

Inflammatory myofibroblastic tumor of the stomach in an adult woman: a rare intermittent cause of gastric outlet obstruction

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ABSTRACT

Background. Inflammatory myofibroblastic tumor is a neoplasm of intermediate biological potential that frequently recurs and rarely metastasizes.

Case report. We report a rare case of intermittent gastric outlet obstruction by an inflammatory myofibroblastic tumor of the cardia.

Results. A 56-year-old woman presented at the gastroenterology department with a two-day history of hematemesis and melena. She had intermittent nausea and vomiting complaints, which had manifested periodically for about five months. Upper gastrointestinal endoscopy demonstrated a mass of 6 cm in diameter, which was resected. Histological examination revealed ulcerated mucosal granulation-like tissue with myofibroblastic spindle cell proliferation in a storiform pattern.

Conclusions. In order to avoid unnecessary aggressive therapy, gastric IMT should be taken into account when a gastric mass accompanied by the various clinical manifestations of IMT is found in an adult. Free full text available at www.tumorionline.it

Introduction

Inflammatory myofibroblastic tumor (IMT) has been defined as “a neoplasm of intermediate biological potential that frequently recurs and rarely metastasizes”¹. IMT is also called inflammatory pseudotumor, plasma cell granuloma, inflammatory myofibroblastoma, and inflammatory myofibrohistiocytic proliferation. It is an inflammatory solid tumor that contains spindle cells, myofibroblasts, plasma cells, lymphocytes, and histiocytes^{2,3}. Inflammatory pseudotumors rarely occur in the stomach^{2,3}. A thorough literature search identified only 6 reported cases in the English-language medical literature so far²⁻⁶. We report, for the first time, a rare cause of intermittent gastric outlet obstruction in the form of an IMT of the cardia.

Case report

A 56-year-old woman presented at the gastroenterology department with a 2-day history of hematemesis and melena. She complained of intermittent nausea and vomiting, which had manifested periodically for about 5 months. There were no other symptoms. On examination, the patient appeared pale. Her body temperature was 36.7 °C, the respiratory rate 16/min, heart rate 110 beats/min, and blood pressure 90/50 mmHg. Findings on cardiopulmonary examination were normal. The abdomen was soft and not tender. Her medical background and family history were unremarkable and physical examination was normal. Blood tests included a hemoglobin level of 5.9 g/dL, a white blood cell count of 11,800/mm³, and a platelet count of

Key words: inflammatory myofibroblastic tumor, endoscopy, spindle cell.

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372,000/ L. Coagulogram, liver function tests, creatinine and BUN were normal. Fluid resuscitation was started immediately and the patient was transfused with 2 units of red blood cells. After stabilization, an esophagogastroduodenoscopy was planned. Upper gastrointestinal endoscopy demonstrated a mass of 6 cm in diameter (Figure 1). The tumor was a large polyp originating from the cardia and extending towards the pylorus. There was no active bleeding. Stacked (bite-on-bite) forceps biopsy specimens were obtained, which showed only normal mucosa. A computed tomography (CT) scan of the abdomen demonstrated a well-defined lesion in the gastric body (Figure 2). On ultrasound and CT no evidence of metastatic disease or infiltration of adjacent organs was found. However, because of the size of the tumor, which was suggestive of malignancy, surgical ex-

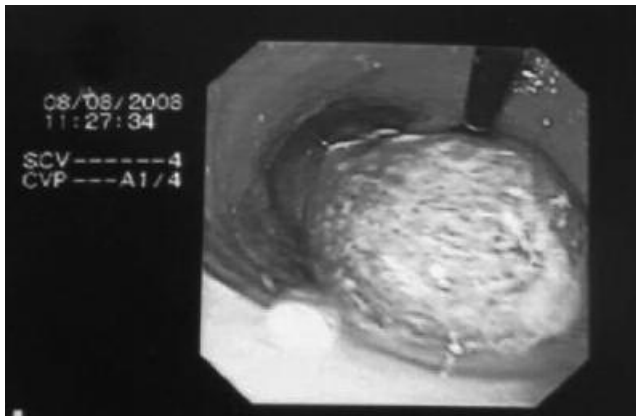


Figure 1 - Endoscopic view showing a large elevated lesion just beneath the cardiac region.

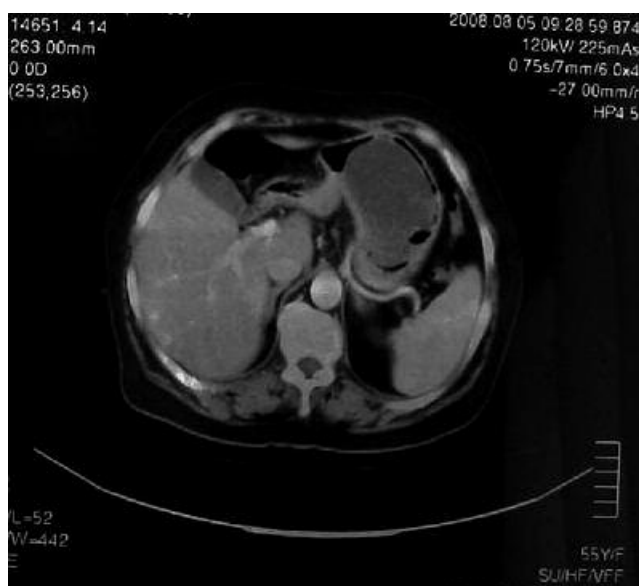


Figure 2 - Abdominal computed tomography showing a large mass located in the gastric wall.

cision was performed. At laparotomy, a round, rubbery mass with an extragastric component was excised; a definite diagnosis could not be made based on frozen-section biopsies, so a partial gastrectomy (Billroth I) was performed. The surgical procedure was completed without complications, and the patient tolerated the procedure well. Macroscopic examination of the resected portion of the stomach revealed a tumor mass that involved the entire thickness of the gastric wall. Most of the mass was a polyp; one portion consisted of a 11 × 7 × 5 cm solid white nodule. Histological sections of the tumor mass showed ulcerated mucosal granulation-like tissue, with myofibroblastic spindle cell proliferation in a storiform pattern. The mitotic activity was low, and cellular atypia was minimal. An increased number of plasma cells occurred singly and in clusters and lymphoid aggregates were present (Figure 3). Immunoperoxidase stains showed reactivity of the spindle cells to vimentin (Figure 4) and smooth-muscle actin, while being negative for c-kit, CD34, desmin, S-100, CD31 and anaplastic lymphoma kinase-1. These findings were consistent with IMT. The size of this gastric inflammatory pseudotumor was very uncommon. The patient has been symptom-free over 8 months of follow-up.

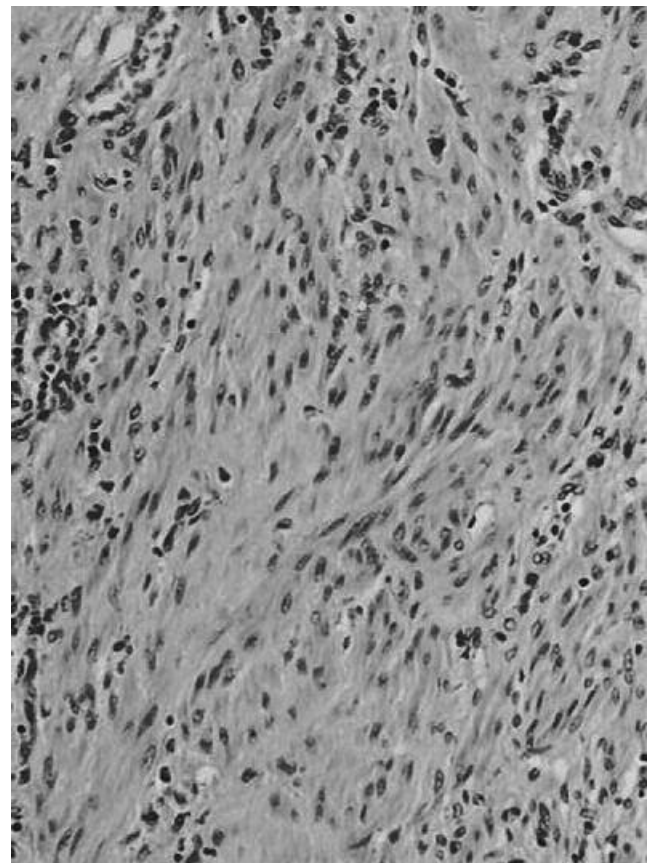


Figure 3 - High-power view of the mass showing inflammatory cells.

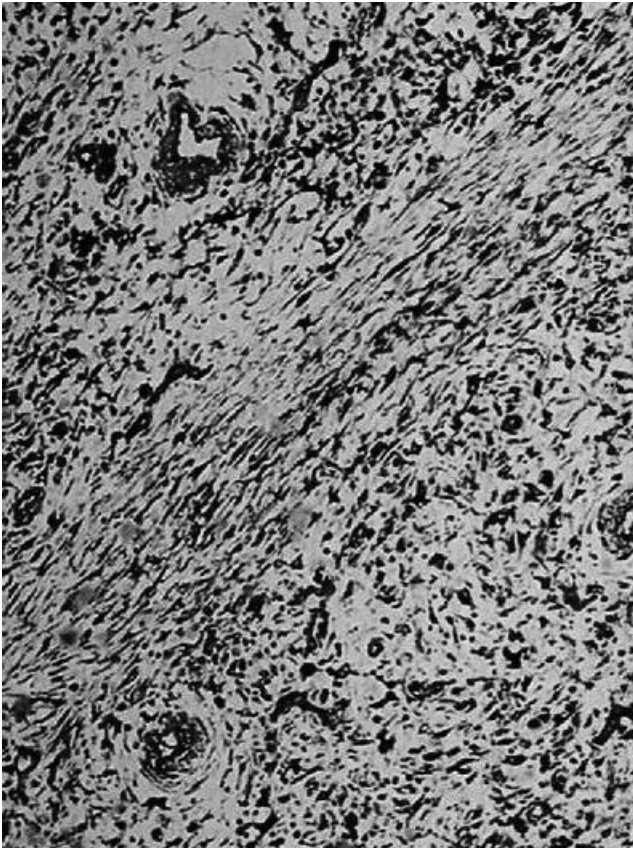


Figure 4 - The tumor cells showed positive immunoreactivity for vimentin ($\times 100$).

Discussion

IMTs have been identified in virtually every organ system but are most commonly reported in the lungs. Intraabdominal sites of the disease are most often the liver, followed by stomach, bowel, and spleen. IMT is primarily a disease of children and young adults. Gastric IMT in adults is very rare and to the best of our knowledge the present case is only the seventh reported in an adult²⁻⁷.

There has been debate as to whether IMT is a pseudotumor or a neoplasm and whether it is benign or malignant. It is currently regarded as a specific neoplasm within the larger descriptive category of inflammatory sclerosing and fibrosing processes. It is locally recurrent but rarely metastasizes¹. The tumors are histopathologically composed of myofibroblastic spindle cells, with an inflammatory cell infiltrate of plasma cells, lymphocytes, and eosinophils⁸.

IMTs are often large, ranging in size from 3 cm to 10 cm. The gastric cases described to date were located at different sites in the stomach: cardia, antrum, and prepyloric region⁹. Our patient presented with a large IMT that had prolapsed through the pylorus, causing symptoms due to gastric outlet obstruction. These tu-

mors have been described by Lazure as whitish or translucent, single or multiple, sometimes ulcerated, endoluminal polypoid nodules¹⁰. They are infiltrative and often extend through the gastric wall, sometimes reaching adjacent organs including the esophagus, duodenum, peritoneal cavity, spleen, pancreas, and liver^{5,9-11}, as well as adjacent structures such as the falciform ligament or greater omentum^{1,4}. These features mimic malignancy on endoscopy and radiological imaging^{5,11}.

Even with a thorough diagnostic workup, which may include ultrasonography, CT, endoscopy, and percutaneous fine-needle aspiration biopsy, it is difficult to make an accurate preoperative diagnosis of IMT. Confirmation with microscopic examination of gastric specimens is frequently required¹². The differential diagnosis of IMT is extensive and depends on tumor site and morphological characteristics, and on the nature of the specimen available for examination^{1,13}.

In conclusion, gastric IMT is a rare lesion in adults. It is, however, surgically curable and should therefore be accurately differentiated from sarcomas at the time of exploration. In order to avoid unnecessary aggressive therapy, gastric IMT should be taken into account when a gastric mass accompanied by the various clinical manifestations of IMT is found in an adult. Resection of the tumor was the treatment of choice in all cases reviewed¹². After surgical excision, endoscopic follow-up is recommended because of the possibility of recurrence at the excision site or development of malignancy in the remote gastric mucosa, as well as the difficulty in predicting the prognosis¹⁴.

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