Resolution of eosinophilic gastroenteritis after resection of uterine leiomyomas

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ABSTRACT

Eosinophilic gastrointestinal disorders (EGIDs) primarily affect the gastrointestinal tract. EGIDs have a broad spectrum of presentations, characterised by prominent eosinophilic infiltration through a variable depth in the gastrointestinal tract in the absence of a known cause for eosinophilia. EGIDs include eosinophilic oesophagitis, eosinophilic gastritis, eosinophilic gastroenteritis, eosinophilic enteritis and eosinophilic colitis. Here, we report EGID in a woman who had co-existing uterine leiomyomas. Her EGID resolved after resection of the leiomyomas. She remained asymptomatic on follow-up 13 months after the myomectomy, with resolution of the eosinophilic infiltrate in the gastrointestinal tract.

Keywords: eosinophilic gastrointestinal disorder, leiomyomas, peripheral eosinophilia

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INTRODUCTION

Eosinophilic gastrointestinal disorders (EGIDs) primarily affect the gastrointestinal tract. EGIDs have a broad spectrum of presentations, which are characterised by prominent eosinophilic infiltration through a variable depth in the gastrointestinal tract in the absence of a known cause for eosinophilia.[1-3] EGIDs include eosinophilic esophagitis, eosinophilic gastritis, eosinophilic gastroenteritis, eosinophilic enteritis and eosinophilic colitis. Diagnosis of EGIDs is based on clinical symptoms, demonstration of eosinophilic infiltration of one or more areas of the gastrointestinal tract from the oesophagus to the colon, or characteristic radiologic findings with peripheral eosinophilia in the absence of parasitic and extraintestinal diseases.[4-6] EGIDs are usually recurrent and chronic, and their histological resolution is slow.[7] The optimal treatment for EGID is still uncertain, and there has been no consensus regarding it. Here, we report a case of EGID in a woman who presented with co-existing uterine leiomyomas; her EGID resolved after resection of the leiomyomas.

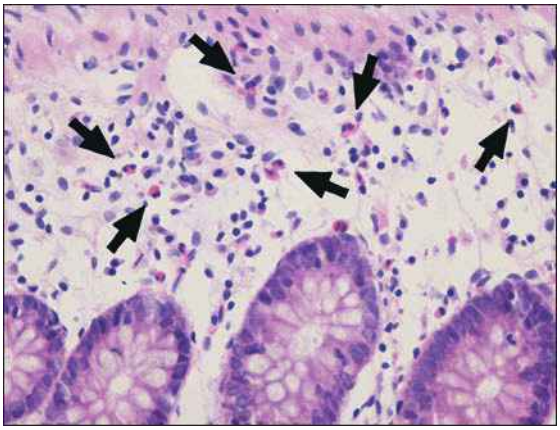

Fig. 1 Photomicrograph of colonic biopsy shows eosinophilic infiltrate (arrows) (Haematoxylin & eosin, × 400).

CASE REPORT

A previously healthy 45-year-old Chinese woman presented with epigastric discomfort for two months. She also complained of diarrhoea five times a day during the same period. Clinical examination was unremarkable except for pallor. Complete blood picture showed hypochromic, microcytic anaemia, with haemoglobin 9.6 (normal range 11.5–15.5) g/dL, mean corpuscular haemoglobin 25.1 (normal range 27.0–35.0) pg and mean corpuscular haemoglobin concentration 30.9 (normal range 32.0–36.0) g/dL. In addition, the patient had a raised eosinophil count of 15.3% (normal range 0.0%–6.5%), with an absolute eosinophil count of 1.04 k/uL. She also had low serum iron of 3.2 (normal range 6.6–30.4) µmol/L, low serum ferritin of 5 (normal range 4–204) ng/ml and a high serum total iron binding capacity of 95 (normal range 44–80) µmol/L.

The patient's liver biochemistry, urea and electrolytes were all unremarkable. Repeated stool tests for ova and parasites (four samples) were negative. Anti-neutrophilic cytoplasmic antibody, anti-nuclear antibody, complement 3, complement 4, immunoglobulin patterns, rheumatoid factor, anti-extractable antibody, anti-cardiolipin antibody, immunoglobulin E, chest radiograph, urine for microscopy and repeated stool for occult blood (three samples) were all unremarkable. Upper endoscopy revealed furrowing of the whole oesophagus with multiple white papules throughout. Multiple nodular-like lesions were observed at the first and second part of the duodenum. Multiple

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biopsies of the mid-oesophagus showed extensive eosinophilic infiltrates in the stroma, with more than 15/high-power field (HPF). Multiple biopsies of the first part of the duodenum also revealed eosinophilic infiltrates in the stroma. Colonoscopy showed a normal-looking mucosa up to the terminal ileum. Multiple biopsies taken from the whole colon and terminal ileum showed minimally inflamed terminal ileum and colonic mucosa with increased eosinophils (> 18/HPF) seen throughout the whole colon and terminal ileum (Fig. 1).

In order to rule out secondary EGID due to diseases such as lymphoma, lymphangiectasia, Hodgkin's disease or carcinoma,[2] a whole body positron-emission tomography computed tomography was performed, which revealed a leiomyoma at the inferior aspect of the uterus (3.0 cm × 3.0 cm in size). The leiomyoma had intense 18fluorodeoxyglucose (18FDG) uptakes with a maximum standardised uptake value of 11.7. Transvaginal ultrasonography of the pelvis confirmed the presence of an intracavitary submucosal leiomyoma of size 2.0 cm × 1.3 cm × 2.1 cm, a left anterolateral intramural leiomyoma of size 1.6 cm × 1.7 cm × 1.7 cm and two intramural leiomyomas measuring 1.5 cm and 1.5 cm in size, respectively.

Allergy testing was unremarkable. The patient was commenced on an elemental diet, proton-pump inhibitor, ketotifen, topical fluticasone, zyrtec and monteleukast. However, her epigastric discomfort, diarrhoea and peripheral eosinophilia failed to improve despite ten weeks of medical therapy. Although she was persistently symptomatic, she did not wish to be started on systemic corticosteroid. Furthermore, despite taking oral iron tablet replacement at 600 mg/day, she was still anaemic from her menorrhagia (haemoglobin of 9.0 g/dL). Thus, ten weeks after she was initially started on medical therapy for EGID, laparoscopic and hysteroscopic myomectomy was performed for treatment of her leiomyomas in order to control her menorrhagia.

Post surgery, the patient's peripheral eosinophilia returned to normal, with resolution of her epigastric discomfort and diarrhoea. Ketotifen, topical fluticasone, zyrtec and montelukast were discontinued four weeks after the myomectomy. She remained asymptomatic on follow-up 13 months after the myomectomy, with resolution of the eosinophil infiltrate in the oesophagus, duodenum and colon.

DISCUSSION

EGID was first described in the 1930s and has since garnered increasing attention in the last ten years.[1,2] The incidence of EGID is estimated to be 20–60 per million/year, with studies showing an increasing prevalence in the last 16 years.[8] However, the low incidence of EGID has precluded any large prospective randomised therapeutic trials. This has resulted in a lack of general consensus on the optimal therapy for EGID. Some experts have suggested that clinicians should initially consider allergy avoidance, and if this fails to improve symptoms, topical glucocorticoid should be initiated. Systemic glucocorticoids should be administered if there is no response to topical glucocorticoids.[9] An elemental diet with the aim of avoiding protein antigen exposure has also been recommended.[9] However, our patient did not wish to be started on systemic glucocorticoid, although her symptoms and peripheral eosinophilia were unresponsive to elemental diet, proton-pump inhibitor, ketotifen, topical fluticasone, zyrtec and monteleukast. Surprisingly, her symptoms, eosinophilic infiltrate in the oesophagus, duodenum and colon as well as peripheral eosinophilia resolved spontaneously after the myomectomy. No recurrence of EGID was detected even after all therapy had ceased.

Resolution of EGID due to medications in this patient was unlikely; there was no improvement in her symptoms and peripheral eosinophilia during the ten weeks of medical therapy. Furthermore, although the natural history of EGID was waxing and waning, the patient did not develop any recurrence of symptoms or peripheral eosinophilia even after 13 months post-myomectomy. This may mean that a true association exists between uterine leiomyomas and development of peripheral eosinophilia and EGID. Resection of the leiomyomas may have resulted in sustained remission of the EGID.

Peripheral eosinophilia associated with uterine leiomyomas was first described by Buka in 1965. In his report, peripheral eosinophilia resolved after hysterectomy for treatment of the uterine leiomyomas.[10] The association between uterine leiomyomas and peripheral eosinophilia is, however, unclear. Buka postulated that peripheral eosinophilia in women with uterine leiomyomas may be the result of an autoimmune response to the myomas.[10] As the pathogenesis of EGID is associated with increased eosinophils in the gastrointestinal tract, the decrease in peripheral eosinophil count after myomectomy in this case may have resulted in the resolution of EGID, thus suggesting an association between peripheral eosinophilia and uterine leiomyomas.

In conclusion, this is the first report of EGID associated with uterine leiomyomas, which resolved after subsequent myomectomy. More studies to determine whether and how such an association exists are warranted.

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