LEIOMYOSARCOMA OF THE STOMACH: BRIEF REVIEW INCLUDING REPORT OF A CASE WITH MULTIPLE METASTASES.

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Leiomyosarcoma (or malignant leiomyoma) is one of the rarest tumours of the stomach. In 1946, when reporting four cases of their own, Schindler *et al.* were able to find only 94 other cases in the literature. The discovery of five clinically significant leiomyomata in a series of 5,183 examinations of stomach led Elliot and Wilson (1952) to conclude that non-epithelial tumours of stomach are not uncommon, but none of their cases appeared to be leiomyosarcoma. Of 60 cases of smooth muscle tumours of gastrointestinal tract and retroperitoneal tissues seen over a period of 26½ years by Golden and Stout (1941), 33 involved the stomach. Of these, however, 20 were incidental findings at autopsy, and 5 were incidental findings at operation; only 5 were malignant, and in only 2 were there metastases.

Golden and Stout conclude from their survey of the literature that metastases are found in 30 per cent of malignant cases, most often in liver, and rarely in lungs or peritoneum. They had not observed spread to lymph nodes, although this has occasionally been mentioned by previous authors. Multiple easily palpable metastases are uncommon: Fleming (1951) reported a case of leiomyosarcoma of the stomach with invasion of lymphatic glands, but there was no palpable abdominal mass. Greathead (1949) described a fatal case of leiomyosarcoma in which a large mass involved the posterior wall of stomach and the liver, but no deposits were found elsewhere.

Leiomyosarcoma has an importance greater than its infrequency would suggest, since its slow rate of growth and dissemination allows of better surgical results than are achieved in carcinoma. Lahey (1938) describes successful total gastrectomy where multiple leiomyosarcomatous masses occupied practically the whole of the stomach. Resection of the growth was possible in 3 of Schindler's 4 cases, in 3 of Golden and Stout's 5 cases, and in Fleming's case.

The main clinical features are not pathognomonic. They are anaemia, upper abdominal pain or discomfort, and an upper abdominal mass. The anaemia is secondary to gastrointestinal bleeding, which may be manifest as haematemesis or melaena, or may be occult. There may be epigastric or left upper quadrant pain, sometimes colicky, or other dyspeptic

symptoms such as heartburn or flatulence may be present without pain. Schindler *et al.* (1946) state that each of the three main clinical features is present in about 50 per cent of the collected cases.

Although the most recently reported cases of leiomyosarcoma of stomach have been in young women (Greathead's case was 21 years of age and Fleming's was 16 years old), the majority of previous cases have been over 40 years of age, and there is no preponderance in either sex. Thus leiomyosarcoma should be considered in the differential diagnosis of gastric neoplasms irrespective of the age or sex of the patient. The radiological syndrome of filling defect with central niche, together with fistulae into the filling defect, is said to be highly suggestive of leiomyosarcoma, distinguishing it from carcinoma, and from lymphosarcoma, which involves the stomach more diffusely, and which is the only one of the three to respond to radiotherapy. Though these features are commonly not all present, Schindler *et al.* claim the possibility of accurate diagnosis before laparotomy, by means of the combined results of radiology and gastroscopy.

The histological features of leiomyosarcoma have been described by Golden and Stout (1941) and by Schindler *et al.* (1946). Elliot and Wilson (1952) emphasize that histology is the only accurate means of differentiating malignant mesenchymal from malignant epithelial neoplasm of stomach. Schindler *et al.*, however, stress the difficulty of pronouncing on microscopy alone whether a leiomyoma is benign or malignant. Calcification on histological section of simple leiomyoma of stomach has frequently been mentioned, and though gross calcification visible on straight X-ray of abdomen is rare, cases showing this feature have been reported by Garbini and Price (1950) and by Leigh (1950). However, none of the cases of malignant leiomyoma mentioned above showed calcification of this degree.

Very few instances of secondary deposits in lung from leiomyosarcoma are on record, and gross calcification in such secondary deposits must be extremely rare. Golden and Stout describe, without noting calcification, one case showing pulmonary metastases on X-ray. Even in simple leiomyoma of bronchus calcification was absent in the case of Turkington *et al.* (1950), and only microscopic in that of Williams and Daniel (1950).

The following case is presented as an addition to the literature and because of certain unusual features.

CASE REPORT.

Miss B. W. was 18 years of age when she was admitted to Gartloch Hospital on 3rd December, 1944, under the care of one of us (J. J.), two months after a severe haematemesis. Apart from childhood fevers she had been well until 1943, when, while in the Services, she began to complain of lack of energy and lassitude. Blood count on 27th July, 1944, had been Hb. 68%, R.B.C. 3,840,000/cu.mm., W.B.C.

6,000/cu.mm. On admission she had a hard slightly tender mass, 3″×3″, in the upper abdomen, to the right of the midline, and barium meal examination showed a large filling defect in the body of the stomach, best demonstrated in the recumbent position. Hb. was 40%.

Operation findings—1. At laparotomy on 9th January, 1945, large masses were found projecting into the posterior wall of the stomach, extending from the cardiac end, along the lesser curvature, and up into the portal fissure. One such mass occupied almost three-quarters of the left lobe of the liver. Several nodules were present in the wall of the pyloric antrum. The sub-pyloric and the aortic lumbar glands were enlarged. Peritoneal seeding was present in the pelvis and both ovaries were involved in the tumour growth. As pyloric obstruction seemed imminent due to glandular enlargement, a posterior gastroenterostomy was performed, no technical difficulty being encountered. A lumbar gland and a portion of a nodule in the gastric wall were removed for histology. The post-operative course was uneventful.

Histological findings—1. The material taken from the lumbar region and that from the subserous tissue of the stomach presented the same histological features. It consisted of a mass of round and elongated cells showing so little structural differentiation that it was impossible to form an opinion as to their normal prototype. It was considered to be a malignant tumour and the designation of spindle-cell sarcoma was suggested. The sections were seen by Professors J. W. S. Blacklock and R. W. Scarff, who agreed on the difficulty in diagnosis, but concluded that it was probably a myomatous tumour of low malignancy.

Further progress. Her progress was observed at intervals thereafter, and on 16th July, 1945, screening of the chest revealed a rounded opacity at the hilum of the right lung and a smaller one just above it. These were thought to be secondary deposits, though when a film on 7th November, 1946, showed no change in their size, and demonstrated flakes of calcification in the lower mass, the possibility of tuberculosis was raised by the radiologist.

During the years until 1950 she kept reasonably well apart from tiredness and slight flatulence, although blood count on 5th September, 1949, was Hb. 50%, R.B.C. 3,180,000/cu.mm., W.B.C. 4,600/cu.mm., and the blood film indicated severe anisocytosis and ring-staining. There was little response to intensive oral therapy with iron and cod liver oil or to parenteral liver therapy.

On 14th April, 1950, she was admitted to Glasgow Royal Infirmary under the care of Dr. J. H. Wright, complaining of fatigue and flatulence, and of dyspnoea on exertion of recent origin. For two months she had had recurrent bouts of upper abdominal colic and nausea ; vomiting occurred once only.

She was a well nourished young woman (weight, 8 st. 3½ pounds). Her complexion and mucosae were pale. The abdomen was slightly distended and the liver edge was hard and irregular, 3-4 cm. below the costal margin. Three large firm masses were palpable in the upper abdomen, and the lower abdomen was resistant to palpation. Several hard nodules in the pelvis were felt on rectal examination. Blood count was Hb. 64%, R.B.C. 4,050,000/cu.mm., W.B.C. 7.400. Blood film showed anisocytosis and ring-staining, and differential count was normal. B.S.R. (Westergren) was 13 mm. in 1 hour, 26 mm. in 2 hours. Sternal marrow was cellular, active and normoblastic. On barium meal examination several large filling defects were seen on the posterior wall of the body of the stomach, and mucosal defects over some of them were consistent with ulceration. The first inch of the gastroenterostomy was infiltrated. X-ray of chest showed the two masses near the right hilum to be unchanged.

During her stay in hospital faeces were repeatedly positive to tests for occult blood. She was sent home on 9th May, 1950, symptomatically improved, but with blood count little changed. She was readmitted on 17th November, 1950, and again on 30th March, 1951, because of severe anaemia (on 17th November, 1950, Hb. was 34%, and R.B.C. 2,100,000/cu.mm.), resulting mainly from repeated melaena. There was slight haematemesis once only. Two pints of blood were given on 20th November, 1950, and the response to iron therapy thereafter was fairly good. In April, 1951, abdominal examination and barium meal showed little change, though on X-ray of chest the calcification in the lower thoracic mass seemed more dense.

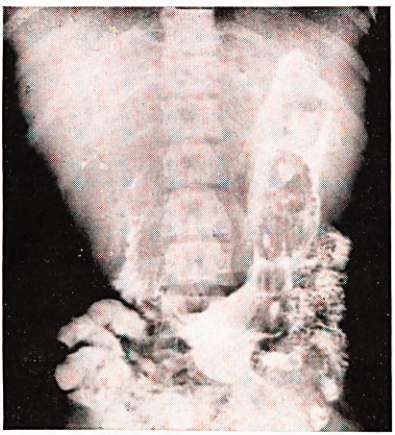

Fig. 1. Barium meal, April, 1950, showing multiple large filling defects in stomach with central niche in several.

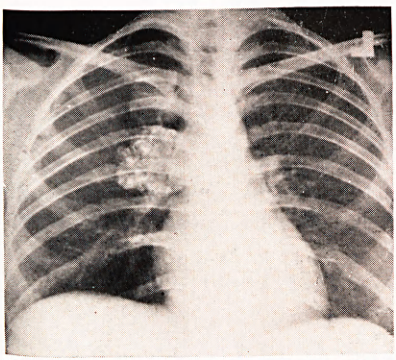

Fig. 2. Straight X-ray of chest, August, 1952, showing masses in R. hilum, the lower one calcified.

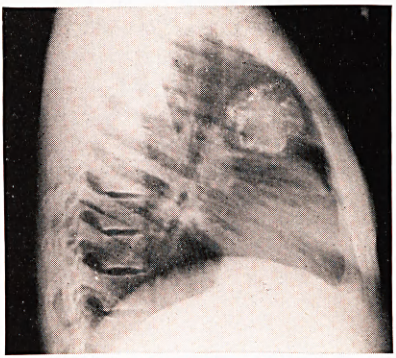

Fig. 3. Right lateral view of chest, August 1952.

Operation findings—2. As her main disability was anaemia secondary to gastric haemorrhage, Professor Illingworth's advice was sought with regard to the possibilities of surgical treatment and particularly gastrectomy. An exploratory laparotomy was done by him but as can be seen from his report which follows, gastrectomy was impossible.

'The stomach was occupied by a massive tumour some 6" in its long diameter. This tumour appeared to be arising from the posterior wall of the stomach in the region of the incisura, and at that point it had erupted posteriorly on to the surface of the stomach and was beginning to envelop the gastreonterostomy stoma. The remainder of this main growth was lying within the lumen of the stomach extending upwards to the region of the cardia.

'The anterior wall of the stomach was normal. In the region of the pylorus, however, there were three or four small intramural growths each a few millimetres in diameter. One such growth was present in the first part of the duodenum, though the small intestine appeared to be free of them.

'There were multiple secondary deposits present within the abdomen. Two large masses some 3" or 4" in diameter appeared to have originated from glands in the pyloric region. One of these growths was almost completely cystic. The liver contained a large number of metastases scattered through both lobes. In addition there were numerous metastases in the pelvis, some of which appeared to have developed on the ovaries. Finally the omentum contained numerous nodules. Portions of the main tumour mass from the vicinity of the gastroenterostomy stoma, from nodules in the omentum, and from one nodule in the liver were removed for histological examination.'

Histological findings—2. The following histological report was supplied by Dr. John R. Anderson :—'The biopsy material from the stomach consists of neoplastic tissue in which many of the cells are elongated, while others are distended by a large clear vacuole which does not give a positive reaction for fat, mucin or glycogen. The tumour contains many thin-walled vascular spaces. Mitoses are not numerous. The nodule from the liver is enclosed in a dense fibrous capsule, and has been present for some considerable time ; it is more highly cellular than the gastric tumour and the cells tend to be arranged in groups and show a whorled arrangement. The peritoneal nodules show all transitions in histological appearances between the gastric and hepatic tumours. Professor Cappell considers the tumours to be of low grade malignancy, and considers that they may be leiomyosarcomata.' Professor D. F. Cappell showed the sections to Professor R. W. Scarff, who had seen the sections from the previous laparotomy, and he agreed with the probable diagnosis of leiomyosarcoma.

The patient quickly recovered from the laparotomy, and when seen again on 8th August, 1952, was feeling well and able to move about freely. She had put on weight and there had been no further haematemesis or melaena. The opacities in the right hilar region on X-ray of chest were unchanged. The masses in the abdomen, however, felt, if anything, bigger than before, and the blood count was Hb. 68%, R.B.C. 3,600,000/cu.mm.

SUMMARY.

The literature on leiomyosarcoma of the stomach is briefly reviewed.

An unusual case is described of leiomyosarcoma of the stomach in a young woman with multiple gross secondary deposits known to have been present for 7½ years. The patient is still alive and relatively well, showing no emaciation despite extensive ulceration of the growths in the stomach. X-ray of chest revealed two masses of uncertain nature near the right hilum ; one of these shows considerable calcification.

ACKNOWLEDGEMENTS.

We wish to thank Dr. J. H. Wright for permission to publish and for helpful advice ; Professor C. F. W. Illingworth for operation notes ; Dr. J. R. Anderson for histological report ; Professors J. W. S. Blacklock, D. F. Cappell and R. W. Scarff for permission to quote their opinions on histological sections ; and Mr. Towler for X-ray reproductions.

REFERENCES.

Elliot, G. V. & Wilson, H. M. (1952). *Arch. intern. Med.* **89** : 538

Fleming, C. M. (1951). *Glasg. med. J.* **32** : 3, 77

Garbini, J. & Price, H. P. (1950). *New Engl. J. Med.* **243** : 406

Golden, T. & Stout, A. P. (1941). *Surg. Gynec. Obstet.* **73** : 784

Greathead, E. (1949). *Brit. med. J.* **2** : 422

Lahey, F. H. (1938). *Surg. Gynec. Obstet.* **67** : 213

Leigh, T. F. (1950). *Radiology.* **55** : 419

Schindler, R., Blomquist, O. A., Thompson, H. L. & Pettler, A. M. (1946). *Surg. Gynec. Obstet.* **82** : 239

Turkington, S. I., Scott, G. A. & Smiley, T. B. (1950). *Thorax.* **5** : 138

Williams, R. B. & Daniels, R. A. (1950). *J. thorac. Surg.* **19** : 806