

Case Report

Coexistence of Celiac Disease and Systemic Lupus Erythematosus: A Case Report

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Received: 14.01.2021

Accepted: 27.02.2021

Published: 17.03.2021

Journal homepage:<https://www.easpublisher.com>**Quick Response Code**

Abstract: Celiac disease (CD) is a chronic inflammatory disorder of the small intestine triggered by the ingestion of gluten. It has been associated with auto-immune disorders. Although many similarities exist between the pathogenesis of CD and systemic lupus erythematosus (SLE), their association has been rarely reported. We describe a case of a 23-year-old woman diagnosed with CD, and one year later developed SLE.

Keywords: Coexistence, Celiac Disease, Lupus Erythematosus.

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INTRODUCTION

Systemic lupus erythematosus (SLE) and celiac disease (CD) are complex and clinically heterogeneous autoimmune disorders. Their pathogenesis is still not completely understood; genetic and environmental factors are implicated [1-3]. SLE involves the gastrointestinal tract which is well documented [4] but its association with CD is rare [5, 6]. Nevertheless, some data suggest that patients with SLE develop CD and vice versa, highlighting a possible association between these diseases [3, 7]. We report the case of a 23-year-old woman with CD, who developed SLE one year after the diagnosis.

CASE PRESENTATION

A 23-year-old woman presented with pallor, abdominal pain, diarrhea, and steatorrhea and weight loss. Physical examination showed meteorism, bilateral leg edema and muscle atrophy.

The laboratory evaluation revealed abnormal results, which included iron deficiency anemia at 6g/dl, hypocholesterolemia at 0,6 g/l, and hypoalbuminemia at 20 g/l. IgA anti-endomysial antibodies were positive (1/10). While anti-tissue transglutaminase antibodies (IgA and IgG) and IgG anti-endomysial antibodies were negative. Mucosal histopathology revealed villous atrophy, classified as 3A according to Marsh's scale [8].

The diagnosis of CD was established, and a gluten-free diet was initiated. The six months follow-up showed a resolution of gastrointestinal symptoms and a weight gain. Additionally, laboratory parameters were within normal range. One year after, the patient presented with polyarthralgia, photosensitivity, alopecia, ascites and lymphopenia (700 cells/mm³). Serum anti-nuclear antibodies were positive with a titer of 1/160 and a homogeneous pattern. The titer of anti-dsDNA was high at 55 IU/l. The kidney function and cardiac exploration did not show any abnormality. The diagnosis of SLE associated with CD was made, and the patient started antimalarial drugs in association with oral prednisone 20 mg/day followed by a gradual taper. At two-year follow-up, a clinical remission was achieved, gastro-intestinal symptoms were improved and the laboratory parameters were normal.

DISCUSSION

The frequency of malabsorption in SLE is estimated at 10%. It is certainly underestimated due to the small number of studies [9]. Gastrointestinal manifestations are commonly presented in up to 50% of SLE patients [10], and include mesenteric arteritis, intestinal perforation and obstruction, ascites, peritonitis, enteritis, ulcerative colitis, pancreatitis, esophageal and hepatic involvement [4, 5]. In contrast, about 50% of adults with CD have extra-intestinal

manifestations; such as iron deficiency anemia, osteoporosis, infertility, cerebellar ataxia, and peripheral neuropathy [11]. Many autoimmune diseases can coexist in a single patient. However, the association of SLE with CD is still unclear [6]. A combination of histology, serology, morphometry and HLA typing may be helpful in equivocal cases [12].

Several theories have been proposed to explain this association, including the duration of exposure to gluten which would increase the risk of developing autoimmune diseases [13], and a genetic predisposition (HLA B8 DR3) [9]. High frequency of histocompatibility antigens HLA-DR3 is observed in both CD and SLE [2, 5, 14, 15]. Likewise, SLE is associated in 40 to 70% of cases with HLA-DR3 [15].

Mader *et al.* reported a study of 21 SLE patients. A history of abdominal pain and occasional diarrhea was elicited in 2 patients. In one of them, histologic examination revealed flattened and deformed villi with an inflammatory infiltrate [10]. The retrospective study of Benghorbal *et al.* enrolled 340 patients with SLE. Six presented with CD. The mean age at diagnosis of the association SLE-CD was 28 years (16–47 years). CD occurred ten and twenty-three years before SLE respectively, in two cases. CD occurred concomitantly in four cases. The laboratory finding showed malabsorption. Mucosal histopathology revealed villous atrophy, classified as 4 according to Marsh's scale in two cases, stage 3 in one and stage 2 in two, and was normal in one case. All the patients were tested positive for antigliadin, anti-endomysial and anti-transglutaminase antibodies. Four patients had a cutaneous and articular form of SLE without visceral involvement. Lupus nephritis occurred in one case [16].

Zitouni *et al.* reported five cases over a 4-year period. CD and SLE occurred concomitantly in one patient, whereas CD preceded SLE in one patient and was diagnosed afterwards in the remaining 3 patients. Villous atrophy on duodenal biopsy specimens with a favorable response to a gluten-free diet was noted in all the 5 patients. Only 4 patients had positive serological tests for celiac disease and only three had abdominal symptoms [17].

Due to its rarity, the association of CD with SLE is considered by some authors to be fortuitous. The study of Rensch *et al.* included 103 patients with SLE, who were tested for the serological presence of IgA and IgM antigliadin and IgA anti-endomysial antibodies. Those with positive serology underwent esophagogastroduodenoscopy with duodenal mucosal biopsy. Twenty-four patients (23.3%) tested positive for antigliadin antibody. None of them were found to have endoscopic or histological evidence of CD, making the false positive rate of antigliadin antibody 23%. According to the authors of this study, there seems to be no association between CD and SLE

since false positive antigliadin antibodies in patients with SLE is common [5].

CONCLUSION

The association between systemic lupus erythematosus and celiac disease deserves the attention of researchers. Further researches are needed in order to determine the true prevalence of this coexistence.

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Cite This Article: Jaouad Yousfi *et al* (2021). Coexistence of Celiac Disease and Systemic Lupus Erythematosus: A Case Report. *East African Scholars J Med Surg*, 3(3), 44-46.