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CASE REPORT

Russell body gastritis/duodenitis: A case series and description of immunoglobulin light chain restriction

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Summary Russell body esophago-gastro-duodenitis is an unusual form of chronic inflammation, with only 22 cases being reported in PubMed. However, the prevalence and clinical significance remain unknown. This report describes the clinico-pathological characteristics of nine cases of Russell body gastritis (RBG) and one case of Russell body duodentitis (RBD), with nonspecific endoscopic appearance. The Mott cells (plasma cells with Russell bodies) showed κ light chain restriction in eight gastritis cases and λ light chain restriction in the duodentitis case, and there were no histological features that suggested lymphoma. Thus, a diagnosis of monoclonal RBG/RBD was made. *Helicobacter pylori* infection was found in 55.6% of RBG cases and in the RBD case. And, the clinical follow-up evaluations were uneventful. This report is the first study to describe this benign disease entity with monoclonality on a large-scale basis. In addition, the monoclonality of Mott cells cannot be used as evidence of an existing neoplastic lesion, and taken together, these findings may indicate a reactive process.

Introduction

The infiltration of plasmocytes is a hallmark of gastric chronic inflammation. Under the stimulation of antigens, e.g., *Helicobacter pylori* (*H. pylori*), plasmocytes produce immunoglobulins (Igs) involved in the immune response. The aggregation of Igs within the rough endoplasmic reticulum of plasmocytes results in Russell body formation [1]. In 1998, Tazawa and Tsutsumi [2] first described the rare benign disease, Russell body gastritis (RBG), in which the lamina propria of the gastric mucosa is excessively infiltrated by plasma cells containing Russell bodies (RBs). Subsequently, Russell body Barrett's esophagitis (RBBO) [3] and Russell body duodenitis (RBD) [4] were reported, respectively. To date, there are only 22 Russell body esophago-gastro-duodenitis cases reported in PubMed, including 18 cases of

Abbreviations: RBG, Russell body gastritis; RBD, Russell body duodentitis; RBBO, Russell body Barrett's esophagitis; RBs, Russell bodies; Igs, immunoglobulins; HIV, human immunodeficiency virus.

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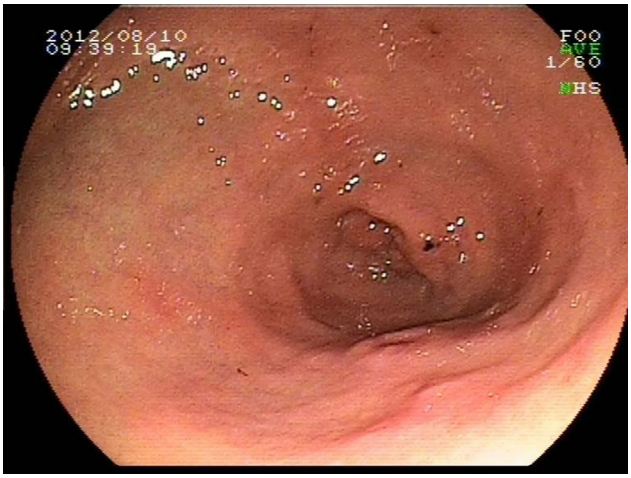

Figure 1 Upper gastrointestinal endoscopic findings of case No. 6. Endoscopic image shows multiple flat and raised erosions in the antrum of the stomach.

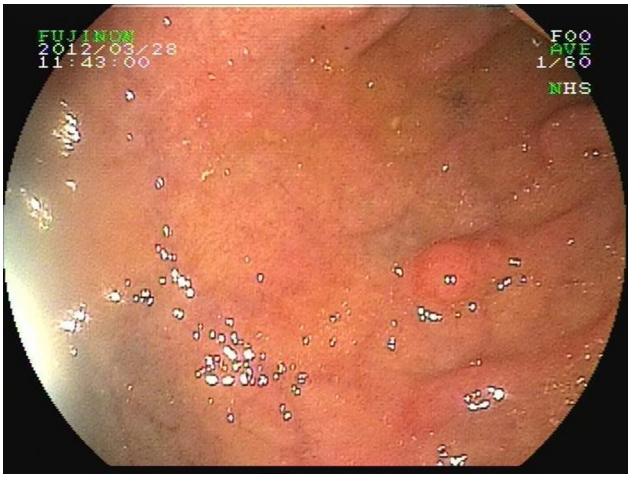

Figure 2 Upper gastrointestinal endoscopic findings of case No. 10. Upper endoscopy revealed multiple nodules within the duodenal bulb, which were biopsied.

RBG [2,5—19], two cases of RBBO [3,20] and two cases of RBD [4,21]. In this study, we describe nine RBG cases and one RBD case, and we review the related literature.

Materials and methods

Between 2007 and 2013, 10 cases of chronic gastroduodenitis with an accumulation of numerous RBs and plasma cells containing RBs in the lamina propria were detected using the keyword ''Russell body'' in archives, including nine cases of gastritis and one case of duodenitis, and no cases of Barrett's esophagitis were found. Clinical records and endoscopic and pathologic findings were retrospectively obtained.

All biopsy specimens were orientated on filter paper and immediately fixed in formol saline. Paraffin-processed sections were cut and stained with hematoxylin and eosin, Warthin-Starry staining, and immunohistochemistry, respectively. According to the Sydney system for the classification of gastritis, the biopsy specimen was evaluated semi-quantitatively. Immunohistochemical staining using AE1/AE3, CD45, CD79α, CD20, CD138, CD68, κ light chain and λ light chain antibodies (Invitrogen Corporation, California, USA) was performed using the conventional avidin-biotin-peroxidase complex method according to the manufacturer's instructions. All of the experiments were performed in triplicate. All patients signed their informed consent.

Results

A summary of the clinical and histological findings is provided in Table 1. The mean age was 60.80 ± 20.76 years (range 24—78 years), and the ratio of male-to-female was 6:4. Most patients presented with nonspecific gastrointestinal symptoms, and the esophago-gastro-duodenoscopic features were nonspecific (Figs. 1 and 2). Five cases underwent colonoscopy, three cases showed colonic polyps, and two cases were normal.

Histological examinations of gastric/duodenal biopsies revealed chronic gastritis/duodenitis with infiltration of lymphoplasmocytes and numerous mononuclear cells containing spherical eosinophilic globules in the lamina propria (Figs. 3 and 4). Polymorphonuclear leukocyte infiltration, glandular atrophy and intestinal metaplasia were observed in some cases. Under high power, mononuclear cells with eosinophilic globules were observed to contain large intracytoplasmic globules, which were mainly round, homogeneous, resembled the Russell body, and pushed the cell nucleus to the periphery (Figs. 3B and 4B). RBs were widely distributed in the stomach. Immunohistochemistry was performed to confirm the cell type. Sufficient biopsy tissue was absent in case No. 9 to permit immunohistochemical analysis. In all analyzed cases, mononuclear cells containing eosinophilic globules (RBs) showed CD45, CD79α, and CD138 expression but no immunoreactivity for AE1/AE3, CD20 and CD68, which confirmed that the cells were plasma cells, known as Mott cells. The morphological plasma cells exhibited polytypic light chain expression. However, the Mott cells in the cases of gastritis only showed κ light chain expression in their scant cytoplasm as a rim of RBs (Fig. 3C and D). RBs showed weak focally positive κ light chain expression in case Nos. 3, 5, 7 and 8. In the case of duodenitis, the Mott cells and RBs were strongly positive for λ light chain and no immunoreactivity for κ light chain was observed (Fig. 4C and D).

The lesions were focal and non-progressive. Absence of cellular atypia and mitotic activity in proliferating Mott cells and plasma cells, the lack of monocytoid cells and centrocyte-like cells, an absence of lymphoepithelial lesion, and the polyclonal pattern of the morphological plasma cells, ruled out lymphoma. In addition, serum electrophoresis was performed in case nos. 4 and 5, and no monoclonal peak was observed. Immunonegativity with AE1/AE3 ruled out signet-ring cell carcinoma. The diagnosis of monoclonal RBG and monoclonal RBD was made. None of the cases underwent repeat gastric biopsy, and clinical follow-up evaluations were uneventful in all cases.

Spiral-shaped bacteria consistent with *H. pylori* were observed on the gastric foveolar epithelial surface in case