



Nodular Lymphoid Hyperplasia of the Gastrointestinal Tract with Selective IgA Deficiency in an Adult Patient: A Case Report

Selektif IgA eksikliği Olan Erişkin Hastada Gastrointestinal Nodüler Lenfoid Hiperplazi: Olgu Sunumu

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ABSTRACT

Nodular lymphoid hyperplasia (NLH) of the gastrointestinal tract is a rare and benign condition which may accompany immunodeficiency. Although benign, it carries a risk for intestinal and extraintestinal lymphoma. There is no specific and definitive treatment for NLH. Thus, treatment is dependent on the management of the associated conditions. Duodenal and terminal ileal biopsies from a 54-year-old woman with multiple mesenteric lymphadenopathies and gastrointestinal lymphoid hyperplasia revealed nodular lymphoid hyperplasia as well as a paucity of plasma cells. Based on histopathological findings and laboratory results, a diagnosis of selective IgA deficiency was made. It is important to evaluate patients with nodular lymphoid hyperplasia for additional histopathological findings due to the association with possible immune deficiency, such as absence or scarcity of plasma cells.

Keywords: Gastrointestinal tract, Nodular lymphoid hyperplasia, Selective IgA deficiency.

ÖZET

Gastrointestinal kanalın nodüler lenfoid hiperplazisi (NLH) nadir ve benign bir durum olup immün yetmezlikler ile ilişkili olabilir. Benign bir durum olmasına rağmen artmış intestinal ve ekstraintestinal lenfoma riski söz konusudur. NLH'nin kesin ve spesifik bir tedavisi yoktur. Bu nedenle, NLH'nin tedavisi ilişkili durumların yönetimine bağlıdır. Multiple mezenterik lenfadenopati ve gastrointestinal traktta diffüz lenfoid hiperplazi tespit edilen 54 yaşında kadın hastada duodenal ve terminal ileum biyopsilerinde nodüler lenfoid hiperplazi yanı sıra plazma hücrelerinde belirgin azalma tespit edilmiştir. Histopatolojik bulgular ve laboratuvar sonuçları esas alınarak selektif IgA eksikliği tanısı almıştır. Gastrointestinal trakt biyopsilerinde nodüler lenfoid hiperplazi bulguları izlenen hastalarda ilişkili olabilecek antiteler açısından plazma hücre azlığı gibi diğer histopatolojik bulguların da değerlendirilmesi önemlidir.

Anahtar Sözcükler: Gastrointestinal trakt, Nodüler lenfoid hiperplazi, Selektif IgA eksikliği.

Introduction

Nodular lymphoid hyperplasia (NLH) of the gastrointestinal tract is characterized by multiple small nodules distributed along the tract. The size of the nodules usually varies between 2-10 mm. While it is often asymptomatic, it can cause abdominal pain, chronic diarrhea, intestinal obstruction, or bleeding.

The pathogenesis is still unknown. Although it can affect all age groups, it primarily occurs in children and young adults with or without immunodeficiency. It has been reported that cases associated with common variable immunodeficiency, selective IgA deficiency, celiac disease, besides infectious diseases such as *H. pylori*, *Giardia*, and *Human Immunodeficiency Virus (HIV)* (1,2).

We herein present an adult patient with immunodeficiency who had nodular lymphoid hyperplasia in duodenal and terminal ileum biopsies.

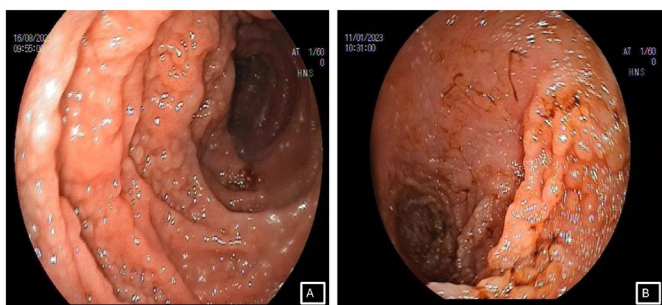


Figure I. Esophagogastroduodenoscopy (EGD) and colonoscopy of the patient showed numerous diffuse mucosal nodular lesions in the duodenum (A) and terminal ileum (B)

Case Report

A 54-year-old woman with complaints of chronic diarrhea applied to the gastroenterology clinic. She also had night sweating and weight loss. Multiple mesenteric lymphadenopathies were defined in computed tomography (CT). In esophagogastroduodenoscopy and colonoscopy, linear erosions in the stomach and cracked soil appearance in the duodenum were described, and biopsies were taken. Terminal ileum mucosa was seen as edematous and nodular (Figure I). In microscopic examination of biopsies, findings were compatible with *H. pylori* gastritis in the stomach samples. The biopsies from the duodenum and terminal ileum

showed nodular lymphoid hyperplasia (Figure II). In higher magnification, almost no plasma cells were detected in the lamina propria (Figure III). *Giardia* and *H. pylori* were not detected in the duodenal biopsy. It was recommended to investigate the possible presence of immunodeficiency in the case with the current histomorphological findings. Serum IgA was extremely low (0.065 g/L; normal 0.7–4.0 g/L), and serum IgE was decreased (<1.00 IU/mL; normal 1.9–170 IU/mL). Serum IgG and IgM ratios were within normal limits. Based on the laboratory results, the patient was diagnosed with selective IgA deficiency, and intravenous immunoglobulin (IVIG) treatment was given. Symptoms of the patient regressed. Surveillance of the patient continues at regular intervals. Written informed consent was obtained from the patient for publication of this case report.

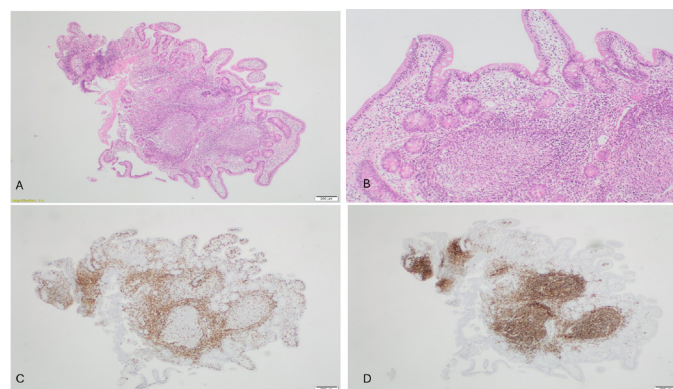


Figure II. (A) and (B). Duodenal and terminal ileal biopsies demonstrated nodular lymphoid hyperplasia (H&E x4), (C) and (D). Immunohistochemical staining in reactive lymphoid hyperplasia with CD3 and CD20, respectively (x4).

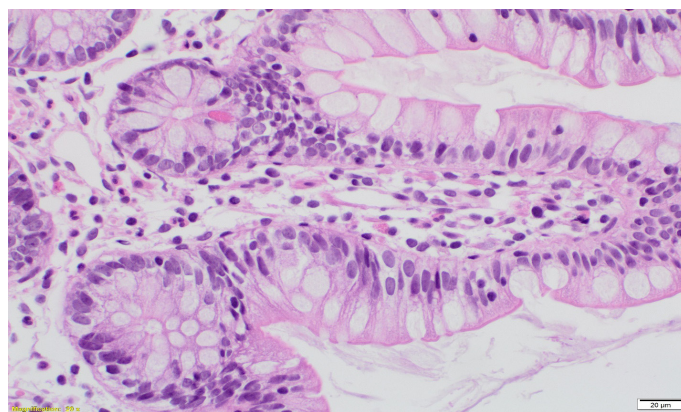


Figure III. In higher magnification, nearly zero plasma cells were detected in the lamina propria of the duodenum (H&E x20)

Discussion

During endoscopic imaging, nodular lymphoid hyperplasia (NLH) is seen as nodules ranging in size from 2 to 10 mm along the gastrointestinal tract. Nodular lymphoid hyperplasia primarily involves the small intestine, but it can also involve the colon and stomach (1). Lymphoid nodules localized in the colon may appear as red macules, as circumferential target lesions (halo sign), or as raised papules (2). Nodular lymphoid hyperplasia can be observed in a focal or diffuse form. In children, it is usually limited to the rectum, colon, and terminal ileum, has a mild clinical course, and often shows spontaneous regression. Cases associated with refractory constipation, viral infection, and food allergies have been reported (3). It has been emphasized that prognosis determination is more uncertain in adults (4). Although case reports and series have increased in the last years, there is no epidemiological data to determine incidence and prevalence (1,2).

Pathogenesis is largely unknown. However, there are some theories explaining the condition according to the presence/ absence of immunodeficiency. It is thought that in the case of immune deficiency, the accumulation of plasma cells and their precursors due to maturation defects of B lymphocytes occurs during the compensation of functionally inadequate intestinal lymphoid tissue (1). It is believed that NLH in the absence of immunodeficiency may be related to repetitive immune stimulation of gastrointestinal lymphoid tissue (2,3).

Chiaramonte et al suggested that NLH could be a transitional stage in the development of malignancy (5). It has been emphasized that NLH carries a risk for intestinal and extraintestinal lymphoma (1,2). Considering the risk of malignant transformation, regular endoscopic control becomes important (1). Although the duration and interval of these controls are not defined, it has been reported that a biopsy should be taken from growing lymphoid nodules to exclude transformation (2).

The treatment of NLH is directed to associated conditions (1). The diagnosis of NLH requires histopathological confirmation following endoscopic imaging or contrast barium examination. This is necessary to distinguish entities that may be confused with NLH and to prevent overtreatment resulting

from misdiagnosis.

In conclusion, although NLH is a rare and benign condition, it is clinically important due to its associated conditions and possible complications such as malignant transformation. Therefore, detecting the underlying causes and monitoring these patients are necessary for treatment and prevention of complications. It is important to keep in mind the possible associated conditions in a patient with nodular lymphoid hyperplasia and to evaluate all possibilities in the differential diagnosis.

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