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# Eosinophilic Gastroenteritis with Serosal Involvement and Cyclic Neutropenia

## *Serozal Tutulumlu Eozinofilik Gastroenterit ve Siklik Nötropeni*

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**Özet**

Eozinofilik gastroenterit gastrointestinal kanalın farklı alanlarının eozinofilik infiltrasyonuna bağlı olarak farklı klinik tabloların görüldüğü nedeni bilinmeyen bir hastalıktır. Eozinofilik gastroenteritin seyrinde görülen hematolojik bulgu sadece eozinofilidir. Biz eozinofilik gastroenterit ve eozinofilik asit nedeniyle değerlendirilen ve siklik nötropeni görülen bir hastayı sunduk. Her ne kadar siklik nötropeni solid organ veya hematolojik malignensileri desteklerse de steroid tedavisi ile asit, nötropeni ve periferal eozinofilide düzelme olması tablonun eozinofilik gastroenterite bağlı olduğunu doğrulamıştır. Eozinofilik gastroenteritin seyrinde siklik nötropeni görülebileceği akılda tutulmalıdır.

**Anahtar Kelimeler:** eozinofilik gastroenterit, eozinofilik assit, siklik nötropeni, eozinofili, serozal tutulum.

**Abstract**

Eosinophilic gastroenteritis is a disease of unknown etiology presenting with different clinical pictures due to eosinophilic infiltration of different areas in gastrointestinal tractus. Eosinophilia is the sole hematologic finding seen in the course of eosinophilic gastroenteritis. We presented a case evaluated for eosinophilic ascites and diagnosed as eosinophilic gastroenteritis in which cyclic neutropenia was seen. Although cyclic neutropenia suggests solid organ or hematologic malignancy no sign of malignancy on investigations, improvement in ascites, peripheral eosinophilia and neutropenia with steroid therapy confirmed that this picture was completely due to eosinophilic gastroenteritis. It should be kept in mind that cyclic neutropenia may be seen during the course of eosinophilic gastroenteritis.

**Keywords:** eosinophilic gastroenteritis, eosinophilic ascites, cyclic neutropenia, eosinophilia, serosal involvement.

## Introduction

Eosinophilic gastroenteritis (EG) is a rare clinical entity characterised with gastrointestinal symptoms (GIS), peripheral eosinophilia when there is no other explanation for eosinophilia, eosinophilic infiltration and no involvement in other organ systems (heart, brain, kidney etc.). Clinical picture may vary with the layer of the gastrointestinal tract (e.g. mucosal, muscular, and serosal) involved. Eosinophilic ascites is the typical finding of serosal involvement (1). In this article we will discuss an EG case who presented with eosinophilic ascites.

## Case Report

A 28-old male was admitted to the hospital with the complaints of abdominal pain and distension lasting for a month. His medical history was unremarkable. On physical examination mild abdominal distension with positive fluid sign was noted but there was no peripheral edema. No other positive physical examination finding could be noted. Laboratory findings revealed leucocytosis (marked peripheral eosinophilia) and an elevated IgE level (292 kU/l, normal value < 100 kU/l). Laboratory and clinical findings are shown on Table–1. Ultrasonographic examination revealed moderate ascites. No finding suggesting portal hypertension, liver or renal abnormality could be detected. Abdominal paracentesis yielded exudative fluid with a serum-ascites albumin gradient of 0.6 mg/dl, white blood count of 1319 cells/mm³( 6% neutrophils, 4% lymphocytes, 90% eosinophils ( figure 1) ) and negative cultures. Computerized tomographic (CT) scan of abdomen showed ascites, extensive peritoneal mass and marked wall thickening of distal esophagus and the jejenum.

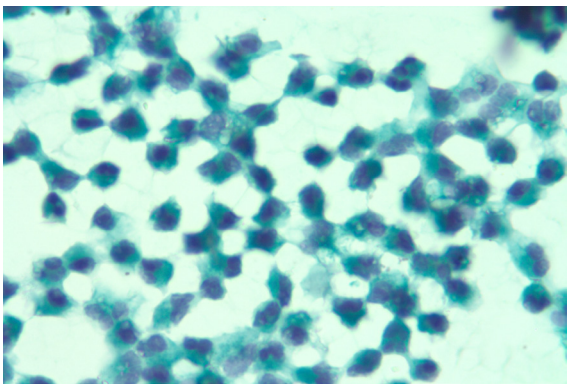

**Figure 1.** Eosinophilia in ascites fluid (Giemsa x 1000)

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At gastroscopy normal esophagus and marked bulbitis was observed (figure 2).

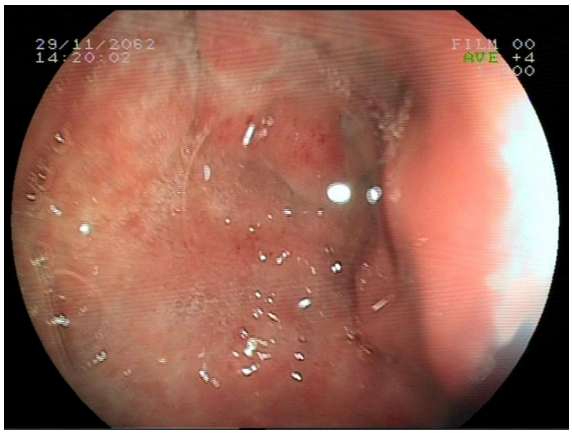


**Figure 2.** Nodular patern in bulbus.

Bulbar biopsy revealed inflammation with non-specific focal eosinophilic infiltration (figure 3).

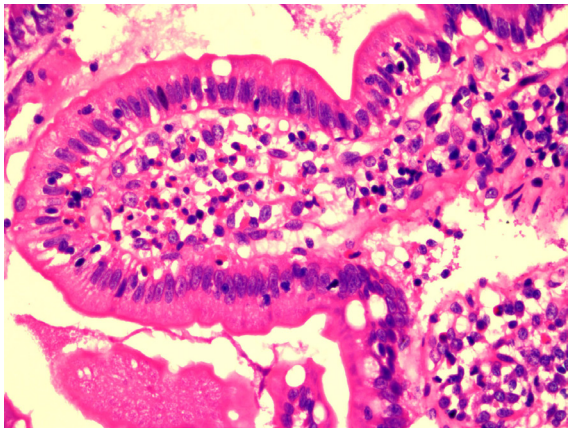

**Figure 3.** Inflammation with non-specific focal eosinophilic infiltration in bulbus biopsy (HE x 40)

Normal colonic mucosa and terminal iletis was seen at colonoscopy. Because of peritoneal involvement, peritoneal viewing was normal in laparoscopy, peritoneal biopsy was performed to exclude malignancy and chronic inflammation was the sole finding.

During the follow-up period since the decline in white blood count together with marked neutropenia and eosinophilia pursued, bone marrow aspiration and biopsy was performed and pathological examination revealed marked increase in granulocytic cell lineage (largely eosinophilia) but no malignancy. Both CD 3 and CD 20 were negative. Solid organ neoplasm was excluded by means of physical examination and radiological imaging. Given the presumptive diagnosis of eosinophilic gastroenteritis, allergic skin testing to common food antigens was performed with negative results.

After excluding the other possible causes of eosinophilia (e.g. malignancy, parasitic infestation, connective tissue disease, inflammatory bowel disease, and autoimmune disease), diagnosis of eosinophilic gastroenteritis was defined. The diagnosis was supported by the presence of peripheral eosinophilia, abdominal symptoms (abdominal pain and distension) and findings (eosinophilic ascites) and wall thickening of distal esophagus and the jejunum detected by abdominal CT.

A low dose of steroid (20 mg/ day) was started. Seven days later symptoms of the patient (abdominal pain, distension) relieved and he was discharged. 20 days after discharge a control examination was performed and the patient was found to have no complaints and no ascites. Laboratory findings (Table 1) revealed a WBC of 4270/mm³ (neutrophils 1620/mm³, lymphocytes 1730/mm$^3$, eosinophils 516/mm³). Other laboratory findings were within normal ranges. Steroid was discontinued on a tapered manner. After discontinuation prednisolone symptoms did not relapse.

## Discussion

Although no histopathologic finding suggesting GIS involvement was seen in this case, the fact that there was no other cause (malignancy, parasitic infestation, drug use, bacterial infection, connective tissue disease) for peripheral eosinophilia and eosinophilic ascites, the presence of jejunal wall thickening on abdominal CT, and rapid improvement in symptoms and findings with treatment strongly favours the diagnosis of EG with serosal involvement. Striking neutropenia suggesting especially malignancy is an interesting finding and improvement in neutropenia with treatment is notable. EG is a disease characterised with eosinophilic infiltration in GIS and the presence of GIS symptoms (1). But the presence of all these is not necessary (2). In a study, peripheral eosinophilia could be found only in % 77.5 of EG cases (3).

**Table 1**. Laboratory and clinical findings.

| | Addmission | Day 8 | Posttreatment |
|---|---|---|---|
| White blood cells (/mm$^3$) | 22900 | 11300 | 4270 |
| Neutrophil (/mm$^3$) | 2360 (%10.3) | 686 (%6.07) | 1620 (%38) |
| Lymphocyte (/mm$^3$) | 3040 (13.2) | 2280 (%20.2) | 1730 (%40.7) |
| Monocyte (/mm$^3$) | 320 (%1.43) | 402 (%3.56) | 321 (%7.53) |
| Eosinophill (/mm$^3$) | 17100 (%74.6) | 7830 (%69.3) | 516 (%12.1) |
| Basophill (/mm$^3$) | 103 (%0.447) | 90 (%0.067) | 74 (%1.73) |
| Red blood cell (/mm$^3$) | 4.60 x 10$^6$ | 4.90 x 10$^6$ | 5.07 x 10$^6$ |
| Hemoglobin (g/dl) | 14.5 | 14.4 | 15.6 |
| Hematocrit (%) | 39.4 | 39.7 | 44.7 |
| Platelet (/mm$^3$) | 301000 | 291000 | 237000 |
| Erythrocyte sedimentation rate mm/h | 3 | 2 | 2 |
| C-Reactive Protein (mg/dl) | 0.76 | 0.1 | 0.138 |
| Total Ig E (kU/l) | 292 | 242 | 4.6 |
| Glucose (mg/dl) | 92 | 85 | 105 |
| Blood Urine Nitrogene (mg/dl) | 11.7 | 16.4 | 13.3 |
| Creatinin (mg/dl) | 1.0 | 0.9 | 0.8 |
| Alanine aminotranspherase (U/L) | 20 | 25 | 18 |
| Aspartate aminotranspherase (U/L) | 19 | 17 | 16 |
| Gamaglutamil transpeptidase (U/L) | 10 | 16 | 14 |
| Alchalen phosphatase (U/L) | 64 | 66 | 87 |
| Lactic dehydrogenase (U/L) | 312 | 172 | 179 |
| Total protein (g/dl) | 5.7 | 6.6 | 7.8 |
| Albumin (g/dl) | 3.6 | 4.2 | 4.5 |
| Bilirubin (mg/dl) | 0.43 | 1.05 | 0.71 |
| Sodium (mEq/L) | 140 | 136 | 136 |
| Potassium (mEq/L) | 4.3 | 4.6 | 4.2 |
| Urinalysis | Normal | Normal | Normal |
| Symptoms | Abdominal pain-dyscomphort, di- | Abdominal pain-dyscomphort, | No |
| Ascites | ++ | ++ | No |

Besides cases of EG with serosal involvement but without eosinophilic infiltration has also been reported as in our case (4). There may not be sufficient histopathological evidence in mucosal biopsies of cases with serosal involvement (5). In such cases abdominal CT findings (especially wall thickening) are important for supporting the diagnosis as in our case (6).

EG, is classified pathologically in three major groups; mucosal, muscular and serosal. Mucosal disease usually presents with findings like bleeding, malabsorption and protein losing enteropathy (7) and high serum IgE levels are noted (8). In the case of muscular layer involvement wall thickening and obstructive symptoms predominate. Serosal involvement is seen in 10 % of this group of patients and is characterised with exudative eosinophilic ascites (7). Even though it is easy to diagnose mucosal disease, biopsies may be inadequate for the diagnosis of muscular and serosal involvement as in our case.

The characteristic CT features include bowel wall thickening with halo sign or 'aranedilimb-like' sign or intra-luminal granuloma, extraluminal lymphadenopathy or granuloma with necrosis and luminal narrowing without obstruction. All of these CT features suggest an inflammatory disease rather than either a malignant or benign neoplastic process. Moreover, CT features combined with the typical clinical features, including an allergic history, peripheral eosinophilia and a rapid relief of symptoms by steroid therapy will likely lead to correct diagnosis (9). Even though findings on biopsy were inadequate to confirm the diagnosis of EG histologically in our case, esophagial and jejunal wall thickening, peripheral eosinophilia, eosinophilic ascites (Given that other possible causes of eosinophilia are excluded) and especially the dramatic response to steroid therapy strongly favours the diagnosis of EG.

Even if EG is related to allergy to some food and immunologic abnormalities by some authors; the exact etiology is still obscure. Food allergy and intolerance is detected mostly in patients with mucosal involvement (3). In this case eosinophilia and serum Ig E elevation may suggest an allergic cause, no allergen could be defined. Possible causes of this picture like malignancy, parasitic infestation, drug use are excluded by means of history, physical examination, laboratory and imaging procedures.
EG is a disease with good response to steroid therapy (3). But before starting steroid therapy parasitic infection should be excluded. Steroid therapy rapidly improves ascites and eosinophilia but relapses are not uncommon (8). Surgical treatment should not be considered except in cases of obstruction or to exclude primary or metastatic peritoneal malignancy when there are findings suggesting peritoneal involvement as in our case (5).

When literature is reviewed, no haematological complication except eosinophilia is seen in EG patients. But in our case cyclic neutropenia was observed and this condition improved with steroid therapy. Although eosinophilia together with neutropenia may suggest haematological malignancy or solid organ neoplasm, exclusion of these conditions by means of clinical, laboratory and imaging procedures and improvement of eosinophilia and neutropenia with therapy shows that cyclic neutropenia may be seen in EG patients. Our case is the first EG case that neutropenia is witnessed during the clinical course.

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