

BIZARRE PRESENTATIONS OF LEIOMYOBLASTOMA OF THE STOMACH: A Report of Two Cases.

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ABSTRACT

Two cases of leiomyoblastoma presenting 15 years apart, in the same centre each in a bizarre manner, but with excellent outcome on treatment are presented.

First case, a 52 yr old university don of Ibibio extraction presented at the emergency room in 1987, with a two-day history of abdominal pain, progressive abdominal distension with respiratory distress, fainting and weakness. The patient was optimized while urgent urinalysis; full blood count, electrolytes urea and creatinine were obtained. Exploratory laparotomy was carried out, which revealed a bleeding tumour on the greater curvature of the stomach. The tumour was excised and histology showed it to be leiomyoblastoma of Stout¹. Today, 16 yrs on, the patient is alive and well. The second case, a 60yr old peasant farmer again of Ibibio extraction seen in the surgical outpatient with a mass in the epigastrium of about 6 months duration. The mass had one month before presentation, undergone sudden increase in size, becoming painful in the process. Ultrasound scan of the mass, gave an impression of a pseudocyst of the pancreas. At surgery a solid tumour of gastric origin with central haemorrhagic necrosis was found and was completely excised by partial gastrectomy. Histology revealed it to be leiomyoblastoma (epitheloid leiomyoma) with areas of haemorrhage within the tumour. No other treatment was given. Follow up of the patient for 17 months now remains uneventful.

Key words. Stomach, epitheloid leiomyoma, leiomyoblastoma, leiomyosarcoma

INTRODUCTION

Gastric leiomyoblastomas are very rare and are usually not diagnosed preoperatively². They constitute only about 10% of non-epithelial gastric neoplasm³. Together with leiomyosarcoma, the spindle celled more malignant variant, they constitute less than 1 % of malignant tumours of the stomach⁴. While there are two previous reports of gastric leiomyosarcoma^{2,5}, this seems to be the first report of leiomyoblastoma in the Nigerian literature. It has however been reported from central Africa⁶. We have encountered in our service, two cases over a 15 yr period, each case presenting in a rather bizarre manner with haemorrhage as a prominent feature, we therefore document them as very rare cases of non epithelial gastric neoplasms with frightening presentation and yet excellent outcome on treatment.

CASE No.1 (1987)

A 52yr old university don, O.U.E., Hosp No. 191265 was first seen in the accidents and emergency unit with a two-day history of sudden abdominal pain around the umbilicus which became generalized with associated progressive abdominal distension. There was no vomiting nor nausea and no history of trauma. Clinically he looked well but distressed. In the abdomen there was generalized abdominal distension, no guarding or rigidity but tenderness

the left para-umbilical area. No masses palpated, bowel sounds were distant, few and far between. Chest was clinically clear. Investigations showed results as follows: Urinalysis-normal, Hb 9.9gm/dl, WBC $4.7 \times 10^9/L$, Blood group O⁺, Urea 5.8mmol/L, Creatinine, 92.7 $\mu\text{mol/L}$, Sodium 138mmol/L, Potassium 3.0 mmol/L, Chloride 100mmol/L and Bicarbonate 24mmol/L. The patient was optimized and then under general endotracheal anaesthesia, exploratory laparotomy was carried out. Operative findings were massive haemoperitoneum, with large clots in the lesser sac from a bleeding pedunculated solid tumour measuring about 2cm by 4 cm, on the posterior wall of the greater curvature of the stomach. All other organs appeared normal. Bleeding was arrested, the blood in the free peritoneal cavity recovered, filtered and fed into a blood bag for autotransfusion. The tumour was excised, with a full thickness of the stomach (including the mucosa) and the defect repaired in two layers. Clots from the lesser sac and elsewhere were removed and peritoneum mopped dry. The excised specimen was sent for histology following which a diagnosis of "Bizarre leiomyoma - Vascular leiomyoma (leiomyoblastoma of Stout¹)" was made. The postoperative period was uneventful and the patient was discharged home on the 10th postoperative day. Sixteen years on, the patient is well and happy in his retirement.

CASE No. 2 (2002)

A 60yr old rural subsistence farmer, B.I.B, presented at the surgical outpatient department with a six-month history of abdominal swelling. The swelling was initially small, but increased progressively in size, becoming painful 3 months prior to presentation. The pain, initially mild and intermittent, was biting in character, usually aggravated by eating and drinking and relieved when the stomach was empty. One month to presentation, the mass had undergone a sudden increase in size; the associated pain became constant and more severe, prompting him to present at the GOPD from where he was referred.

Clinical evaluation showed he had started losing appetite and weight. The pain did not radiate to any other part of the abdomen, there was no associated vomiting and no history of trauma or melina stool. Physical examination showed the patient was pale and anicteric with obvious weight loss. The abdomen was scaphoid and asymmetrical, with an irregular; ovoid centrally located mass measuring approximately 15 cm by 16 cm, in the epigastrium, reaching down to the umbilical region. The mass was slightly tender, mobile and generally firm in consistency with some soft areas. The liver, spleen and kidneys were not palpable. Bowel sounds were present and normal. Investigations including Urinalysis, Full blood count, Electrolytes, urea & creatinine and chest x-ray were obtained and the results were all normal. Ultrasound scans of the abdomen however gave an impression of a cystic mass in the area of the pancreas. An impression of Pancreatic pseudocyst was made based on sonologic findings and the patient was prepared for laparotomy.

The abdomen was entered through an upper mid-line incision. Findings were an irregularly shaped cystic mass 15 cm in diameter, attached to the lower edge of the greater curvature of the stomach, the pylorus and greater omentum, with several solid 'seedings' on the omentum. The liver was free. Complete excision of the tumour, with full thickness of the stomach wall and a good tumour margin, as well as the omental 'seedings' was carried out. Division of the tumour showed a haemorrhagic central portion, which created the impression of a cyst on the ultrasound scan. All specimens were sent for histology and the histological diagnosis was given as "Epithelioid leiomyoma (Benign leiomyoblastoma)", a photomicrograph of which is shown in Fig.1. The postoperative period was uneventful and the patient was discharged home on the 9th postoperative day without any other treatment, as the patient could not afford it. Seventeen months

on, the patient remains well with no complaints directly related to the lesion

DISCUSSION

Gastric stromal tumours are generally rare, constituting less than 1% of malignant tumours of the stomach. Ten percent of this is accounted for by Leiomyoblastomas³. Davies⁷ reporting from a British centre recorded 4 cases in a 12-year period, while from another British center, 3 cases of 2799 gastroscopies performed over a 4-year period were found to be leiomyosarcomas⁸. Figures from our department of pathology show that during the period when these two cases were seen, a total of nine biopsy specimens of gastric tumours were received confirming our relatively low incidence of gastric stromal tumours. They present clinical, therapeutic and prognostic features very different from common epithelial tumours³. In a rare report of a large series, from one centre⁹, workers from the Anderson Hospital and Tumour institute in Houston USA, reviewing 50 cases seen over twenty-two years, made certain general observations on the clinico-pathological characteristics of these tumours. They observed that; median age at presentation is above 50yrs, abdominal mass is the commonest physical sign, pain and weight loss are prominent symptoms, jaundice is not encountered, there may be no symptoms referable to the gastrointestinal tract, chest x-rays are normal, diagnosis is made at surgery and the tumours have a high propensity to grow to a large size before discovery.

Findings in our two index cases while conforming largely to these generalizations differed though, in a few areas. The first case did not grow to a large size before manifesting, most probably because of the acute episode of haemorrhage. Its gross appearance at surgery resembled a haemangioma, which was the intraoperative diagnosis. The second case differed in manifesting the gastrointestinal symptoms of poor appetite and worsening of symptoms by eating, but more especially in the mimicry of pancreatic pseudocyst. Histology revealed it to be an epithelioid leiomyoma (leiomyoblastoma), a rare, benign but potentially malignant tumour, with sometimes bizarre presentations. Early and complete excision with full thickness of the stomach wall carries excellent prognosis as demonstrated by our two cases. However when metastatic or when inadequately excised, the prognosis is uniformly poor.

Kamiga et al¹⁰ reported 12 cases of gastric leiomyoblastomas with metastasis to the liver all with very poor prognosis. We therefore report these cases on account of the rarity of the lesion and also to

illustrate the good prognosis that attend adequate and complete excision of the tumour.

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