

Inflammatory Pseudotumour of Stomach in an old lady: a rare case report

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ABSTRACT

Inflammatory pseudotumour (IPT) is a rare benign solid tumor in adults and children. The prevalence, etiology and pathogenesis of this condition are still uncertain. Despite the use of modern laboratory techniques and imaging, it is often difficult to make the diagnosis of IPT. Besides, occasionally the nonspecific morphological appearance and clinical presentation of the mass may mimic other more common primary or secondary neoplasms. IPT is commonly encountered in the lung and mediastinum. Other sites include abdomen (liver, pancreas, stomach, omentum), retroperitoneum, pelvis (bladder) and extremities in children. We report a rare case of gastric inflammatory pseudotumour in a 65-year-old female patient. Clinical presentations and its management along with review of literatures are presented.

KEYWORDS: Inflammatory pseudotumour of stomach, Elderly, Rare

INTRODUCTION

Inflammatory pseudotumour (IPT) is a rare benign lesion in adults and children and it can occur in any site of the body.^{1,2,3} Other synonyms for IPT include inflammatory fibromyoblastic pseudotumor, plasma cell granuloma, plasma cell pseudotumour, omental mesenteric myxoid hamartoma and inflammatory fibrosarcoma.⁴ However, the term IPT being the most descriptive is widely used in the literature.

Gastric IPT in adults is a very rare disease. Only five reported cases of gastric IPT in adults exist in the English literature.^{5,6,7,8,9} We describe a case of a 65-year-old lady who suffered from abdominal discomfort, constipation and abdominal distension for a year due to growing gastric inflammatory pseudotumour. We report this case because of its rarity and its presentation bearing close resemblance to malignancy.

CASE REPORT

A 65-year-old female patient was referred to the surgical clinic for further investigation of abdominal mass. She claimed that she suffered from abdominal discomfort and intermittent constipation for a year. She denied loss of weight or appetite, febrile illness, urinary symptoms or abdominal distension. Clinical examination revealed a huge painless mass over the epigastric region extending to the left hypochondrium

measuring 20 x 15 cm in size. The mass had a smooth surface and well-defined margins. The mass was hard in consistency and moved in both vertical and horizontal directions during the palpation. There were no stigmata of chronic liver disease and no hepatosplenomegaly. Other physical examinations were normal. Blood, renal and hepatic parameters were normal. Hepatitis serology of HbsAg and anti-HCV, a-fetoprotein, and carcinoembryonic antigen were negative.

Computed tomography (CT) scan of the abdomen showed a well-defined cystic mass measuring 12.8 x 9.6 x 12.8 cm in the epigastric region (Figure 1). Mesenteric cyst with probable infection or hemorrhage was suspected. Differential diagnosis at that point of time was gastric lymphoma and gastrointestinal stromal tumour (GIST) of the stomach.



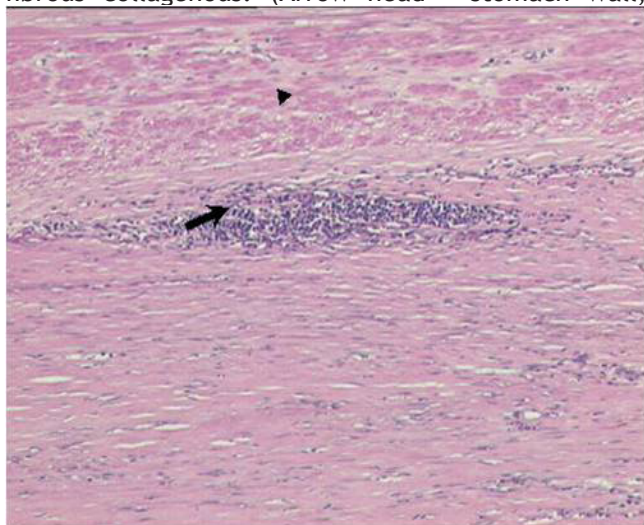
Figure 1: Photography of CT abdomen showing defined cystic mass 12.8 x 9.6 x 12.8 cm in the epigastric region (white arrow). The cystic mass has thickened

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wall and contained protein-like material.

The patient underwent explorative laparotomy, complete excision of the mass and subtotal gastrectomy with gastrojejunostomy. Macroscopically, the tumour was solid and lobulated measuring 20 x 14 x 12 cm in size arising from the greater curvature of stomach. Histopathological study of the excised specimen showed diffuse infiltration of plasma cells in all the specimens, walled by dense hyalinised fibrous collagenous tissue (Figure 2). Giant cells of foreign body type and fibroblastic reaction were noted. There was no evidence of dysplasia or metaplasia from the specimen. Histopathological diagnosis was IPT of stomach.

Figure 2: Photomicrograph showing diffuse infiltration of plasma cells, that walled by dense hyalinised fibrous collagenous. (Arrow head - stomach wall,



black arrow - inflammatory plasma cells, white arrow hyalinised fibrous collagenous matrix).

Postoperative course was uneventful. At follow up nine months later, she was well, with no evidence of recurrence.

DISCUSSION

We highlight a rare case of IPT in elderly patient who presented with vague abdominal pain, change of bowel habit and fast growing abdominal mass within a year. Despite the use of modern laboratory techniques and imaging, making a definite diagnosis of IPT is often difficult. Our patient had non-specific clinical presentation and the mass also had non-specific morphological appearances that mimic other lesions, such as primary or secondary neoplasm.

The IPT is characterized pathologically by a diffuse infiltration of inflammatory cells with fibromyoblastic proliferation. The predominant cells may be plasma cells, neutrophils or lymphocytes.³ The prevalence, etiology and pathogenesis of this condition are still uncertain. The condition is thought to be related to

an unusual tissue response to injury, bacterial, viral and fungal infection and autoimmune causes.¹⁰ There are many reports which associate IPT with *Mycobacterium avium intracellulare*, *Campylobacter jejuni*, *Bacillus sphaericus*, *Coxiella burnetii*, *Escherichia coli*, Epstein-Barr Virus and acute retroviral syndrome.¹¹ Association of IPT and radiotherapy, steroid usage and some genetic factors has also been reported.¹²

There are still two controversial issues regarding IPT. Is IPT purely an inflammatory entity or neoplastic in its origin? If it is neoplastic, is IPT benign or malignant tumour? Few genetic studies on cytogenetic abnormalities such as rearrangements of the ALK gene on chromosome 2p23, clonal chromosome abnormalities and DNA aneuploidy, and the role of oncogenic viruses suggest that IPT is a true neoplasm.¹³⁻²⁰ According to the current classification of the World Health Organization (WHO), IPT is a neoplasm with a tendency for local recurrence and a very low rate of metastasis.²¹ Hussong et al. confirmed that IPT tumors testing positive for p53 showed recurrence or malignant transformation.²² However, Yamamoto et al. do not support the theory that p53 plays a major role in the pathogenesis of IPT.²³

Most IPTs require surgery to obtain definite diagnosis and cure. Complete resection is the preferred option, because incomplete excision has been shown to be a risk factor for recurrence.² According to Hakozi et al, pronounced inflammatory reaction is the main characteristic of IPT. Therefore, the logical option is anti-inflammatory treatment. In fact the trial of NSAID treatment may both confirm the diagnosis of IPT and at the same time successfully treating the tumor.²⁴ There is no uniform recommendation in the literature regarding when to use additional therapy.²⁵ Steroid therapy has been tried for cases in whom diagnosis is made preoperatively, but the outcome is not significant.²⁶ Other adjuvant therapy like radiotherapy and chemotherapy can also be tried. However, additional therapy for IPT that show a high risk of recurrence indicated by ill-defined margins or intra-abdominal, mesenteric, omental, and retroperitoneal localizations is strongly recommended.¹

Recurrences are documented in 18-40% of the cases and appear to be more frequent in the extrapulmonary lesions, which are larger than 8 cm and locally invasive.²⁷ Unfortunately, there are no definitive clinical, histopathological, or genetic features to predict recurrence or metastasis though one of the recent studies suggests that reactivity of ALK to be a favorable prognostic indicator.³ Novosel et al suggest a palette of immunohistochemical agents with p53 or ALK in addition to standard immunohistochemical procedures for prognostic stratification. Negative p53 expression or positive ALK expression should be considered as a favorable IPT diagnosis.¹⁰

A year after surgical IPT resection, our patient is well, with no signs of recurrence. We suggest long term follow up for this patient.

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