blood on four

occasions, and collapsed one hour before admission. He vomited a further litre of altered blood very soon after admission.

The patient was very thin and in a state of shock. Pulse rate, 85 per minute; blood pressure, 115/70 mm. Hg; respirations, 18 per minute; temperature, 97° F. (36.1° C.). The teeth were carious and there was gross gingivitis. The chest was barrel-shaped with poor movement but good air entry; breath sounds were vesicular with moist rales at the lung bases; heart sounds were normal. On abdominal examination a right upper paramedian scar was seen. There was visible peristalsis in the lower abdomen, but no distension. In both upper and lower right quadrants there was marked rigidity, with tenderness in the epigastrium and complete absence of liver dullness; a marked succussion splash could be elicited; bowel sounds were reduced. There was no abnormality on rectal examination.

A radiograph revealed the presence of "loculated" air under the right dome of the diaphragm (Special Plate, Fig. 1). A large portion of intestine was seen extending up the right lateral aspect of the abdomen to lie above the liver. At the time the significance of this subphrenic gas shadow was not appreciated, and the most probable diagnosis seemed to be pyloric obstruction with haematemesis and perforation of a peptic ulcer. A further litre of altered blood and food residue was removed by tube from the stomach. Laparotomy was then performed.

The abdomen was opened through a midline incision. There was no free fluid or gas in the peritoneal cavity. The stomach was grossly distended and occupied both left quadrants, extending into the pelvis (see Plate, Fig. 2, opaque area marked *b*). Above the liver, on the right side, there was a large cystic mass attached to the under surface of the diaphragm. The terminal ileum passed up over the liver into this cystic mass. There was no evidence of any intestinal obstruction. The mass was separated from the diaphragm and the liver, when it was seen to be composed of hundreds of gas-containing cysts surrounding the lower ileum for a distance of 45 cm. (Plate, Fig. 2). The mesentery of the affected part of the ileum was also involved, with cysts ranging from 0.5 cm. to 5 cm. in diameter. Both the caecum and large intestine were unaffected. Owing to the extensive involvement of the terminal ileum and mesentery by the air cysts and absence of involvement of any other part of the gut it was decided to resect the affected segment with its mesentery, leaving a 3-cm. stump at the ileo-caecal end and performing an end-to-end anastomosis. There was an ulcer mass with considerable oedema in the region of the pylorus, but no sign of recent perforation. Partial gastrectomy seemed inevitable owing to the presence of pyloric obstruction and haematemesis, and was performed with the Moynihan II method.

The patient made a good recovery. His general condition improved rapidly and he had no further bronchitis. X-ray examination of the chest showed areas of cystic emphysematous changes at both lung bases. He has now been able to return to light work and is free from pain for the first time for 25 years.

It seemed likely that the cystic loop of bowel above the liver might have become displaced during cleansing of the subphrenic pouches at the operation for closure of the perforated duodenal ulcer four years previously.

Pathological Report

The specimen consisted of approximately 75 cm. of ileum showing numerous thin-walled, translucent cysts varying from 2 mm. to 3 cm. in diameter distributed along its length. The cysts were most numerous about the central portion of the excised ileum, and here the affected gut was coiled upon itself and continuous loops and cysts were adherent to one another, forming an irregularly nodular mass. Less-affected lengths of ileum projected. The cysts, containing gas, were present beneath the mucosa (which was not ulcerated), in the

substance of the muscularis, and beneath the serosa, along both mesenteric and free borders. The main nodular mass was purple-red in colour and congested. The attached mesentery was oedematous and hyperaemic and packed with cysts, many of which were several centimetres from the bowel.

Sections of small intestine showed a number of empty cystic spaces in the submucosa, muscle coat, and particularly in the rather fibrosed subserosal layer. Some cysts had little lining endothelium, while others had a layer of cubical flattened endothelium. In yet others a number of large giant cells were incorporated in the lining endothelium. Sections of lymph nodes showed only sinus catarrh.

Sections of stomach revealed the presence of an active chronic peptic ulcer.

Discussion

Cystic pneumatosis may be found associated with any part of the intestine. It is commonest in the jejunum (Lamont, 1929), but a number of cases have been described where the cysts occurred in the distal ileum. Pybus (1934) reports such a case which resembles the one described above, though in his case cysts were also present in the falciform ligament of the liver. Jones (1948) describes pneumatosis occurring in the large gut in association with a chronic volvulus of the sigmoid colon. Cysts have also been seen at sigmoidoscopy, looking like small grapes, which when ruptured were found to contain gas. Sherwin and Messe (1952), reviewing the American literature, found reports of only 43 previous cases. They added a further case which bears a striking similarity to that now described. Their patient had pyloric obstruction due to peptic ulceration, and the affected loop of ileum was wedged between the dome of the diaphragm and the liver on the right; cyst formation extended to within 6 in. (15.2 cm.) of the ileocaecal junction.

One of the complications of this condition is pneumoperitoneum, due to rupture of the cysts, as described by Somerville (1951), Schorr *et al.* (1951), and Jones (1948). It may be the presenting clinical feature. Pneumomediastinum has also occurred.

It is known that these cysts can disappear spontaneously after relief of the causative factors, but remaining cysts may give rise to post-operative pneumoperitoneum and other complications (Berglund, 1939). The mass of cysts can cause intestinal obstruction due to pressure on the intestinal tract, as described by Thoma and Hennigar (1952). It has been suggested that subserous cysts are of a precancerous nature, although this association is not generally accepted.

If the cysts are localized to one area of the bowel and a laparotomy has been performed, it would seem correct to excise the area containing them. Any coexistent gastric, duodenal, or intestinal condition must be dealt with appropriately as the primary treatment. Excision avoids the possible complication of pneumoperitoneum and intestinal obstruction which may arise subsequently from progressive inflammatory change and fibrosis.

Summary

A case of ileal pneumatosis cystoides which developed in association with prolonged malnutrition due to chronic gastric ulcer is described. The presenting symptoms were due to haemorrhage from the ulcer. The presence of gas below the diaphragm led to a mistaken diagnosis of perforation of the ulcer. Partial gastrectomy and excision of affected ileum relieved the patient's symptoms so that he was able to return to full employment after four years' disability. It is suggested that the affected ileal loop had been displaced to the anterior subphrenic pouch at a previous operation; its fixation here and its subsequent exposure to continuous variations in pressure between liver and diaphragm may explain the localization of the pneumatosis.

There seems little doubt that violent changes of intra-abdominal pressure caused by vomiting have some connexion with the production of this condition. Vomiting so often occurs in association with chronic malnutrition that a nutritional factor may also be operative.

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Medical Memorandum

Use of Papaverine in Traumatic Arterial Spasm after Supracondylar Fracture of Humerus

Threatened ischaemia consequent upon fractures in the upper limb has always presented its difficulties, not only of recognition but also of treatment. The following case report illustrates some of the prevailing views held on the treatment of arterial spasm.

CASE REPORT

On March 20, 1954, a 6-year-old boy was brought to the casualty department with a history of having injured his left elbow.

On examination he was pale and in some pain but not obviously shocked. The left elbow was swollen, heavily contused, and deformed, suggesting a supracondylar fracture of the humerus. The proximal end of the humerus was palpable in the antecubital fossa and appeared to be immediately subcutaneous. The radial pulse was impalpable despite prolonged examination and its absence indicated some form of arterial damage. X-ray examination confirmed the diagnosis of a severe supracondylar fracture with marked posterior displacement and a less severe lateral displacement of the distal fragment.

One attempt at manipulation of the fracture was made in the casualty department in the hope that adequate reduction would restore the blood supply to the distal part of the limb (Griffiths, 1948). Not only was there a failure to restore the radial pulse, but reduction of the fracture was impossible, and it was clear that the proximal fragment of the humerus had buttonholed the soft tissues. As his fingers were developing a cyanotic tinge and fast becoming cool and swollen, the patient was prepared for transfer to the theatre, where both the fracture and the brachial artery could be explored.

At operation a medial longitudinal incision was made because of the severe soft-tissue swelling. The subcutaneous tissues were found to be severely contused and lacerated. The proximal end of the humerus was found to have buttonholed the brachialis muscle and lay just under the skin in the antecubital fossa, in direct medial relationship to the median nerve, which, however, showed no signs of damage.

Initially the brachial artery was difficult to identify by virtue of the extreme spasm for approximately 12 cm. of its

length, which made it resemble one of its own venae comites. When ultimately identified by the recognition of proximal normal pulsation, the vessel was lifted from its bed so far as was technically possible in a vessel so contracted. This procedure produced no obvious change in the condition of the artery, and without further delay a 5% solution of papaverine sulphate was applied superficially to the vessel wall. The response was dramatic. The brachial artery assumed its normal size within two to three minutes, with ample evidence of full and sustained arterial pulsation. The vessel wall appeared undamaged. Reduction of the fracture was not completely successful, for it was virtually transcondylar and very unstable.

The loosely sutured wound was drained and the arm immobilized in a plaster-of-Paris posterior slab and a lateral stirrup. At the conclusion of the operation stellate ganglion and brachial plexus blocks were performed. The radial pulse was now easily palpable at the wrist.

Post-operative treatment included elevation and cooling of the limb. The following morning the radial pulse remained easily palpable; the fingers were warm and the colour was normal. By the same evening the whole limb had begun to swell and the pulse was correspondingly weaker, although the fingers retained their warmth. All dressings were freed, and to ensure maximum vasodilatation of collateral vessels a further brachial plexus block was performed. The pulse none the less continued to be weak and intermittently palpable for the next three days, but at all times the hand remained warm. The view was held that the pulse was difficult to feel owing to the increasing tension created by the injury. Confirmation of this view was provided by inspection of the arm and elbow; for here, in association with the intense swelling, the skin showed the haemorrhagic vesication of impaired viability. Subsequently the wound broke down for this very reason and required skin grafting.

Although his convalescence was slow, he ultimately made satisfactory progress, and when seen six months after the injury there was no evidence of ischaemic changes to be found in the limb.

COMMENT

Griffiths (1948) has shown that, when conservative measures have failed, exploration must be carried out to expose the artery, and, provided there is no arterial damage, the vessel is liberated, mobilized, and, if necessary, a solution of procaine applied. At the conclusion of the operation he advocates sympathetic paravertebral block in the neck, presumably to eliminate spasm in the smaller arteries and in the arterioles.

It has since been shown experimentally by Kinmonth (1949) that smooth-muscle contraction gave rise to local segmental spasm in the large arteries of the body, in the maintenance of which nervous factors did not play a demonstrable part. Following these important findings, Kinmonth (1952) made known the results of directing his attention to paralysing smooth muscle itself by smooth-muscle-relaxing drugs. He found that the local application of papaverine sulphate to the walls of large arteries was the most effective in the volume of its response, in its speed, and in its sustained action.

This case demonstrates the immediate and dramatic effect of papaverine sulphate in relaxing arterial spasm when used locally. In injuries of this nature, where, following exposure of the brachial artery, simple spasm is found, the initial local application of papaverine would appear to be the treatment of choice.

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G. A. GRANT PETERKIN: ERYTHEMA AB IGNE

FIG. 1.—Case 4: Photomicrograph ($\times 68$) from nodular lesion on left leg.

FIG. 2.—Case 4: Photomicrograph ($\times 68$) from small nodule on right leg.

FIG. 3.—Case 4: Photomicrograph ($\times 15$) of section from large nodule on left leg, showing squamous-cell carcinoma.

FIG. 4.—Case 4: Photomicrograph ($\times 68$) of squamous-cell carcinoma.

J. D. GRIFFITHS AND D. F. ELLISON NASH: PNEUMATOSIS CYSTOIDES INTESTINALIS

FIG. 1.—Antero-posterior radiograph of abdomen. (a) Loculated gas beneath right dome of diaphragm. (b) Complete obscuring of central abdominal area by stomach grossly distended with blood clot.

FIG. 2.—Photograph of specimen of resected small intestine showing on left relatively collapsed segment with submucosal cysts, and on right (top) the opened proximal portion containing large intramural cysts. The intervening mass consisted of strictured bowel and contracted mesentery packed with large and small cysts.