UNSUSPECTED OESOPHAGEAL LEIOMYOMA FOUND AT HIATAL HERNIORRHAPHY

REPORT OF TWO CASES

BY

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Leiomyomas of the oesophagus are relatively rare. The chance finding, in seven months, of two leiomyomas during transthoracic repair of two apparently simple sliding hiatal hernias emphasizes again that hiatal hernias do sometimes occur in association with tumours in the lower third of the oesophagus, that some of these tumours may be undetectable despite barium studies and oesophagoscopy until operation, and that only by the transthoracic route can they be readily detected and treated.

Case 1

A farmer aged 56 was admitted to hospital on May 27, 1957. He gave a four-year history of epigastric discomfort, flatulence, dyspnoea, and hiccups which first began with heavy lifting when making concrete paths. His chief disability was severe flatulence and regurgitation with any bending job. Occasionally there were bouts of vomiting. Radiographs in 1954 confirmed the presence of a hiatal hernia, which was at first treated medically. In 1957, however, because the symptoms persisted, he was referred for surgical treatment. His past history was non-contributory.

Physical examination showed a well-built man of 14 st. (88.9 kg.). His blood-pressure was 130/90 mm. Hg, and no physical abnormalities were detectable. As there was clear x-ray evidence of hiatal hernia (Fig. 1), oesophagoscopy on this occasion was not performed.

Operation

On May 29 the left chest was opened through the left eighth-rib bed. After division of the inferior pulmonary

FIG. 1.—Barium swallow, showing hiatal hernia without oesophageal obstruction.

ligament the oesophagus was isolated. This proved unexpectedly difficult because of a large unsuspected tumour which lay within the oesophageal wall, just beyond the upper limit of his large hiatal hernia. After the oesophagus was dissected from the posterior mediastinum, closer inspection showed that the tumour was lobulated, was almost the size of a cricket ball, almost completely encircled the oesophageal lumen without greatly narrowing it, and extended at least 9 cm. up the wall of the lower oesophagus. A small radial incision was made in the diaphragm, and inspection showed no enlargement of the regional lymph nodes along the course of the left gastric artery. Because the growth lay strictly within the oesophageal wall and was in no way attached to any adjoining structure, the most likely diagnosis was thought to be a leiomyoma of the oesophagus.

Excision of Tumour.—A vertical incision was made through the oesophageal muscle layer and the tumour found to lie in the submucosa. With care it was gradually enucleated. There were several ramifications which extended upwards and downwards from the main tumour mass, without invading the mucous membrane. The tumour was ultimately dissected free and several small vertical mucosal splits were closed with fine silk sutures on atraumatic needles. The overlying thickened muscle wall of the oesophagus was finally approximated with interrupted No. 0 chromic catgut sutures placed in two layers.

Repair of Hiatal Hernia.—The hiatal hernia was next reduced by passing the catheter sling around the oesophagus down through the hiatal opening and out again through the radial incision in the diaphragm. The dissected crural fibres of the diaphragm were then approximated behind the oesophagus with four double No. 4 silk sutures. The oesophagus was also sewn to the diaphragm in the hiatal region with a series of interrupted No. 2 silk sutures. The small exploratory incision in the diaphragm was closed with two layers of double No. 2 silk sutures, and the chest wall was closed over water-seal drainage.

Progress

The patient was kept on intravenous fluids for two days, after which he took increasing amounts of fluid by mouth, and solid food after 10 days. He was discharged fit 12 days after operation.

Pathological Report.

—The specimen consisted of a lobulated tumour, 9 by 5 by 2 cm., which had been excised from the submucous layer of the lower oesophagus. Histologically it was composed of interlacing bundles of smooth-muscle fibres which showed no evidence of malignancy (Fig. 2). This was a leiomyoma of the oesophagus.

Follow-up.—When seen two years later he was well. A check oesophagoscopy on June 4, 1959, showed satisfactory function of the cardia, and no evidence from the mucosal surface to suggest any recurrence. Check barium swallow also showed no evidence of tumour recurrence.

FIG. 2.—Leiomyoma discovered in lower third of oesophagus when repairing the hiatal hernia.

Case 2

A labourer aged 59 was admitted to hospital on November 21, 1957, for surgical treatment of a hiatal hernia. He first consulted his private practitioner on July 20,

1957, complaining of severe substernal pain and sweating. For 20 years he had noticed retrosternal "heartburn," generally initiated by leaning forward or lying down. Over the past six months it had become more frequent, occurring as often as two or three times a day. It was relieved by taking sodium bicarbonate or the proprietary preparation "rennies." He had severe flatulence. At no time did he complain of dysphagia. There was no history of melaena, haematemesis, or any other complaint referable to the alimentary system.

In addition to his heartburn, he complained of palpitations while working, together with a crushing precordial pain suggestive of angina of effort which doubled him up and at times radiated down his left arm. His past history and family history were non-contributory.

On physical examination he was a somewhat obese man weighing 13 st. 2 lb. (83.5 cm.). His alimentary, cardiovascular, respiratory, genito-urinary, and central nervous systems were all normal. His blood-pressure was 170/100 mm. Hg, his electrocardiogram was normal, and an oesophagogram showed reflux from the stomach into the lower oesophagus with a hiatal hernia.

Oesophagoscopy on November 22 revealed a lax cardia with no normal sphincteric control. There was free regurgitation of gastric juice into the lower oesophagus without any severe ulceration of the mucous membrane. The oesophagoscope passed easily through into the stomach.

A provisional diagnosis of reflux oesophagitis and possible coronary insufficiency was made, and repair of the hiatal hernia advised to relieve symptoms.

Operation

On November 27 the left pleural cavity was opened through the left eighth intercostal space. Pleural adhesions over the left lower lobe were divided. After division of the inferior pulmonary ligament the oesophagus was isolated in the posterior mediastinum and raised with a catheter sling. Examination at this stage showed not only a hiatal hernia that admitted all the fingers of one hand but also a large lobulated tumour growing within the wall of the lower third of the oesophagus. The tumour arose from the left side of the oesophagus; it penetrated through the musculature and grew in a lobulated fashion throughout the peri-oesophageal tissue of the posterior mediastinum. In its fresh state it measured 7 by 3 by 1.5 cm. It was not firmly adherent to any structure, nor was there any lymph-node enlargement. It had all the pathological characteristics of an extramucous, extramural leiomyoma.

A small radial incision was made in the diaphragm; the hernial sac was opened and the crura of the diaphragm defined. At this stage, after a vertical incision in the oesophageal wall, the tumour was further dissected and was found to extend down to, but not through, the mucous layer of the oesophagus. It was carefully excised without injuring the mucous membrane, and the oesophageal muscular layers were then approximated with interrupted No. 2 silk sutures, placed at 1-cm. intervals. Thereafter the hiatal hernia was repaired by a technique similar to that in Case 1.

Progress

Convalescence was complicated by urinary retention, secondary to minimal lateral-lobe prostatic hypertrophy. At first this was treated by an indwelling Foley catheter. Ankle oedema was detected on December 7, and a further E.C.G. showed a left bundle-branch block.

Some 19 days after this thoracic operation the patient was transferred to the urological department, where a transurethral resection of a median-bar prostatic obstruction was performed. Four days later, while shaving in the bathroom, he dropped dead. Necropsy revealed signs of heart failure secondary to a recently healed intraventricular myocardial infarct, with pulmonary oedema, small bilateral hydrothoraces, and two small pulmonary artery thromboses in the right lower lobe. In addition the myocardium was

hypertrophied, there was severe atherosclerosis of the right and left coronary arteries, and there were two infarcts of the right kidney.

The specimen consisted of a lobulated tumour measuring 6 by 2.5 by 2 cm. The cut surface had a whorled, homogeneous appearance, with occasional flecks of calcium. Histological section showed that the tumour comprised smooth muscle together with a large amount of collagen. This was a leiomyoma of the oesophagus.

Discussion

The occurrence of unsuspected tumours in the oesophagus may be considered rare when viewed in the light of Moersch and Harrington's (1944) findings of 44 cases in 7,459 routine necropsies, yet Meissner's (1944) finding of 46 leiomyomas in 50 random careful necropsy examinations of the stomach suggests far greater frequency.

Malignant tumours in association with hiatal hernias are regularly reported. Carcinomas are the commonest. The relevant literature was cited by Pack *et al.* (1952) when describing two further cases. Robb (1954), in reviewing 138 hiatal hernias, found two cases with an associated carcinoma of the oesophagus, as well as four others with carcinoma of the stomach and one from the sigmoid colon. Adler and Rodriguez (1959) found that 21 (2.5%) out of 814 hiatal hernias had an associated malignancy. Nine were oesophageal, of which two were in the upper third of the oesophagus, one in the middle, and six in the lower third. Twelve were gastric, eight being at the cardia and four spread more distally.

I have encountered five tumours—two carcinomas of the oesophagus, a lipoma, and these two leiomyomas—in a series of 75 hiatal hernias treated surgically. In addition, a fibrosarcoma has been described elsewhere—detected one year after repair of a hiatal hernia (Borrie, 1959).

Benign tumours of the oesophagus fall into two pathological and clinical groups—namely, those arising from the mucosa, "intraluminal tumours," and those arising from extramucosal tissue, "intramural tumours." These latter may remain sessile and entirely intramural, or they may extend extramurally. The mucosal or intraluminal benign tumours may be lipomas, polyps, adenomas, haemangiomas, myofibromas, and fibrolipomas as reported by Moersch and Harrington. Their intramural tumours included leiomyomas, myomas, and cysts. Other tumours reported include neurofibromas and osteochondromas.

The commonest benign oesophageal tumours are leiomyomas. Their association with hiatal hernias has rarely been recorded in the past. Schafer and Kittle (1947) reported one that gripped the entire lower oesophagus like a fist; there was an associated gastro-hiatal hernia, and it required a partial oesophago-gastrectomy. Goldman and Masters (1950) described a similar case.

The age incidence of leiomyomas corresponds to the maximum age incidence of hiatal hernias. Though Rose (1936) in reviewing 49 cases, found the 60–70 age-group most affected, in Myers and Bradshaw's (1951) series of 42 cases the ages ranged from 1½ to 66 years. Sweet *et al.* (1954) recorded a range from 19 to 67 years in patients treated surgically, with an average of 41.2 years. Further, they reported 25 cases in men and 22 in women, indicating that the sex incidence was almost equal.

The commonest site for leiomyomas is the lower oesophagus, so that again they can be expected to occur from time to time in association with the commonest lower oesophageal lesion—namely, a hiatal hernia.

These tumours vary in size from a match-head to one weighing 1,420 g. as reported by Kenney (1953). They are firm and may be round, nodular, or lobulated. Though they can be localized, they tend to encircle the oesophagus and spread upwards in the submucosal plane. Only if there is actual narrowing of the oesophageal lumen will dysphagia occur. Because the oesophageal lumen was not grossly occluded, the two patients in this report had symptoms referable only to their hiatal hernias. Neither barium studies nor oesophagoscopy suggested the presence of an intramural tumour.

The Approach to Hiatal Herniorrhaphy

Several surgical approaches to hiatal herniorrhaphy have been devised. Harrington (1955) has favoured the abdominal approach, Allison (1951) the left thoracic approach, while Collis *et al.* (1954) have advocated an abdomino-thoracic approach. I have on one occasion successfully repaired such a lesion through the right pleural cavity (Borrie, 1958).

Suffice it to say that as a guiding principle the route chosen for this operation should be the one which allows of greatest flexibility—and especially when unsuspected tumours can be discovered by chance in the lower third of the oesophagus or upper third of the stomach. This pathological fact strongly supports the view that the route that will allow inspection of both the lower oesophagus and the upper stomach should be used wherever possible, and that opportunity be taken to inspect the foregut on both sides of the diaphragm. This ideal is possible only via a left transthoracic approach with division of the diaphragm. Had these two patients been treated by the abdominal route, these tumours could not have been detected during the standard transabdominal repair and therefore would have been left to grow until the inevitable onset of dysphagia.

At operation the lobulated appearance of the lesion confined to the oesophagus with no extension into surrounding structures and no lymph-node invasion made the diagnosis of an intramural leiomyoma tolerably certain. Conservative removal was therefore chosen, and a vertical incision through the outer muscle layers gave easy access to the loose submucosal plane in which it lay. Careful dissection was required to avoid injury to the mucous layer. Thereafter the hiatal hernia was reduced and repaired by standard techniques.

Summary

The chance finding within a seven-months period of two leiomyomas of the lower third of the oesophagus during transthoracic repair of two apparently simple sliding hiatal hernias emphasizes again that hiatal hernias do sometimes occur in association with lower oesophageal tumours—benign as well as malignant—that some of these tumours may be undetectable despite barium studies and oesophagoscopy until operation, and that only by the left transthoracic route can they readily be detected and treated.

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EFFECT OF PREDNISOLONE IN BANCROFTIAN ELEPHANTIASIS

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Conflicting reports on the place of steroids in the treatment of Bancroftian elephantiasis have suggested that further trials were necessary before their efficacy in the treatment of this condition could be ascertained. Using corticotrophin, McFadzean (1953) was unable to detect any improvement in his cases in West Africa. and Subramaniam and Mohammed (1957), in India, reported only temporary improvement with cortisone. Markell and Kerrest (1955), in Tahiti, reported considerable improvement with cortisone, and Laigret, March, and Kessel (1957) obtained good results when cortisone or prednisone was combined with bandaging and Unna's paste.

The present communication is a report of a trial in which prednisolone was given to seven persons with Bancroftian elephantiasis due to periodic *Wuchereria bancrofti*.

Material and Methods

The diagnosis of Bancroftian elephantiasis is sometimes difficult, as laboratory tests are rarely of help and clinically the condition is often indistinguishable from other causes of tropical elephantiasis (Jordan, Trant, and Laurie, 1956). Patients were selected for the present series if they came from areas of Tanganyika where Bancroftian infection was known to be common. The condition had been present for periods ranging from one to eight years, and the severity of the disease varied from slight to severe. In some advanced cases the skin was verrucose, and in every case was thicker than normal. In three patients the disease was bilateral.

All patients were examined and questioned with regard to possible peptic ulceration and chest disease. Where necessary an x-ray examination was made. The fluid intake and urinary output were measured for a few days before treatment in order that any diuresis that might occur during treatment could be more accurately assessed.

The legs of all patients were measured at four levels—mid-thigh, mid-calf, at the ankle, and round the foot—before and after treatment. Measurements were made at preselected points at fixed distances from the anterior superior iliac spine. Accuracy in measuring the circum-