Spontaneously Identified Gastric Sarcoidosis: a Report of Three Cases

K KAWAURA1, T TAKAHASHI2, K KUSAKA1, J YAMAKAWA1, T ITOH1 AND T KANDA1

1Department of General Medicine, Kanazawa Medical University, Ishikawa, Japan; 2Division of Infectious Diseases, Advanced Clinical Research Centre, Institute of Medical Science, University of Tokyo, Tokyo, Japan

Sarcoidosis is a systemic granulomatous disease, frequently involving the lungs, lymph nodes, eyes and skin. Gastric sarcoidosis is very rare. We report three patients diagnosed initially with gastric sarcoidosis. Two had no other identified involvement, and one had involvement of the lungs and hilar lymph nodes. Gastroscopy was performed because of abdominal discomfort or as a follow-up examination for partial gastrectomy. This revealed atrophic lesions with nodular mucosal changes in the antrum and granular mucosa, and residual gastritis was found at the site of gastroduodenal anastomosis. Non-caseating epitheloid-cell granulomas were found in all patients following histological analysis. Gastroscopy and histopathological findings in gastric mucosal biopsy samples from suspicious sites are essential in establishing an accurate diagnosis of gastric sarcoidosis.

KEY WORDS: GASTRIC SARCOIDOSIS; GASTROSCOPY; EPITHELOID-CELL GRANULOMA; JAPAN

Introduction

Sarcoidosis is characterized by systemic non-caseating epitheloid-cell granulomas. The Eighth Japanese National Survey on Sarcoidosis1 reported that sarcoidosis most frequently occurs in the bilateral hilar lymph nodes (in 75.6% of cases), eyes (49.0%), lungs (29.2%), skin (18.5%), and subcutaneous lymph nodes (7.7%), although any organ can be involved. Gastric sarcoidosis is a very rare involvement in this illness but Japanese cases have been described.2 Symptoms of gastric sarcoidosis can be pain in the epigastrium, nausea, vomiting and haematemesis, but the condition is often asymptomatic.3

We report two patients with gastric sarcoidosis alone, and one patient found to have involvement of the lungs and bilateral hilar lymph nodes after an initial diagnosis of gastric sarcoidosis.

Case reports

PATIENT 1

A 56-year-old woman complained of mild abdominal discomfort when fasting. Skin reaction to purified protein derivative (PPD) was negative, and serum levels of angiotensin-converting enzyme (ACE; 17.2 IU/l; normal range, 8.3 – 21.4 IU/l) and blood calcium concentration were within the normal ranges. A serological test for syphilis was negative.

Verrucoid lesions at the antrum of the stomach were found following endoscopic analysis of the upper gastrointestinal tract.
Gastroscopy revealed antral atrophic lesions of the antrum anterior wall (Fig. 1A) together with multiple nodular changes. Biopsies were taken from the nodular lesions, and histopathological analysis using haematoxylin and eosin stain showed non-caseating epitheloid-cell granulomas including giant cells. These were surrounded by infiltration of lymphocytes and proliferation of fibroblastic cells (Fig. 1B). Staining for mycobacterium was negative. Radiological analysis of the bowel section from rectum to caecum, following a barium enema, showed no evidence of stenosis, ulcers or polypoid lesions, eliminating a diagnosis of Crohn’s disease.

Chest roentgenogram and computed tomogram showed no evidence of bilateral hilar lymphadenopathy (BHL) or diffuse pulmonary infiltrates. An abdominal ultrasound scan showed normal findings. Bronchoscopy revealed no mucosal lesions, and the bronchial tissue did not contain granulomas. No cutaneous lesions or abnormalities of the optic fundus were found.

The patient was diagnosed with localized gastric sarcoidosis without involvement of other organs. Abdominal symptoms gradually resolved with antacid treatment.

PATIENT 2
A 54-year-old female complained of mild abdominal discomfort after eating. Skin reaction to PPD was negative, and serum ACE level (10.1 IU/l) and blood calcium concentration were within normal ranges. A serological test for syphilis was negative.

Gastroscopy revealed antral atrophic lesions of the lesser curve of the antrum (Fig. 1C) and multiple nodular mucosal changes. Histopathological findings in tissue from the nodular lesions demonstrated non-caseating epitheloid-cell granulomas (Fig. 1D) and no mycobacteria.

Radiological examination of the lower bowel, chest roentgenogram, chest computed tomogram and abdominal ultrasound scan showed normal findings. Bronchoscopy revealed no mucosal lesions, and the bronchial tissue did not contain granulomas. No cutaneous lesions or abnormalities of the optic fundus were found.

The patient was diagnosed with localized gastric sarcoidosis without involvement of other organs. Abdominal symptoms gradually resolved with antacid treatment.

PATIENT 3
Gastroscopy was performed on a 42-year-old man as follow-up after a partial gastrectomy for gastric ulcer 12 years earlier. He had not reported any symptoms. The endoscopy revealed granular mucosal lesions at the site of gastroduodenal anastomosis (Fig. 1E) and residual gastritis of the oedematous mucosa. Histopathological findings in tissue from the granular lesions demonstrated non-caseating epitheloid-cell granulomas including giant cells (Fig. 1F). Mycobacteria were absent.

Skin reaction to PPD was negative, and serum levels of ACE were just outside the normal range (21.9 IU/l). Blood calcium concentration was within the normal range and a serological test for syphilis was negative. A chest roentgenogram and computed tomogram showed evidence of BHL and diffuse bilateral pulmonary infiltrates, suggesting pulmonary involvement of sarcoidosis. A gallium-scintigram revealed abnormal uptake in the bilateral lung fields consistent with the findings of the chest images. Radiological examination of
FIGURE 1: Gastroscopic and histopathological findings. (A) Gastroscopy in patient 1 revealed multiple nodular mucosal lesions of the antrum anterior wall. (B) Histopathology of the biopsy specimen taken from patient 1 shows non-caseating epitheloid-cell granulomas including giant cells, surrounded by the infiltration of lymphocytes and proliferation of fibroblastic cells. (C) Gastroscopy in patient 2 revealed multiple nodular mucosal lesions of the lesser curve of the antrum. (D) Histopathology of the biopsy specimen taken from patient 2 showed non-caseating epitheloid-cell granulomas. (E) Gastroscopy in patient 3 revealed granular mucosal lesions at the site of gastroduodenal anastomosis. (F) Histopathology of the biopsy specimen taken from patient 3 demonstrated non-caseating epitheloid-cell granulomas including giant cells.
Gastric sarcoidosis

the lower bowel, abdominal ultrasound scan and computed tomogram of the abdomen showed normal findings. No cutaneous lesions or abnormalities of optic fundus were observed.

The patient was considered to have gastric sarcoidosis with the involvement of the lungs and hilar lymph nodes. He continued to receive follow-up gastroscopies, without specific therapy for sarcoidosis.

Discussion

Analysis of patients already affected by cutaneous and pulmonary sarcoidosis found gastric involvement in 9.4% (three of 32). Stomach sarcoidosis was identified at a frequency of 0.02% during 5000 routine endoscopies of the upper digestive tract in one report, indicating that sarcoidosis can be present in asymptomatic patients, as in the case reported here.

A previous investigation into gastric sarcoidosis showed that 75% of patients presented with pain, and 25% had bleeding. Gastric haemorrhage from a sarcoid stomach ulcer was reported in one patient, and the ulcer resolved within 5 months with conventional antacid treatment. Another case of gastric sarcoidosis with haemorrhage was described after diagnostic gastrectomy. This patient died, and apart from the stomach, no other areas of granulomatous tissue were identified during the autopsy.

Gastric sarcoidosis appears as diffuse mucosal hyperaemia, nodular lesions and ulcers on endoscopy, but they are extremely variable. In several reports, a single polypoid appearance or mucosal lesions resembling linitis plastica are also described. The latter lesions were shown, by gastroscopic ultrasonography, to result from abnormal hypertrophy of the third hyperechoic layer. Gastric lesions were localized to the mucous membrane in all 17 cases of sarcoidosis evaluated by Ito et al., although the granulomas spread outside the mucous membrane. The epitheloid-cell granulomas could possibly therefore be detected by mucosal biopsy. Gastroscopy, biopsy and histopathological findings from the mucosal samples obtained from suspicious sites are accurate, and essential, in establishing the diagnosis of gastric sarcoidosis.

Clinicians should consider mycobacterial infections, syphilis, histoplasmosis, gastric cancer and lymphoma in the differential diagnosis of gastric sarcoidosis. A paediatric case of gastric sarcoidosis, initially thought to be Crohn’s disease, has been reported. Gastric sarcoidosis was confirmed when the region of the bowel from terminal ileum to rectum was found to be normal. This emphasizes the need to check this part of the bowel by barium radiological examination or colonoscopy.

Surgery is required for 50% of patients with gastric sarcoidosis, while improvement of symptoms is seen in 66% of those taking corticosteroids. In another report, 30 mg/day prednisolone was administered orally to a patient with gastric sarcoidosis in decreasing doses; the maintenance dose was 5 mg/day. Digestive symptoms quickly disappeared, the patient gained weight and returned to work, and macroscopic features in the gastric mucosa regressed. Epitheloid-cell granulomas persisted, however, but to a lesser extent, despite treatment.

One study has evaluated 17 Japanese patients with gastric sarcoidosis. All patients were aged between 20 and 60 years, and there was a female to male ratio of 2:1. The prognosis for all patients was good following steroid treatment.

Acknowledgements

The authors thank Drs Akio Mukawa and Shizuo Odashima for their critical review of the manuscript.
Gastric sarcoidosis

References

Address for correspondence
Dr T Kanda
Department of General Medicine, Kanazawa Medical University,
Daigaku 1-1, Uchinada-machi, Kahoku-gun, Ishikawa 920-0293, Japan.
E-mail: kandat@kanazawa-med.ac.jp