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Russell Body Gastroesophagitis Concurrent With Barrett's Esophagus

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Abstract. *Background: Russell body gastroesophagitis is a rare entity characterized by the accumulation of immunoglobulins within the cytoplasm of plasma cells. Case Report: Here, we present the case of a 41-year-old male with history of gastroesophageal reflux disease who presented with nausea, vomiting, and altered mental status. Candida esophagitis was noted on upper endoscopy. After treatment, a surveillance endoscopy revealed salmon colored mucosa in the distal esophagus and mild gastric erythema. The biopsy confirmed Barrett's esophagus that was negative for dysplasia and mild chronic inactive gastritis. Interestingly, diffusely infiltrating Russell body-containing plasma cells (Mott cells) were present in the distal esophagus and extending into the gastric cardia. The Mott cells were highlighted on CD138 immunostaining and Periodic acid-Schiff stain. Immunostaining for cytokeratin AE1/AE3 was negative. There was no evidence of Helicobacter pylori organisms on the gastric mucosa. Conclusion: This is the first report on Russell body-containing plasma cells diffusely involving both esophagus and gastric cardia with concurrent Barrett's esophagus.*

Russell bodies are eosinophilic globular intracytoplasmic accumulations of nondegradable immunoglobulins in the endoplasmic reticulum of plasma cells (1, 2). It was first described in 1890 by William Russell (1). The term "Mott cells" has been used to describe these plasma cells with cytoplasmic accumulation of Russell bodies, which indicates that either the secretory or the degradative function is impaired in these plasma cells (3). The presence of Russell bodies and Mott cells is a rare finding in the gastrointestinal tract, and the first case was reported by Tazawa and Tsutsumi in 1998 (4). Most patients present with non-specific symptoms, and Russell bodies are usually incidental findings on biopsies. For the Russell bodies and Mott cells, gastric antrum is the most commonly reported location, and they're exceedingly rare in the esophagus that only 2 cases with Barrett's esophagus have been reported to date (5, 6). Here we report the first case on Russell body-containing plasma cells diffusely involving both esophagus and gastric cardia with concurrent Barrett's esophagus.

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Case Report

A 41-year-old male with past medical history of significant gastroesophageal reflux disease, asthma, mental retardation, schizophrenia and bipolar disorder presented to the emergency department for concerns of altered mental status. He had three episodes of non-bloody emesis and persistent hiccups with no complaints of abdominal pain. The patient was admitted to the hospital. Abdominal CT scan revealed an incidental intrapancreatic spenule. A biopsy during an upper endoscopy revealed candida esophagitis and mild chronic inactive gastritis. The patient was treated for Candida esophagitis and followed up with a surveillance endoscopy a month later.

On surveillance endoscopy, there was a segment of non-nodular salmon colored mucosa in the distal esophagus from 35 to 38 centimeters suggestive of Barrett's esophagus. Mild erythema was noted in the gastric cardia, fundus/body and antrum. Cold biopsies were taken from distal esophagus and gastric cardia, and sent to the pathology department for review.

The biopsy of the distal esophagus showed squamocolumnar mucosa with intestinal metaplasia, consistent with Barrett's esophagitis. No evidence of dysplasia was identified. Diffusely infiltrating plasma cells were identified in the lamina propria within the esophageal mucosa. These plasma cells had eccentric nuclei and intracytoplasmic eosinophilic globules (Russell bodies). These Russell body-containing plasma cells were highlighted on CD138 immunohistochemical staining and Periodic acid-Schiff (PAS) stain. Immunohistochemical staining for cytokeratin AE1/AE3 was negative (Figure 1). Histological

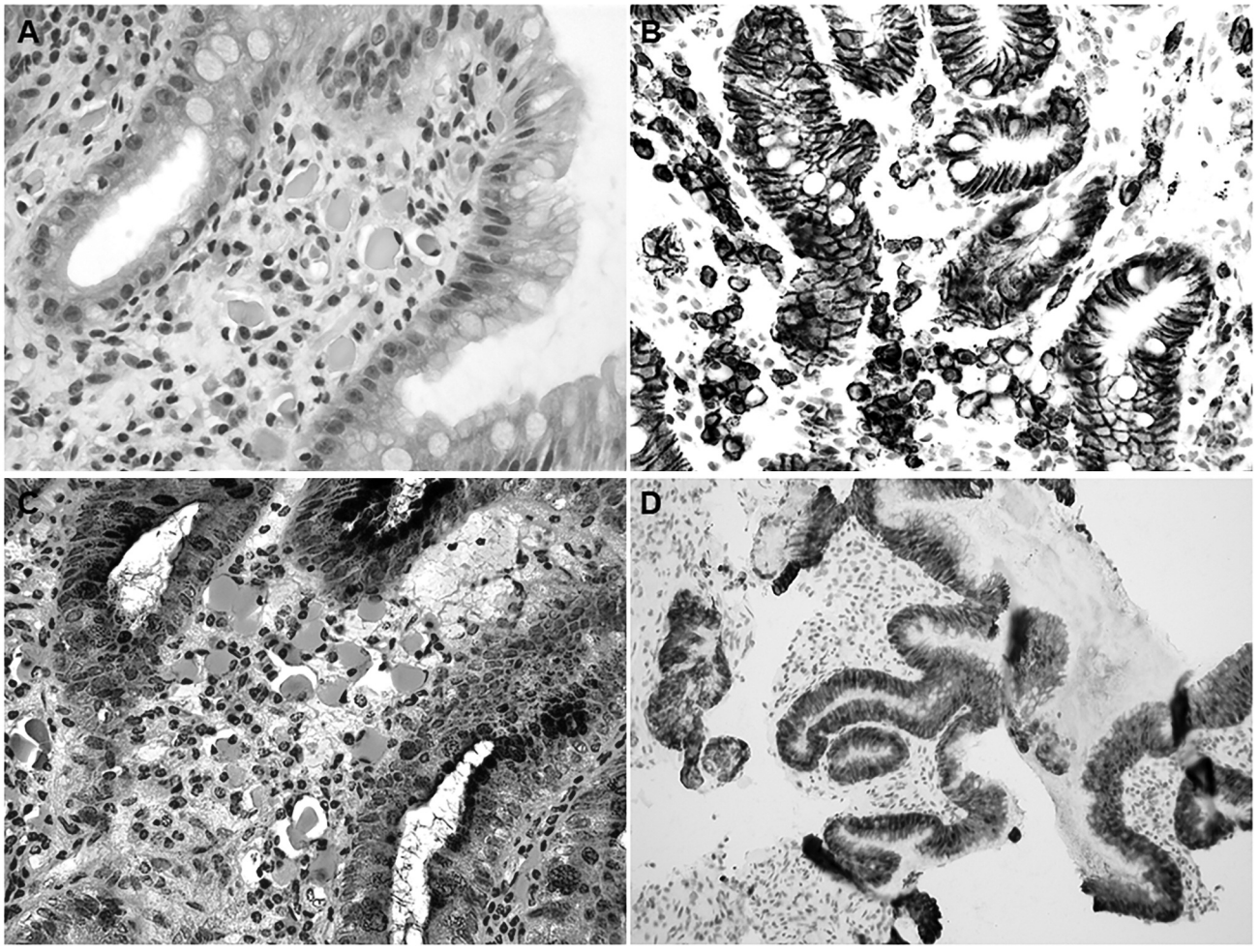

Figure 1. *Russell body Barret's esophagitis. (A) H&E stain; (B) Russell body plasma cells positive for CD138; (C) PAS stain highlights Russell body plasma cells; (D) Russell body plasma cells negative for cytokeratin AE1/AE3.*

examination of the gastric cardia biopsy showed mild chronic inactive gastritis, and there was no evidence of intestinal metaplasia or *Helicobacter pylori* organisms on the gastric mucosa. Interestingly, the infiltrating Russell body-containing plasma cells were also present in the lamina propria of the gastric mucosa, suggesting the direct extension of Mott cells from distal esophagus to proximal stomach. These Mott cells were also positive for CD138 and PAS, and negative for cytokeratin AE1/AE3 (Figure 2). The clinical presentation and histopathologic findings support the diagnosis of Russell body gastroesophagitis with coexisting Barrett's esophagus.

Discussion

Russell bodies are eosinophilic, non-degradable cytoplasmic inclusions within the polyclonal plasma cells (also known as Mott cells) (1, 2). Gastric antrum is the most commonly reported site for Russell body gastroenteritis, and the association with *Helicobacter pylori* infection is present in up to 82% of the Russell body gastritis patients (7). Complete regression of Russell body gastritis upon *Helicobacter pylori* eradication has been reported (8). Russell bodies may also be present in patients with autoimmune disorders, infections (such as HIV), hematopoietic tumors, and gastric cancer (9-12). Mott cells associated with inflammatory conditions are mostly polyclonal, but immunoglobulin light chain restriction has been reported in Russell body gastritis and duodenitis cases (13). Russell body gastritis can even present as a tumor like lesion (14). The morphology and infiltrating pattern of Mott cells can mimic gastric signet ring cell carcinoma on the biopsy, therefore it's important to be able to recognize this rare entity. A detailed work up is often necessary. Compared to signet ring cell carcinoma, Mott cells lack the features of malignancy such as nuclear atypia and mitosis. Immunohistochemical and special stains could be helpful to reach the right diagnosis.

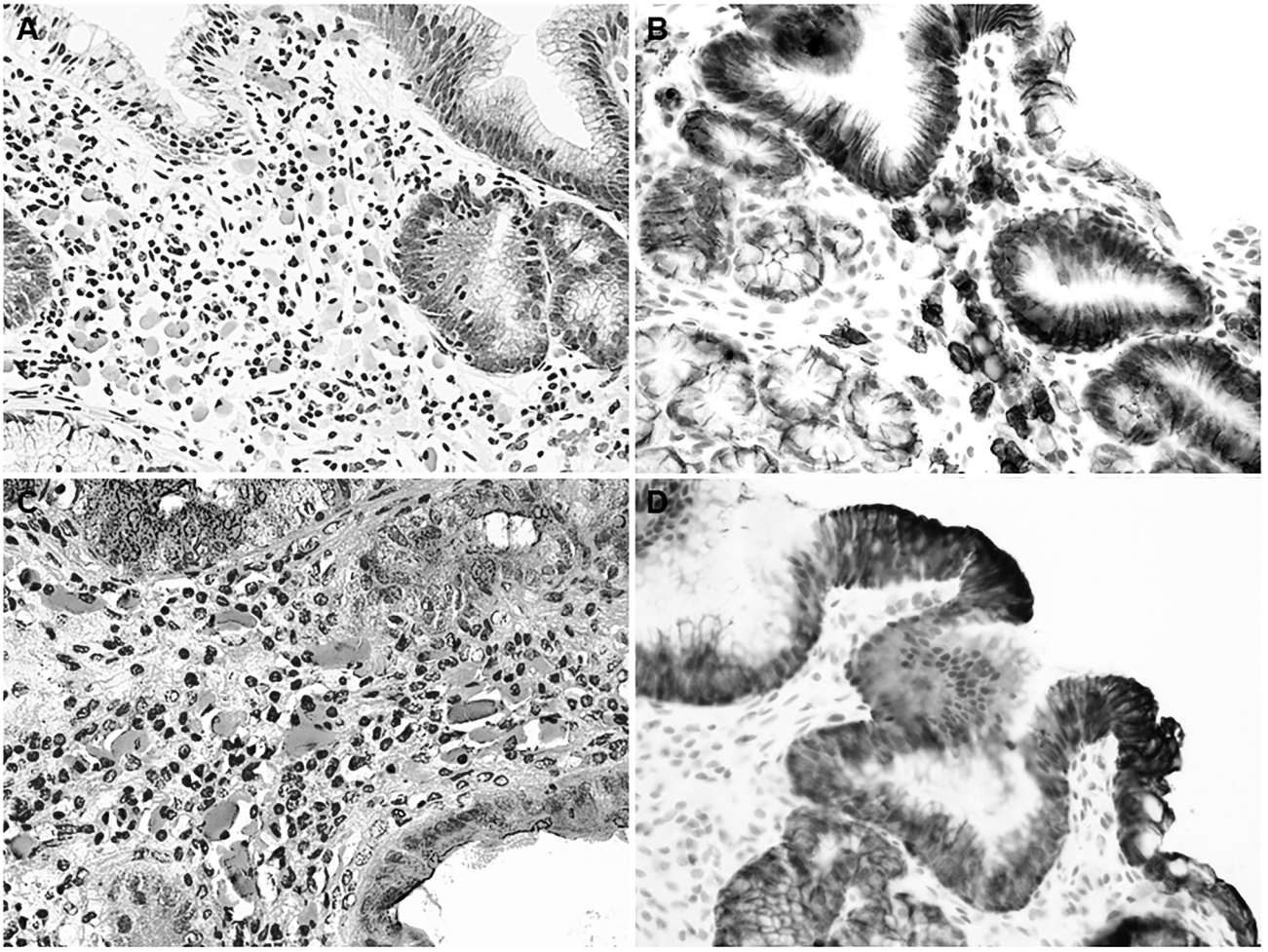

Figure 2. *Russell body gastritis. (A) H&E stain; (B) Russell body plasma cells positive for CD138; (C) PAS stain highlights Russell body plasma cells; (D) Russell body plasma cells negative for cytokeratin AE1/AE3.*

Our case is unique. Barrett's esophagus is a precancerous medical condition frequently associated with gastroesophageal reflux disease. Its potential association with ingestion of nitrites-containing food products has also been proposed (15). Here we present the first case report on Russell body-containing plasma cells extending from esophagus to stomach with the coexisting Barrett's esophagus. Notably, the first case report on Russell bodies Barrett's esophagus claimed that the presence of Russell bodies was not a widespread phenomenon and instead they were only localized to the Barrett's mucosa. In their study, Russell bodies were not identified in the gastric mucosa and in the urinary bladder (5). The underlying cause for this discrepancy is at present unknown. Russell body gastritis is commonly associated with *Helicobacter pylori* infection, but apparently this is not the case for our patient. In the esophagus, Russell body has been found in relation to long-standing gastroesophageal reflux disease. Russell body esophagitis has also been associated with Candida esophagitis in one report (14). Interestingly, the patient in our case also had a history of Candida esophagitis status post treatment. Whether Candia infection with concurrent Barrett's esophagus could promote the extension of Russell bodies remains unclear. Vice versa, whether Russell body gastroesophagitis could promote the development of Barrett's esophagitis also awaits further investigation.

The clinical significance of Russell body plasma cell infiltration has not yet been identified. Clinicopathological correlation is of utmost significance along with endoscopic surveillance. Overall, Russell bodies and Mott cells are incidental benign findings. In esophagus, they may represent a sequela of healed esophageal injury or a metaplastic change in association with Barret's esophagus. Notably, rare cases of Russell bodies associated with certain malignancies including gastric signet ring cell carcinoma and Epstein-Barr virus-associated gastric carcinoma have been reported (10,

11). A long-term follow up and endoscopic surveillance might be applicable. Interestingly, the Mott cells associated with neoplastic process are different from those with inflammatory conditions. Specifically, the Mott cells associated with neoplasms are usually monoclonal and stain positive for Concanavalin A (Con A) by lectin immunohistochemistry. In contrast, the Mott cells associated with chronic inflammatory conditions are usually polyclonal and do not stain for Con A (16).

In conclusion, this is the first reported case of Russell body Barrett's esophagitis with concurrent Russell body gastritis. Although it has been considered as a benign condition, rare reported association with certain malignancies raises the potential clinical value for a long-term endoscopic surveillance. It's also important to be aware of this rare entity to avoid diagnostic pitfalls.

Conflicts of Interest

The Authors declare no competing interests regarding this study.

Authors' Contributions

JA and FY performed the histological examination, researched the literature, and wrote the manuscript. JN researched the literature and edited the manuscript. All Authors read and approved the final manuscript.

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