

Inflammatory myofibroblastic tumour presenting as a gastric submucosal tumour

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A 56-year-old male presented to our gastroenterology department with a 1-month history of abdominal discomfort. He had a 5-year history of interstitial lung disease and was regularly taking pirfenidone, prednisone, cyclosporine, and hydroxychloroquine. He denied any family history of malignancy. Laboratory tests, electrocardiography, and physical examination revealed no significant abnormalities. Gastroscopy identified a 40-mm submucosal tumour (SMT) in the gastric angle (Fig 1a). Abdominal computed tomography revealed an SMT with intraluminal growth (Fig 1b). Endoscopic ultrasound gastroscopy showed a well-defined and hypoechoic lesion originating from the muscularis propria (Fig 1c). The SMT was completely resected en bloc via endoscopic submucosal excavation (ESE) [Fig 1d]. Post-ESE histopathological examination revealed the tumour to be composed of spindle fibroblasts arranged in fascicles, with a background of lymphocytes, plasma cells, and some eosinophilic

infiltration (Fig 2a and b). Immunohistochemical staining showed tumour cells positive for vimentin (Fig 2c) and CD34 (cluster of differentiation 34) [Fig 2d]. The frequency of Ki-67 positive proliferating cells was very low (1%). CD117, DOG-1, S100, SOX-10, SMA, desmin, and calponin were all negative. Histopathological and immunohistochemical findings confirmed the diagnosis of an inflammatory myofibroblastic tumour (IMT).

Inflammatory myofibroblastic tumour is a rare type of mesenchymal tumour, first reported in the lungs in 1937.¹ Primary gastric IMT is extremely rare and its biological behaviour remains poorly understood. Due to its non-specific endoscopic and radiographic features, it is challenging to differentiate from other SMTs. On immunohistochemistry, IMT is positive for anaplastic lymphoma kinase (ALK), vimentin, and CD34, and negative for S-100, DOG1 (discovered on gastrointestinal stromal tumours [GIST] 1), and CD117 (cluster of differentiation 117).

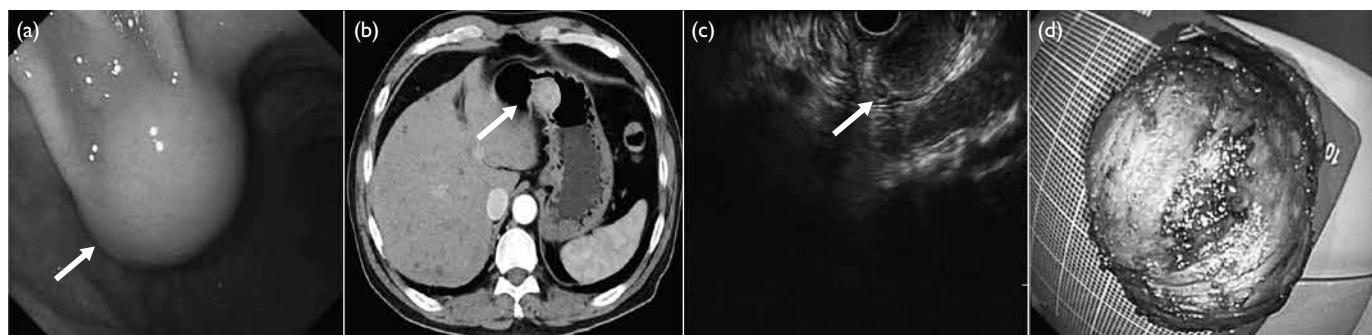


FIG 1. (a) The submucosal tumour is located at the gastric angle (arrow). (b) Abdominal computed tomography shows a mass in the gastric angle (arrow). (c) Endoscopic ultrasound reveals a hypoechoic lesion (arrow). (d) The submucosal lesion was completely resected

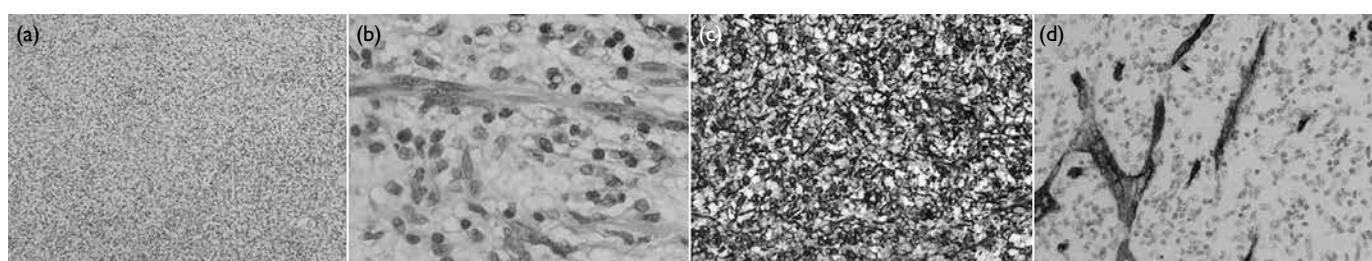


FIG 2. (a, b) Histopathological examination of the postoperative specimen showed infiltration of lymphocytes, plasma cells, and some eosinophils (a: haematoxylin and eosin, $\times 100$; b: haematoxylin and eosin, $\times 400$). Immunohistochemistry was positive for (c) vimentin ($\times 200$) and (d) CD34 ($\times 200$)

Gastrointestinal stromal tumours, which are also composed of fascicular spindle cells, can be easily confused with IMTs. However, GISTs are strongly positive for CD117, DOG1, and CD34, but negative for ALK.² Unlike IMT, an inflammatory background is not typical in GISTs. Another differential diagnosis is an inflammatory fibroid polyp, a SMT composed of spindle cells and inflammatory cell infiltration, predominantly eosinophils. These lesions are usually CD34-positive and CD117-negative. Anaplastic lymphoma kinase positivity is helpful in diagnosing IMT but is detected in only 50% to 60% of cases.³ Anaplastic lymphoma kinase negativity has been associated with a higher risk of distant metastasis,⁴ therefore long-term follow-up is required. Although our case was ALK-negative, the histological features were typical of IMT.

Endoscopic submucosal excavation can completely excavate the tumour in the muscularis propria along the lesion's margin.⁵ The procedure is comparable to traditional endoscopic submucosal dissection, with the main difference being the depth of dissection. No evidence of recurrence was observed in our patient during 3 years of follow-up. To the best of our knowledge, this is the first report of gastric IMT treated with ESE.

Author contributions

Concept or design: L Ren.

Acquisition of data: S Li.

Analysis or interpretation of data: H Zhang.

Drafting of the manuscript: H Zhang, S Li.

Critical revision of the manuscript for important intellectual content: L Ren.

All authors had full access to the data, contributed to the study, approved the final version for publication, and take responsibility for its accuracy and integrity.

Conflicts of interest

All authors have disclosed no conflicts of interest.

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Ethics approval

This study was conducted in accordance with the Declaration of Helsinki and was approved by the Ethics Committee of The First People's Hospital of Lianyungang, China (Ref No.: LW-20230612001-01). Informed patient consent was waived by the Committee due to the retrospective nature of the study, with the patient anonymised and no identifiable information included.

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